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An Investigation of Psychosocial Aspects of Adolescent Idiopathic Scoliosis: Adolescent and Parent Perspectives

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Thesis submitted to University College Dublin in fulfilment of the requirements for the degree of Doctor of Philosophy in Psychology

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November 2021

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Publications and Presentations

Publications

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Motyer, G., Dooley, B., Kiely, P., & Fitzgerald, A. (2021). Parents' information needs, treatment concerns, and psychological well-being when their child is diagnosed with adolescent idiopathic scoliosis: A systematic review. *Patient Education and Counseling, 104*(6), 1347 – 1355. https://doi.org/10.1016/j.pec.2020.11.023

Presentations

Motyer, G., Kiely, P., & Fitzgerald, A. (Poster). The psychosocial implications of living with adolescent idiopathic scoliosis: A qualitative study with a pre-surgical patient cohort. *European Paediatric Psychology Conference, Stockholm, Sweden (Online),* Oct 4 – 6, 2021.

Motyer, G., Kiely, P., & Fitzgerald, A. (Oral). A Qualitative Exploration of Body Image Among Adolescents with AIS. *Appearance Matters Conference Online,* July 13-15, 2021.

Motyer, G., & Fitzgerald, A. (Oral). Wellbeing of young people with chronic medical conditions during COVID-19: Findings from the SCOLI Study. *Disseminating Research on the Impact of COVID-19, UCD School of Psychology Virtual Conference*, Dublin, June 18, 2021.

Motyer, G., Kiely, P., & Fitzgerald, A. (E-Presentation). The Lived Experiences of Adolescents with Idiopathic Scoliosis in Ireland. *Society for Research on Adolescence Biennial Meeting, San Diego, USA, (Online),* March 2021.

Motyer, G., Dooley, B., Kiely, P., & Fitzgerald, A. (E-Presentation). Parenting an Adolescent with Idiopathic Scoliosis: A Multimethod Study. *Society for Research on Adolescence Biennial Meeting, San Diego, USA, (Online),* March 2021.

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Motyer, G., & Fitzgerald, A. (Oral). Adolescents' Adjustment to Idiopathic Scoliosis: What we know and what we need to learn. *UCD Graduate Research Symposium,* May 4, 2017.

Motyer, G., & Fitzgerald, A. (Poster). Exploring Adolescent Idiopathic Scoliosis: Project Plan. *EU COST Action IS1012 'Appearance Matters' International Summit,* Slovenia, April 20, 2017.

Statement of Original Authorship

I hereby certify that the submitted work is my own work, was completed while registered as a candidate for the degree of Doctor of Philosophy in Psychology at University College Dublin, and I have not obtained a degree elsewhere on the basis of the research presented in this submitted work.

____

November 2021

Abstract

This thesis presents a multimethod programme of research that aimed to further current understanding of psychosocial aspects of adolescent idiopathic scoliosis, a paediatric spinal condition characterized by a sideways curvature of the spine. The first study comprised a qualitative exploration into adolescents' experiences of living with AIS at the presurgical stage of treatment (*N* = 14). Four key themes were developed, including "Proceeding with Caution," "Am I Different?" "An Emotional Journey," and "No Pain, No Gain." The second study examined the contribution of psychosocial processes to variation in health-related quality of life and body image outcomes among adolescents with idiopathic scoliosis (*N* = 115). Findings demonstrated that coping strategies were significantly associated with health-related quality of life among this patient group, and several appearance-related cognitions were significantly associated with body image satisfaction and disturbance. The third study comprised a systematic review and synthesis of the literature pertaining to parents' information needs, treatment concerns, and psychological wellbeing in the context of their child's adolescent idiopathic scoliosis. The fourth and final study was a qualitative exploration of parents' experiences in relation to their child's AIS from diagnosis to presurgical preparation (*N* = 20), which resulted in three main themes including "Fear of the Unknown," "Maintaining Normality," and "Navigating the Patient Pathway." Together, these studies make a number of significant contributions to the literature on psychosocial aspects of adolescent idiopathic scoliosis, and provide implications for research and practice in line with a patient and family-centered approach.

Chapter 1

Thesis Overview

1.1. Introduction to the Research

Receiving a diagnosis of adolescent idiopathic scoliosis (AIS) is a significant life event for young people and the parents of those affected. Affecting approximately 1 – 3% of adolescents, AIS is a spinal condition characterised by a sideways curvature of the spine that develops with no known cause around the onset of puberty (Weinstein et al., 2008; Altaf et al., 2013). Physically, the spinal curvature can result in postural asymmetries including imbalanced shoulders and hips, and rib prominence. Hresko (2013) described a typical clinical presentation of AIS in the following case vignette from the orthopaedic department of a children's hospital:

"A 12-year-old girl presents with her parents after a positive school screening for scoliosis. Physical examination reveals shoulder and torso asymmetry with trunk imbalance (i.e., shift from the midline). Neurologic and skin examinations are normal. How should the patient be evaluated and treated?"

For adolescents presenting with suspected idiopathic scoliosis as described here, radiographic assessment is typically conducted to measure the size of the spinal curve and a diagnosis is confirmed by an orthopaedic specialist (Horne et al., 2014). Although the cause of AIS is uncertain, growth spurts during adolescent development are understood to contribute to progression of the condition (Beauchamp et al., 2019). What ensues for many adolescents and their parents is months or years of potential progression of the spinal curve. Available treatment aimed at reducing progression can include orthotic bracing, or for curves that progress severely, surgical intervention may be required. The diagnosis and treatment of AIS during this time is likely to pose psychosocial challenges for affected adolescents and for their parents who often take on the role of managing their child's healthcare.

1.2. Overarching Research Questions

Under the broad aim of furthering current understanding of psychosocial aspects of adolescent idiopathic scoliosis from a patient and family-centered approach, this thesis sought to address three main research questions in relation to adolescents and their parents:

(i) How do adolescents experience living with idiopathic scoliosis?

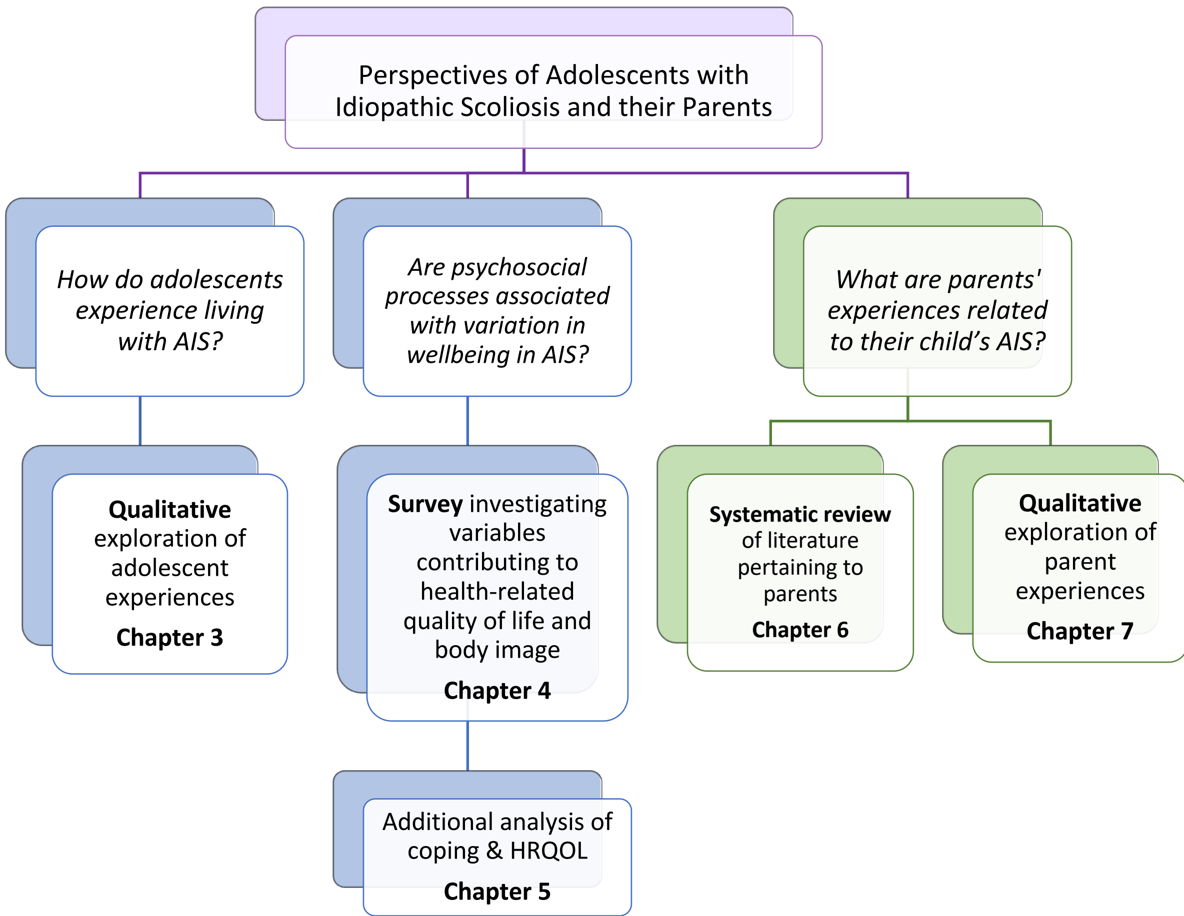
(ii) Are psychosocial processes associated with variation in wellbeing (i.e., health-related quality of life and body image) among adolescents with idiopathic scoliosis?

(iii) What are parents' experiences related to their child's adolescent idiopathic scoliosis?

To address these research questions, a programme of research studies were conducted as presented in Figure 1.1. Specific aims and objectives for each study are presented in the corresponding chapters.

Figure 1.1

Components of the Research Project Including Overarching Research Questions & Programme of Studies



1.3. Outline of Thesis Content

Following on from this overview chapter, Chapter 2 provides an introductory literature review intended to provide a background to the research topic and position the studies included in this thesis. The aims and an introduction to each of the studies conducted in the thesis is provided. This is followed by five chapters detailing the programme of research that was conducted. Chapters 3 to 5 pertain to research conducted with adolescents while Chapters 6 and 7 report on data collected with parents.

Chapter 3 presents the qualitative interview study conducted with adolescents with presurgical AIS to explore their experiences of living with AIS (*N* = 14). Four key themes were developed: *"Proceeding with Caution," "Am I Different?" "An Emotional Journey,"* and *"No Pain, No Gain."* Detailed in Chapter 4 is the cross-sectional quantitative survey study named the "SCOLI Survey" which investigated the contribution of a number of psychosocial processes (i.e., coping and appearance-related cognitions) to variation in health-related quality of life and body image among adolescents with AIS, as well as their potential support needs (*N* = 115). Following this, Chapter 5 presents a cluster analysis of the coping data collected as part of the SCOLI Survey to further examine the role of coping in health-related quality of life.

Chapter 6 presents the systematic literature review, the first of two studies that focused on the parents of adolescents with idiopathic scoliosis. The systematic review was undertaken to synthesize and evaluate previously published literature pertaining to parents' experiences in the context of their child's AIS, specifically their information needs, treatment concerns, and psychological wellbeing. Next, Chapter 7 describes the qualitative interview study conducted to explore parents' experiences across the trajectory of their child's AIS from diagnosis to presurgical preparation (*N* = 20). Three key themes were developed: *"Fear of the Unknown," "Maintaining Normality,"* and *"Navigating the Patient Pathway."* Finally, Chapter 8 concludes the thesis by reviewing the research studies and their main findings. The contributions of this programme of research are reviewed along with consideration of the implications for practice and future research directions.

1.4. Unique Contributions of the Research

The overarching aim of this thesis was to further current understanding of psychosocial aspects of adolescent idiopathic scoliosis, from adolescent and parent perspectives. Through a multimethod programme of research, this thesis makes a number of unique contributions to the literature on adolescent idiopathic scoliosis. These contributions include:

(i) It provided an in-depth qualitative exploration of the experiences of adolescents living with presurgical AIS from a psychosocial perspective (Chapter 3).

(ii) It examined the contribution of psychosocial variables to variation in health-related quality of life and body image among adolescents with AIS (Chapter 4).

(iii) It provided a comprehensive investigation of coping among adolescents with AIS and how coping patterns relate to health-related quality of life (Chapter 5).

(iv) It took a systematic approach in reviewing what is known about parents' experiences in the context of their child's AIS, providing a critical review of the literature base (Chapter 6).

(v) It provided an in-depth qualitative exploration of parents' experiences across the trajectory of their child's AIS from diagnosis to presurgical preparation. (Chapter 7).

(vi) Overall, this thesis furthers understanding of the experiences of adolescents with AIS and their parents and considers implications of this research for support provision.

Chapter 2

Introductory Literature Review

Chapter Overview

This chapter includes the introductory literature review, presenting relevant literature on adolescent idiopathic scoliosis and a background to the research topic in order to position the research conducted for this thesis. Firstly, adolescent idiopathic scoliosis is described in detail in terms of the diagnosis, aetiology, prognosis, and treatment for this condition. Current understanding of the psychosocial impact of AIS is reviewed considering the adolescent perspective and in particular, key psychosocial outcomes health-related quality of life and body image. Furthermore, the importance of a patient and family-centered care approach to AIS is introduced, recognising the value of patient and parent perspectives. This chapter ends with a description of the programme of research conducted for this thesis.

2.1. Adolescent Idiopathic Scoliosis (AIS)

2.1.1. Diagnosis

Adolescent Idiopathic Scoliosis (AIS) is characterized by vertebral rotation and a lateral curvature of the spine of at least 10 degrees (°) that develops with no identifiable cause (Weinstein et al., 2008; 2019). AIS is one of the most common spinal conditions seen in paediatric orthopaedics, affecting approximately 1% – 3% of adolescents, with a higher prevalence among females (Weinstein et al., 2008; Konieczny et al., 2013). The female to male incidence ratio is estimated at 2:1 and this increases for larger curvatures (Konieczny et al., 2013). Typically, AIS is identified at the peripubertal period before the age of 16, when affected adolescents present with postural asymmetries such as unlevel shoulders or rib prominence (Altaf et al., 2013). The primary indicator of scoliosis severity is a radiological measurement of the major spinal curve known as the Cobb angle presented in degrees (Greiner, 2002; Goldberg et al., 2008). AIS curvatures can be categorised by severity to determine appropriate treatment, whereby those between 10° – 24° can be considered mild, those that progress to 25° – 44° are moderate, and curves of 45° or above are severe (Janicki & Alman, 2007; Altaf et al., 2013). Curvatures can be categorised as thoracic, lumbar, or

thoracolumbar, depending on where they occur in the spine, with the most common AIS curvatures occurring in the thoracic or thoracolumbar spine (Konieczny et al., 2013; Zadeh & Gleiber, 2015). AIS can be detected via school screening programmes. However, screening practices vary widely and have been discontinued in many countries (Altaf et al., 2017).

2.1.2. Aetiology

The cause of AIS is largely unknown, and the diagnosis is therefore one of exclusion when other non-idiopathic forms of scoliosis have been ruled out (Weinstein, 2019). Other non-idiopathic forms of scoliosis include congenital scoliosis caused by vertebral anomalies and neuromuscular scoliosis which can develop secondary to neurological or muscular conditions such as cerebral palsy or spinal muscular atrophy (Janicki & Alman, 2007; Konieczny et al., 2013). Idiopathic scoliosis has no clear cause and is categorized into juvenile (<10 years) and adolescent onset (>10 years until completion of growth), with adolescent onset accounting for approximately 90% of all idiopathic scoliosis cases (Konieczny et al., 2013). Several potential aetiological factors for AIS have been investigated including genetic factors, as studies indicate increased risk of developing AIS if a first degree relative is affected, and higher concordance rates among monozygotic twins (Addai et al., 2020). Other potential aetiological factors proposed have included hormones and metabolic dysfunction, nervous system abnormalities, and skeletal length asymmetries (Wang et al., 2011). Overall, no clear cause has been identified and it is generally accepted that the aetiology of AIS is multifactorial and heterogenous (Newton Ede & Jones, 2016).

2.1.3. Prognosis

The course of AIS can be unpredictable, but often the severity of the spinal curvature will progress over the course of adolescence (Greiner et al., 2002). Pubertal growth spurts during adolescent development are associated with increases in curve severity that can be rapid in nature, and curve progression subsequently diminishes when growth is complete (Altaf et al., 2013; Beauchamp et al., 2019). Other known risk factors for curve progression include having a thoracic spinal curve type, pre-menarchal status at diagnosis, and having a Cobb angle of more than 25 - 30° around the onset of puberty (Weinstein et al., 2008; Altaf et al., 2013; Beauchamp et al., 2019). Epidemiological studies also demonstrate that girls are more likely

to develop larger spinal curvatures than boys, with the female to male ratio estimated to be approximately 7:1 for curves exceeding a Cobb angle of 40° (Konieczny et al., 2013).

While patients' clinical presentation will vary and change over the course of their AIS, symptoms resulting from their spinal curvature can include back pain, altered functional ability or mobility, and pulmonary problems (Weinstein et al., 2008). Pulmonary symptoms such as shortness of breath are typically only associated with larger curves (greater than 80° - 90° Cobb angle) (Asher & Burton, 2006; Goldberg et al., 2008). A significant and often problematic symptom arising from AIS is the appearance of the spinal curvature (Weinstein et al., 2008). AIS is known to have a considerable impact on the physical appearance of the upper body, particularly for curves that progress severely. The vertebral rotation and consequent rotation of the ribcage that occur in AIS can lead to visible asymmetries of the back and chest, truncal shift, rib prominence, waistline asymmetry, and unlevel shoulders and hips (Altaf et al., 2013). The impact that these postural asymmetries can have on the adolescents' body shape and appearance are often the first noticeable signs of AIS. While AIS will affect 1 – 3% of the population, it is estimated that 0.3 to 0.5% will have curves that progress to a stage requiring treatment with bracing or surgery (Weinstein et al., 2013). AIS can be considered a chronic health condition (musculoskeletal) in line with definitions indicating an expected duration of at least three months and a need for medical attention above that ordinarily expected (Perrin et al., 1993; Sawyer et al., 2007).

2.2. Treatment for AIS

Treatment options for AIS are primarily dependent on the severity of the spinal curve and the amount of skeletal growth remaining (Beauchamp et al., 2019). Guidelines for the treatment of AIS include observation, bracing, and surgical treatment. Adolescents with minor curves may remain under observation to monitor progression. The two main treatment approaches are conservative methods (i.e., orthotic bracing and physiotherapy) to prevent progression of mild to moderate scoliosis, and surgical intervention to correct more severe scoliosis (Weinstein et al., 2008). Approximately 10% of AIS cases will progress to a stage requiring surgical intervention (Altaf et al., 2013).

2.2.1. Conservative Treatment

Observation. For adolescents with minor spinal curvatures (i.e., Cobb angle 11° - 24°) a “wait and see” approach is typically adopted. Radiographic and clinical assessment to monitor possible curve progression is recommended every 4 - 6 months, depending on the patients’ age and growth potential (Altaf et al., 2013). Adolescents who are younger and have more skeletal growth remaining are likely to be at higher risk of curve progression and may need to be monitored closely, while those with lower risk of progression may be reviewed less frequently (Altaf et al., 2013; Choudhry et al., 2016).

Orthotic Bracing. Growing adolescents with moderate curves of 25° or more may undergo conservative orthotic bracing on advice of their treating physician. Braces for scoliosis are designed to exert an external force to guide the growth of the spine during growth (Schiller et al., 2010). While a variety of braces have been developed, thoracic-lumbar-sacral orthoses (TLSOs), worn underneath clothing for 18 hours or more per day, are most common (Weinstein et al., 2008; Schiller et al., 2010). Patients who have reached or are near to reaching skeletal maturity are unlikely to benefit from a brace (Altaf et al., 2013). The primary goal of bracing is to prevent significant curve progression until the patient reaches skeletal maturity, at which point the risk of progression decreases (Weinstein et al., 2008).

Bracing can help to prevent significant progression of the spinal curve and reduce the possibility of the patient requiring surgery (Schiller et al., 2010). However, the success of bracing as a treatment for AIS has remained a contested issue among scoliosis clinicians (Weinstein et al., 2008; Altaf et al., 2013), with evidence for bracing efficacy typically of low quality (Negrini et al., 2015). However, encouraging evidence in favour of conservative treatment for AIS has been provided by a multicentre randomized clinical trial known as the BrAIST trial (Weinstein et al., 2013). This trial demonstrated that 72% of adolescents who were treated with TLSO braces reached skeletal maturity without the need for surgical intervention (i.e., curves remained <50°), compared to only 48% of adolescents who had remained under observation alone. Importantly, the effectiveness of bracing is highly reliant on patients’ adherence to the prescribed hours of brace wear, and poor adherence is a commonly documented problem (Schiller et al., 2010). Research indicates that factors including age, the appearance of the brace, social settings, and psychological aspects such as

motivation to avoid surgery can affect compliance with bracing treatment (Rahimi et al., 2019).

Physiotherapy. Physiotherapeutic, scoliosis-specific exercise (PSSE) may also be recommended for adolescents with mild and moderate spinal curves, alone or in combination with bracing treatment with the aim of minimizing curve progression (Marti et al., 2015; Monticone et al., 2014). One of the most popular PSSE approaches for AIS is the Schroth method (Weiss, 2011), which involves exercises to strengthen and lengthen the asymmetrical muscle groups, along with posture correction and breathing techniques, performed under physiotherapist supervision, and using at-home programmes (Day et al., 2019). Randomized controlled trials have reported that participation in a six-month Schroth program stopped or significantly reduced curve progression in patients with AIS aged 10 – 18 (Schreiber et al., 2016; Kuru et al., 2016). As is the case with bracing, the success of physiotherapy relies largely on patient compliance with treatment (Beauchamp et al., 2019). Relatedly, supervised physiotherapy exercise sessions are found to be more beneficial for curve management than at-home exercises which rely on the patient alone (Kuru et al., 2016). While interest in PSSE is increasing, higher quality evidence is required to support the recommendation of this treatment approach for mild to moderate scoliosis (Marti et al., 2015).

2.2.2. Surgical Intervention

For adolescents whose curvatures progress severely, exceeding a Cobb angle of 45° - 50°, spinal fusion surgery is considered as a treatment option. This is considered a standard threshold because curves that exceed 50° at skeletal maturity typically continue to progress slowly throughout adulthood, whereas smaller curvatures are less likely to worsen following completion of growth (Weinstein & Dolan, 2015). The main aims of surgical treatment are to stop further progression of the spinal curve and achieve the maximum possible correction of the existing curvature (Weinstein et al., 2008). Posterior spinal fusion with instrumentation is the standard surgical approach in the correction of AIS (Beauchamp et al., 2019). This surgery is considered a major and highly invasive operation, during which fixation devices (i.e., pedicle screws, rods) are inserted to hold the spine in place and the corrected vertebrae are fused to prevent further growth or movement of the corrected area (Goldberg et al., 2008). Curve correction of more than 60% to 70% is typically achieved using modern surgical techniques

(Imrie et al., 2011), with an average correction of 80% reported in one study (Roberts et al., 2014). After surgery, some degree of the patient's spinal mobility and flexibility may be lost depending on the number of vertebral levels that are fused.

Advancement of surgical procedures over time has greatly improved the safety of undergoing surgery for AIS, and the risk of complications is considered low (Altaf et al., 2013). The incidence of neurologic complications following surgical treatment of AIS is reported to be less than 1% (Altaf et al., 2013). Other potential complications can include non-union (i.e., the vertebrae do not heal or fuse), and persistent postsurgical pain (Perry et al., 2018), and wound infection (Tarrant et al., 2015). The main surgical concerns reported by adolescents prior to surgery are possible neurologic injury, postsurgical pain, and their ability to take part in physical activities in the future (Chan et al., 2017). While postsurgical experiences will vary among patients, it is advised that patients can be discharged from hospital three to six days following their surgery (SRS, 2020). Limited physical activity is advised in the immediate postsurgical period, with a gradual increase to non-contact, low-impact activities. On advice of the treating physician, unrestricted physical activity may resume between 6 to 12 months post-surgery (Tarrant et al., 2014).

Issues with Timely Care. In the treatment of AIS, issues regarding timely access to care are recognized and wait times for spinal surgery for AIS have been a topic of major concern. Best practice guidelines recommend a 3 – 6 month timeframe from the decision to operate (Ahn et al., 2011), however reported wait times have included up to 9 months in the UK, 12 months in Canada, and 13 months in Ireland, with many adolescents waiting much longer than this (Clark, 2008; Miyanji et al., 2015; Tarrant et al., 2016; Ombudsman for Children, 2017). Continued curve progression during prolonged surgical wait times can have a detrimental effect on patients leading to more complex surgical procedures (Fallatah et al., 2015; Tarrant et al., 2016). In addition, there is no clear consensus on screening guidelines and screening is varied internationally (Labelle et al., 2013), which can lead to delayed scoliosis referral and detection. Most cases of AIS are first detected by a non-medical professional, typically the patient or a family member (Ali & Edgar, 2006; Anthony et al., 2021). Concerningly, research indicates that a trend of delayed referral can mean that many

patients present too late for conservative management and may be already approaching the surgical threshold at diagnosis (Anthony et al., 2021).

2.3. Psychosocial Aspects of Adolescent Idiopathic Scoliosis

2.3.1. Biopsychosocial Approach

The biopsychosocial (BPS) model originally introduced by Engel (1977) has underpinned the growth of a holistic approach to the understanding of health conditions that goes beyond a sole focus on the underlying biomedical causes or mechanisms involved. The BPS approach draws attention to the whole person, acknowledging that psychological and social (psychosocial) factors can be implicated in the prognosis and clinical management of health conditions (Bolton & Gillett, 2019). Central to the holistic BPS approach is understanding the person's lived experience of their health condition, and the impact that the condition can have on psychosocial domains of their lives (Epstein & Borrell-Carrio, 2005; Langberg et al., 2019). In the context of paediatric health conditions, psychosocial domains of interest can include activity limitations and physical functioning, pain, emotional functioning (e.g., depression and anxiety symptoms), body image, self-esteem, school or social functioning, sleep, parental responses, and family functioning (Law et al., 2017; Duncan et al., 2017; Rapoff et al., 2017).

As highlighted recently by Płaszewski et al. (2019), little is known about adolescents experiences of living with AIS, despite the significance of developing a serious spinal condition at the sensitive development phase of adolescence. Authors have acknowledged that generally, a substantial amount of previous research on AIS has been biomedical in nature, with sporadic publications relevant to psychosocial aspects of the condition available (Płaszewski et al., 2019). However, the importance of psychosocial wellbeing in the treatment of patients with AIS has been highlighted. For example, the international Scientific Society on Scoliosis Orthopaedic and Rehabilitation Treatment (SOSORT) published a consensus statement advocating for a multidimensional treatment approach to AIS, identifying that patients' appearance concerns and quality of life are priority treatment outcomes in AIS care (Negrini et al., 2006). The growing appreciation for the importance of examining the psychosocial impact of AIS is also demonstrated by literature reviews examining research on this topic (Tones et al., 2006; Gallant et al., 2018). Research into the psychosocial impact of

AIS has focused on health-related quality of life and considering the appearance-altering nature of AIS, there has also been a focus on body image among this patient group.

2.3.2. Adolescent Perspective

The challenges of living with a chronic health condition in adolescence can represent a significant psychosocial burden (Yeo & Sawyer, 2005; Sawyer et al., 2007). The timing of the AIS diagnosis and the subsequent treatment of this condition occurs during a critical developmental stage. Adolescence is a rapid period of biological, psychological and social change, and a time when young people experience puberty, develop their personal identity, and gain independence (Christie & Viner, 2005; Alderman & Breuner, 2019). On top of the normative developmental demands of this period, adolescents with AIS are faced with a multitude of challenges considering the unpredictable progression of their condition over time, changes to appearance, and medical treatment including the possibility of invasive spinal surgery. Moreover, adolescence is an important period of body image development and a time when individuals may be vulnerable to body image dissatisfaction (Levine & Smolak, 2002). It is recognised that the appearance changes resulting from scoliosis can be particularly challenging for AIS patients who are already managing the heightened appearance-related pressures and desire for peer conformity typical of the teenage years (Carrasco & Ruiz, 2014; Reichel & Schanz, 2003; Rumsey & Harcourt, 2007).

Previous research concerning the psychosocial impact of AIS has indicated that adolescents with AIS are more likely to experience appearance self-consciousness, associated problems with social interactions (Auerbach et al., 2014), and higher levels of body image dissatisfaction compared to their non-affected peers (Tones et al., 2006; Rushton & Grevitt, 2013). Authors have also indicated that adolescents with AIS may be more at risk of having depressive symptoms, a poorer sense of self-worth, and a less positive attitude toward life in general (Reichel & Schanz, 2003). Concerns have also been raised in more recent research that adolescents with AIS may be vulnerable to depression and anxiety symptoms (Gallant et al., 2018). In particular, previous studies have indicated that patients with scoliosis are almost twice as likely to develop depression in comparison to non-affected individuals (Chang et al., 2016). Sanders et al., (2018) reported that 32% of those living with AIS that were surveyed experienced clinically significant psychological distress, and that the AIS sample experienced

similar levels of anxiety symptoms to paediatric oncology patients. Together, this previous literature underscores the need to consider psychosocial aspects of AIS. Among adolescents with AIS, two key dimensions of psychosocial wellbeing within the literature are health-related quality of life and considering the appearance-altering nature of AIS, there has also been a particular focus on body image among this patient group (Weinstein et al., 2008; Tones et al., 2006; Gallant et al., 2018).

Health-Related Quality of Life. Assessment of health-related quality of life (HRQOL) has become common in research on paediatric health conditions, reflecting the widening of focus from biomedical outcomes, to recognise psychosocial dimensions of health (Sawyer et al., 2007). While objective medical characteristics (e.g. curve size) are key considerations in management and treatment throughout the course of a paediatric health condition, the quality of those years from the patient perspective should also be understood (Buckloh & Schilling, 2015). HRQOL is a multidimensional construct which describes an individual's perception of how their condition affects them in relevant physical, psychological, and social domains (Spieth & Harris, 1996). AIS is understood to affect multiple domains of HRQOL, specifically function/activity, self-image, pain, and mental health (Asher et al., 2000). The study of HRQOL in AIS has centered around the Scoliosis Research Society (SRS) self-report measure, designed to assess each of these four domains. The original version of this measure (Haher et al., 1999) has been modified and refined since its inception to improve internal consistency and reliability, resulting in its current version, the SRS-22r (Asher et al., 2006). Originally developed as an outcome measure to assess treatment efficacy, it is now also used to assess HRQOL throughout the presurgical period (Rainoldi et al., 2015). The function/activity domain captures physical capability and limitations to school or social activity, while the self-image domain assesses satisfaction with back shape and how this affects attractiveness or relations to others. The pain domain covers levels of pain experienced, and the mental health domain taps into feelings of low mood or anxiety.

Research using the SRS measure has demonstrated that adolescents with AIS report lower HRQOL in comparison to non-affected controls (Tones et al., 2006), and a review of studies identified that these differences are reported most consistently for the pain and self-image domains (Rushton & Grevitt, 2013). Notably, gender and age have been shown to influence

HRQOL. In studies with the general adolescent population, males have been shown to report better scores in overall HRQOL (Verma et al., 2010), and in the mental health domain (Verma et al., 2010; Daubs et al., 2014). In an AIS sample, males were found to have better scores in the pain and mental health domains compared to females (Diarbakerli et al., 2019). In a surgical cohort of patients with AIS, preoperative HRQOL scores demonstrated that males had better self-image and mental health than females (Roberts et al., 2011). In terms of age, lower age has been shown to correlate weakly with better scores across all HRQOL domains in the general adolescent population (Daubs et al., 2014). Looking to specific age categories, those aged 13 to 15 years reported significantly better activity, pain, and mental health compared to those aged 16 to 19 years (Daubs et al., 2014). Similarly in a sample of females with AIS of varying severity, age was weakly to moderately correlated with all HRQOL domains, with younger patients reporting better scores (Parent et al., 2009). Those who identified as Caucasian were also shown to report higher HRQOL scores in comparison to other ethnic groups (Verma et al., 2010; Daubs et al., 2014).

Among adolescents with AIS, curve severity and treatment status are factors which have previously been examined in relation to variation in HRQOL. Research has demonstrated that the severity of the spinal curve (i.e., increasing Cobb angle) is associated with HRQOL, such that patients with larger curves experience poorer HRQOL (Asher et al., 2003; Watanabe et al., 2005, Cheung et al., 2019). In particular, adolescents whose spinal curvatures progress severely to the stage of surgical consideration may be at increased risk of experiencing reduced HRQOL. Comparison of SRS scores among adolescents with curves of varying severity and in different treatment subgroups has indicated that presurgical patients with curves exceeding 50° report worse pain, self-image, and overall HRQOL compared to those with smaller curvatures (Parent et al., 2009). Berliner et al. (2013) reported similar results for adolescents with curvatures over 40°. Furthermore, some researchers have posited that length of diagnosis and the implications of a clinical diagnosis (e.g. treatment plans) may contribute to lowered HRQOL as adolescents did not perceive their HRQOL to be impaired when they were assessed just prior to diagnosis or when newly diagnosed (Rainoldi et al., 2015; Kontodimopoulos et al., 2018).

Undergoing bracing treatment has also been suggested to influence HRQOL among patients with AIS. Findings are mixed, with some studies reporting that bracing negatively impacts HRQOL and others reporting no difference between braced an unbraced patients (Pham et al., 2008; Weinstein et al., 2013). Cheung et al. (2019) reported that braced patients reported poorer HRQOL in all SRS domains compared to patients with smaller curves under observation. Conversely, Schwieger et al. (2016a) reported no differences in HRQOL or body image between braced and observed patients in a randomized controlled trial for bracing effectiveness. Despite the importance of clinical indicators in the treatment of AIS, it is notable that associations observed between AIS severity and HRQOL are small to moderate at best, highlighting the need to identify other factors that may influence HRQOL among patients with AIS (Asher et al., 2004; Parent et al., 2010; Berliner et al., 2013).

Body Image. Body image is a multifaceted construct that encompasses an individual's subjective perceptions, attitudes, and behaviours toward their body (Cash, 2004). A central component of body image is self-evaluation of one's appearance in terms of satisfaction – dissatisfaction (Cash & Pruzinsky, 2002; Kling et al., 2019). While adolescence is recognised as a vulnerable time period for body dissatisfaction due to factors such as increased sociocultural pressures and peer influences, varied trajectories of body image development have been identified, with some adolescents demonstrating high satisfaction throughout adolescence (Lacroix et al., 2020). Female adolescents have previously been shown to be more susceptible to body image dissatisfaction than males (Wertheim & Paxton, 2011), however research indicates that this gap is narrowing, perhaps due to increased sociocultural shifts in body acceptance and diversity (Karazsia et al., 2017).

Body image among individuals with chronic health conditions has received increasing attention as the significance and complexity of body image concerns among such populations has become apparent (Pruzinsky et al., 2004; Rumsey & Harcourt, 2004; Pinquart, 2013; Crerand et al., 2017). Adolescents who have a chronic health condition, particularly one that affects appearance such as scoliosis, have been shown to be less satisfied with their appearance in comparison to non-affected peers (Pinquart et al., 2013). In a meta-analysis including various chronic health conditions, Pinquart (2013) reported that children and adolescents with obesity, scoliosis, or cystic fibrosis were least satisfied with their bodies in

comparison to controls, with small to moderate effect sizes. Furthermore, research using the SRS measure of HRQOL demonstrates that self-image (or appearance satisfaction) is the domain that is most often found to differentiate adolescents with AIS from their non-affected peers (Ruston & Grevitt, 2013). While body image is partly captured under the self-image domain of the SRS-22r measure of HRQOL, the multifaceted nature of this concept warrants further investigation.

While having a health condition that changes physical appearance is recognized as a factor that can contribute to body image (dis)satisfaction, Cash's cognitive-behavioural framework of body image development asserts that there are many other factors that can have an influence on this outcome (Cash, 2012). Factors particularly relevant to the condition itself include severity of the condition, as well as the individuals' subjective perception of the appearance of the condition and its' visibility to others (Cash, 2012; Feragen & Stock, 2017). Aside from the condition itself, another key influence on body image satisfaction is appearance investment, defined as the salience of appearance to one's self concept, encompassing the cognitive, behavioural, and emotional importance that an individual places on their physical appearance (Cash et al., 2004; Jarry et al., 2019). Cash (2012) emphasises that while satisfaction with appearance is one key dimension of body image, investment is the second key dimension which has a core and direct influence on body image development and associated functioning. It is suggested that among individuals with appearance-altering conditions, greater investment in appearance may increase the likelihood of negative body image evaluations (Rumsey et al., 2008; Crerand et al., 2017). Other more widespread influences include sociocultural standards or appearance ideals (Cash, 2012) that can exacerbate the pressures on those with conditions affecting appearance (Rumsey & Harcourt, 2004), while more proximal events such as teasing or situations where appearance is emphasized or scrutinized can also impact upon satisfaction with one's appearance. Furthermore, interpersonal influences through peers (e.g. importance of conformity) and family (e.g. parental modelling) are also likely to be particularly relevant toward body image development and satisfaction among adolescents (Wertheim & Paxton, 2012). Overall, this range of factors highlights the multidimensional nature of body image and associated appearance satisfaction, which needs to be considered in order to better understand the experiences of those with chronic conditions such as AIS.

Clinical observation suggests that patients with AIS often wish to hide the appearance of their condition from others, which could lead to social difficulties associated with the fear that others will notice their spinal condition (Reichel & Schanz, 2003). As described by Crerand et al. (2017), comprehensive investigation of body image among adolescents with appearance-altering conditions should include appearance-related distress and impairment in functioning. To comprehensively understand the impact of the appearance-altering nature of AIS, the Body Image Disturbance Questionnaire-Scoliosis (BIDQ-S) was developed by Auerbach et al. (2014). It was based on the original Body Image Disturbance Questionnaire, whereby the measurement of disturbance goes beyond “dissatisfaction” with appearance and incorporates the impact of a negative body image on psychosocial functioning (Cash, 2004). Specific to those living with scoliosis, the BIDQ-S assesses the extent of appearance-related distress and its’ impact upon social or daily functioning (e.g., avoidance of activities, problems with friends, family, school, or work), and has been identified as an important measure in AIS research (Gallant et al., 2018). Research has shown that adolescents with AIS report higher levels of body image disturbance compared to non-affected peers (Auerbach et al., 2014; Wetterkamp et al., 2017), however previous results regarding the relationship between increasing curve size and levels of disturbance have been mixed. While Auerbach et al. (2014) originally identified no significant relationship between curve size and disturbance levels, subsequent studies identified a moderate relationship between increased Cobb angle and higher levels of disturbance (Wetterkamp et al., 2017) and higher disturbance in patients with curves exceeding 40° compared to those with smaller curves (Bao et al., 2015). Other research using this measure has shown that surgical intervention results in reduced levels of body image disturbance (Lonner et al., 2019).

While the literature continues to highlight body image as a significant issue in the treatment of patients with AIS (Gallant et al., 2018), a better understanding of the nature of appearance concerns in AIS is required. Carrasco & Ruiz (2014) have suggested qualitative research strategies to explore the specific and subjective aspects of appearance concerns and body image in patients with AIS. Importantly, the study of body image in the context of chronic medical conditions should seek to align itself with developments in body image research more generally, in terms of understanding the inherently subjective and multidimensional nature of body image concerns (Pruzinsky, 2004). Considering the multidimensional nature of body

image and the various factors that can contribute to body image development (Cash, 2012), research is needed to identify other contributors to variation in this outcome among adolescents with AIS, aside from clinical characteristics alone. This would allow better understanding as to why some adolescents with AIS may experience greater levels of body image satisfaction or disturbance.

2.3.3. Patient and Family-Centered Care

Authors highlight that a biopsychosocial orientation toward chronic conditions should include a focus on the family system as the most immediate social context to the patient (Rolland, 2004; Alderfer & Stanley, 2012; Kazak et al., 2017). Building on the broadening BPS perspective that highlights the centrality of the patient experience, is the concept of Patient and Family-Centered Care (PFCC). Indeed, PFCC emerged as an important health-related concept in the latter half of the 20th century (Eichner et al., 2012), and is particularly relevant in the context of paediatric health conditions given the role of parents in their child's healthcare. It is acknowledged that a paediatric health condition affects not only the child or adolescent, but also the family as a whole and the individuals within it (Kazak et al., 2017). PFCC values mutually beneficial partnerships among patients, families, and healthcare providers in addressing the medical and psychosocial health of patients and the needs of their families (Eichner et al., 2012; Kokorelias et al., 2019).

Within the paediatric and broader health-related literature, various "centeredness" terms including patient-, child-, person-, or family-centered have been used to describe the focus of care. Reviews of these centeredness terms reveal many similarities, and while person-, child-, or patient-centered approaches emphasise the perspectives and needs of the individual, family-centered care highlights the need to collaborate with and support the family to help meet their needs and in turn, meet the needs of their child (Hughes et al., 2008; Coyne et al., 2008). The concept "patient and family-centered care" (PFCC) is adopted in order to explicitly integrate the focus on the patient's individual experience and the role of their family in care (Eichner et al., 2012; Gallo et al., 2016). Importantly, PFCC does not take away from the importance of the individual patient perspective, rather it emphasizes the broader family context that the patient is situated in (Clay & Parsh, 2016). Of note, while the term "family" is a broad definition that captures varied family structures and generations, the focus of

family-centeredness in paediatric psychology has typically been directed toward parents given their responsibilities in their child's care (Alderfer, 2017).

A PFCC approach acknowledges that the perspectives of paediatric patients and their families are essential to informing high quality clinical care (Gallo et al., 2016). Key components of PFFC include tailored information provision, involving families and patients in decision-making, providing or ensuring formal and informal support to patients and their families, and involvement of patients and their families in research (Eichner et al., 2012). Many benefits of the PFCC approach are acknowledged and these include improving the experiences of patients and parents impacted by paediatric health conditions, improving patient outcomes, increased satisfaction with care, and more effective use of healthcare resources (Eichner et al., 2012; Alderfer, 2017). In relation to AIS, previous literature has not yet adopted the term of patient and family-centered care, however a move toward this approach is suggested by studies which have begun to incorporate parent perspectives (e.g., Narayanan, 2008; Bull & Grogan, 2010).

2.3.4. Parent Perspective

As described, when taking a PFCC approach to paediatric health conditions, parents are typically the focus as key members of the family unit who are often responsible for aspects of their child's healthcare. It is understood that parents of children with a chronic health condition face illness-specific demands that can present responsibilities and challenges above those ordinarily associated with parenting (Smith et al., 2013). Parents play a central role in their child's development, and parental mental health and wellbeing are identified as primary concerns after diagnosis of a paediatric health condition (Kazak et al., 2017). It is acknowledged that the diagnosis of a paediatric health condition can affect the wellbeing of the parents, with increased levels of stress, depression, and adverse health compared to parents of non-affected children reported in the literature (Pinquart, 2018; 2019; Miodrag et al., 2015). Specific issues that could impact upon parents include emotional distress caused by the diagnosis itself, changes in family roles, illness management responsibility, and social disruption (Smith et al., 2013). A recent study by Wang et al., (2019) examined mental health among parents of adolescents who were due to undergo surgery for AIS or were undergoing bracing treatment. Compared to a control sample, parents in the AIS group reported

heightened levels of anxiety and depressive symptoms, suggesting the importance of attending to the psychosocial needs of this parent group (Wang et al., 2019).

The potential impact of AIS on the parents of those affected can be understood from a family systems lens which emphasises the interrelations between family members (Alderfer & Stanley, 2012). In the context of a paediatric health condition, the diagnosis phase can be particularly challenging for parents which may involve complex feelings of loss, guilt, or fear, while longer-term adjustment to condition management can lead to stress over time (Smith et al., 2013; Cousino & Hazen, 2013; Emerson & Bögels, 2017). As described by Rolland (2004), the family-systems illness model suggests that the impact of a condition or illness on the family can be conceptualised in terms of a timeline with different 'phases,' in order to appreciate the changing demands, landmarks, or transitions that occur. Depending on the nature of the condition (e.g., fatal, nonfatal, acute, gradual), such a timeline can include a crisis phase surrounding prediagnosis and diagnosis, an adjustment period, a chronic 'long haul' adaptation, and a terminal phase with mourning and loss (Rolland, 1994). Considering AIS in this way, families could be expected to go through the initial diagnosis phase when their child's AIS is detected, followed by an adjustment or adaptation phase during which the condition can progress. If surgery is required, the challenges specific to the pre to postoperative periods are also likely to be significant periods on this timeline.

A number of previous studies related to parents of children with AIS have investigated parents' perceptions of their child's HRQOL and appearance in comparison to child reports, to determine the level of agreement between the two perspectives. While a study with newly diagnosed adolescents and their parents indicated no significant differences between child and parent reports of HRQOL (Brewer et al., 2014), a study with a surgical sample showed that parents tended to positively overestimate patients self-image and satisfaction scores at pre and postoperative time points (Rinella et al., 2004). In terms of scoliosis appearance, research is also mixed, as some studies have indicated high correlation or no significant differences between patient and parent perceptions of appearance (Sanders et al., 2003; Misterska et al., 2014), with another study indicating that parents rated the appearance of the scoliosis as worse in comparison to the child at the preoperative stage (Sanders et al., 2007). While these studies provide insight into levels of agreement between parents and their

children, or potential for differing perspectives, they do not provide much insight into parents' personal experiences or needs.

Other research relevant to parents of children with AIS has appeared to be relatively heterogenous in terms of topics covered. For example, Bull & Grogan (2010) qualitatively explored the experiences of parents throughout their child's surgery for AIS, while Wang et al. (2019) examined depression and anxiety symptoms among parents during the presurgical phase. There is a need to synthesize what is known about parents' experiences in order to more comprehensively understand parental perspectives, concerns, and needs in the context of their child's AIS and to guide future work on this topic. While literature reviews detailing the psychosocial impact that AIS can have on affected adolescents are available (Tones et al., 2006; Gallant et al., 2018), no review on any aspect of parents' experiences in the context of AIS have been completed. Moreover, greater investigation into parental experiences relevant to their child's AIS would provide the insight needed to inform and enable a more PFCC approach to AIS care.

2.4. Summary

Living with AIS is challenging for adolescents and their parents, considering the often progressive nature of the condition, physical symptoms, and treatments involved. In line with a biopsychosocial perspective, it is important to understand the lived experiences of this patient group and the psychosocial impact that AIS can have on those affected. Overall, limited previous research has sought to understand AIS from a psychosocial perspective. Across the previous literature concerning this topic, health-related quality of life (HRQOL) and body image have been emphasized as two key indicators of psychosocial wellbeing among the AIS patient group. However, it is evident that previous research has focused on validating the measurement of these constructs and how clinical characteristics can impact upon them. Clinical characteristics such as size of the scoliosis curvature can influence the psychosocial wellbeing of adolescents with AIS, in terms of their HRQOL and body image. However, it should be noted that these relationships are typically small to moderate, thus highlighting the need to identify other factors or processes leading to variation in these outcomes. Furthermore, the value of adopting a patient and family-centered approach was highlighted,

specifically acknowledging the role of parents in their child's AIS care. This places importance on capturing perspectives of both adolescents and their parents.

Under the broad aim of furthering current understanding of psychosocial aspects of adolescent idiopathic scoliosis from a patient and family-centered perspective, and considering limitations of previous literature, the thesis sought to address three main research questions: (1) How do adolescents experience living with idiopathic scoliosis? (In the presurgical period); (2) Are psychosocial processes associated with variation in wellbeing among adolescents with idiopathic scoliosis? and (3) What are parents' experiences related to their child's adolescent idiopathic scoliosis?

2.5. Multimethod Approach

A programme of four complementary research studies was conducted to address the research questions. Multiple differing methods were used, including two qualitative interview studies, a survey-based quantitative study, and a systematic literature review. In the literature, a multimethod approach refers to research that involves the use of two or more studies using different methods, which address interrelated research questions or contribute toward the same programmatic goal (Anguera et al., 2018). An advantage of this approach is the flexibility it affords to use different types of research data and analysis that are most suitable for the study aims (Hesse-Biber, 2015; Frost & Shaw, 2015). For example, while qualitative methods represent a valuable approach to gain insight into lived experiences of patients and their family members (Wu et al., 2016), quantitative approaches carry different advantages by allowing statistical inferences to be made regarding the population of interest. The four studies, which are detailed across five chapters, are briefly introduced below.

2.5.1. Programme of Research

Qualitative Study with Adolescents (Chapter 3). The aim of this study was to explore adolescents' experiences of living with AIS at the presurgical stage of treatment. Despite the challenging nature of AIS in terms of its' physical and psychosocial consequences (Weinstein et al., 2008), limited previous research concerning AIS has sought to understand the patient perspective. This study addressed the need for more in-depth insight into the lived

experiences of adolescents, focusing on those with significantly progressed curvatures at the stage of surgical consideration.

Quantitative Survey with Adolescents (Chapters 4 & 5). This study addressed the need to better understand the presentation of patients with AIS, in terms of their HRQOL and body image, by building on previous research which has focused on how clinical characteristics may influence these outcomes. Specifically, the contribution of coping strategies to HRQOL was examined, as coping is a psychosocial process shown to influence outcomes among paediatric populations (Compas et al., 2012; Oppenheimer et al., 2018), yet has not been comprehensively investigated in AIS. The contribution of appearance-related cognitions (i.e. perceived visibility, subjective perception of appearance, and appearance investment) to body image outcomes was also examined, recognising the multidimensional nature of body image development (Cash, 2012). A secondary objective was to assess the patient perspective on their support preferences, as little is known on this topic (MacCulloch et al., 2009).

Systematic Review relevant to Parents (Chapter 6). This study sought to synthesize what is known about parents' experiences in relation to their child's AIS. In line with PRISMA guidelines (Moher et al., 2009), a mixed studies systematic review was completed to comprehensively capture the available literature on this topic (Pluye & Hong, 2014). Considering the potential impact that AIS can have on the wellbeing of parents of those affected (e.g., Wang et al., 2019), the systematic review addressed the need to evaluate previous research concerning the role of parents and their needs in order to continue progress toward a patient and family-centered approach to research and care in AIS.

Qualitative Study with Parents (Chapter 7). The aim of this qualitative study was to explore parents' experiences in relation to their child's AIS from diagnosis to presurgical preparation. The review presented Chapter 6 highlighted that most previous research has tended to focus on the immediate surgical period (e.g., Chan et al., 2017; Bull & Grogan, 2010), and that understanding of parents' experiences throughout the trajectory of their child's AIS prior to surgery is particularly limited. Therefore, this study addressed the need for further investigation into the period prior to surgery, from parents perspectives.

Chapter 3

Living with Adolescent Idiopathic Scoliosis: A Qualitative Study of Adolescents' Psychosocial Experiences prior to Surgery

Chapter Overview

This chapter presents the qualitative interview study that was conducted with a cohort of adolescents with adolescent idiopathic scoliosis (AIS), who were in the presurgical stage of treatment (*N* = 14). This study gained an in-depth insight into the lived experiences of adolescents with presurgical AIS and furthered current understanding of the psychosocial impact of this paediatric condition. Adolescents participated in semistructured interviews exploring the impact of their spinal condition on their lives and data were analysed using reflexive thematic analysis to construct meaningful themes. Firstly, relevant background for this study is provided, followed by a detailed account of the method and analytic approach. The resulting themes are then presented and discussed in relation to their research and practical implications. This study has been published in similar format in the *Journal of Pediatric Psychology* (Appendix A).

3.1. Introduction

Adolescent idiopathic scoliosis (AIS) can pose a unique set of challenges for affected adolescents, who are already experiencing the normative developmental challenges of adolescence including pubertal changes and a desire for peer conformity (Reichel & Schanz, 2003; Christie & Viner, 2005). Adolescents with AIS are more likely to report appearance self-consciousness (Auerbach et al., 2014) and higher levels of body image dissatisfaction compared to nonaffected peers (Tones et al., 2006; Rushton & Grevitt, 2013). Considering that AIS can be associated with body image disturbances, concerns have also been raised that this group may be vulnerable to other psychosocial difficulties such as symptoms of depression or anxiety (Gallant et al., 2018). Sanders et al. (2018) reported that those living with AIS experience similar levels of anxiety symptoms to paediatric patients with cancer.

Adolescents whose spinal curvatures progress to the stage of surgical consideration may be at increased risk of experiencing psychosocial difficulties. Previous studies comparing SRS-22 scores among adolescents with curves of varying severity at different stages of treatment have indicated that overall health-related quality of life and self-image are more negatively affected in presurgical patients compared to those not requiring surgery (Parent et al., 2009; Berliner et al., 2013). The asymmetry of the spine and trunk caused by scoliosis can be particularly prominent in adolescents who progress to surgical consideration, and the desire to look more 'normal' is a factor shown to influence adolescents' decision to have surgery (MacCulloch et al., 2009). Furthermore, the time period in the lead up to surgical intervention can be an anxiety-provoking time, as adolescents engage in preoperative preparation and are informed about the extensive surgical procedure (Rullander et al., 2016).

Despite the challenges of living with AIS and the seriousness of undergoing spinal fusion surgery, limited research has sought to explore adolescents' psychosocial experiences specifically during the presurgical stage of treatment. Recently, Lonner et al. (2020) surveyed 44 patients about the most important aspects of their lives that were affected by AIS in the presurgical period, and showed that sports, general function, and general fitness were the most common concerns, and that improving their self-esteem and pain were the most common operative aspirations. Studies by Chan et al. (2017) and Bridwell et al. (2000) surveyed presurgical patients with AIS specifically about their surgical concerns and identified primary surgical concerns as postoperative pain, their ability to return to activities after surgery, and the possibility of surgical complications (e.g., neurologic injury).

One qualitative study by MacCulloch et al. (2009) investigated information and support needs among 11 adolescents with AIS who had undergone or were anticipating spinal surgery. Participants endorsed development of a customised website including information on topics including what to expect at the hospital, managing recovery at home, and how school or peer relationships may be impacted post surgically. To meet support needs, adolescents wanted the resource to include the opportunity for peer support and interactive discussion where personal stories could be shared. While MacCulloch et al. highlighted important considerations for informed surgical decision-making, the focus on surgical information needs precluded a more holistic exploration of adolescents' experiences throughout the presurgical

period. The remainder of qualitative studies of AIS have described challenges associated with treatment (Donnelly et al., 2004) or explored adolescents' experience of the postsurgical period, focusing on postoperative pain and recovery (Rullander et al., 2013; Bray & Craske, 2015; Honeyman & Davison, 2016; Perry et al., 2018). These findings demonstrated how difficult adolescents found the postoperative recovery period, in terms of pain, limited capacity to partake in activities, and losing contact with friends (Rullander et al., 2013; Perry et al., 2018). However, adolescents also felt relieved that they had undergone surgery and that the challenging perioperative experience had been 'worth it' (Bray & Craske, 2015; Honeyman & Davison, 2016). Although these studies focused on postsurgical patients, some detail of the presurgical phase was discussed, including adolescents' recollections of experiencing shock at the need for surgery and fear about possible surgical complications (Rullander et al., 2013; Honeyman & Davison, 2016).

Overall, a comprehensive and in-depth exploration of the psychosocial experiences among adolescents living with presurgical AIS is lacking. Importantly, understanding the experiences and perspectives of adolescents throughout the presurgical period is a key step in informing patient-centered care throughout this challenging stage of treatment. Such insight would allow identification of possible psychosocial support needs and how they could be addressed, information provision that is tailored to adolescents' concerns, and improvement of preoperative preparation for this patient group. Furthermore, the presurgical period is an opportune time to provide psychosocial support, as adolescents are typically reviewed clinically approximately every 6 months to monitor curve progression during growth (Altaf et al., 2013) and are linked in with a multidisciplinary team.

3.1.1. The Present Study

Adolescents at the presurgical stage of AIS treatment are living with significantly progressed spinal curvatures that have potential to negatively impact their psychosocial functioning and wellbeing. However, limited research has explored the psychosocial impact of AIS from the patient perspective and how those with AIS manage the consequences of their spinal condition during their adolescent years. In particular, to our knowledge no study has explored the lived experiences among a presurgical cohort of patients with AIS. The present study therefore employed a qualitative approach to explore the experiences of adolescents living

with presurgical AIS, in order to enhance our understanding of the impact that this condition can have on adolescents' lives from a psychosocial perspective. Through this exploration, this study will gain important insights into possible psychosocial support needs of this group and findings will help to inform clinical practice.

Aim and Objective

The aim of this study was to understand adolescents' experiences of living with presurgical idiopathic scoliosis, with a focus on their psychosocial wellbeing. The objective was:

(i) To explore and describe the experiences of adolescents living with AIS at the presurgical stage of treatment.

3.2. Method

This study adopted a qualitative approach to explore the experiences of presurgical adolescents with AIS. Qualitative methods are recognised as a valuable approach in gaining an in-depth understanding of the perspectives and needs of paediatric patient groups (Wu et al., 2016; Berlin et al. 2017) and this approach has thus far been underutilised in AIS research.

3.2.1. Participants and Recruitment Procedures

3.2.1.1. Eligibility Criteria

Adolescents were eligible to participate if they were aged 12 to 18 years, diagnosed with AIS, and were presurgical candidates who had not yet undergone surgical intervention for AIS. A purposive sampling technique was employed, an approach that recruits participants who share specific characteristics relevant to the research topic (Patton, 2002). Both males and females meeting the inclusion criteria from urban and rural locations across Ireland were invited.

3.2.1.2. Recruitment

Adolescents with AIS were recruited to participate via the orthopaedic department of an Irish children's hospital, a national tertiary referral centre for paediatric scoliosis care. Between October 2018 and May 2019, eligible adolescents and their parent or guardian(s) attending a routine presurgical outpatient appointment for their scoliosis were informed about the study

by a spinal nurse specialist to assess their interest in participation. The purpose of the study, to understand the experiences of adolescents with presurgical AIS, was explained to potential participants. During recruitment, the parents of 16 adolescents agreed to be contacted by the research team with further information and, of these, 14 chose to participate. Reasons for nonparticipation included undergoing surgery before an interview date could be scheduled and unsuitable timing of the study. The first author followed up with each parent by phone or email and arranged a meeting for data collection either in the hospital or participant's home setting. Participants and their parents were provided with detailed age-appropriate study information materials (printed and/ or via email) prior to data collection and were given the opportunity to discuss any queries. Parents provided written consent and adolescents provided written assent to participate.

3.2.1.3. Participants

The final sample included fourteen adolescents (ten females and four males) with thoracic or thoracolumbar idiopathic scoliosis, aged 12 – 17 years (*M* = 14.6). The higher proportion of females in the sample was reflective of the higher incidence of AIS among females (Konieczny et al., 2013). All participants identified as White Irish. Mean time since diagnosis was 12.7 months (range: 3 – 26 months). At time of interview all participants were considered surgical candidates based on the size of their spinal curve and/or potential for future curve progression and had discussed prospective surgery with their medical team. Cobb angles of the major scoliotic curves on participant's most recent radiographs ranged from 46 - 100° (*M* = 68°, *SD* = 14.8). At the time of the study, two participants had a confirmed surgery date. The remainder were waiting to receive a date or anticipated that they would be scheduled for surgery in future. Two participants had also worn an orthotic brace as part of their treatment. Participant characteristics are summarised in Table 3.1, aggregated for confidentiality purposes.

Table 3.1

Participant Characteristics for Qualitative Study with Adolescents (N = 14)

Variable		
	Range	*M*
Age (years)	12 - 17	14.6
Curve Size[a]	46 - 100°	68°
	n	*%*
Gender		
Male	4	28.6
Female	10	71.4
Home Setting[b]		
Urban	5	35.7
Rural	9	64.3
Length of Diagnosis		
<1 year	5	35.7
1-2 years	8	57.1
2-3 years	1	7.1
Curve Type		
Thoracic	6	42.9
Thoracolumbar	8	57.1
Treatment Details		
Bracing	2	14.3
Awaiting surgery	10	71.4
Surgery scheduled	2	14.3

[a] Curve size is the surgeon assessed Cobb angle measurement of the major scoliotic curve. [b] Based on the Central Statistics Office geographical classification guidelines.

3.2.2. Data Collection

Prior study approval was obtained from the University College Dublin Human Research Ethics Committee (HS-17-05-Motyer-Fitzgerald) and from the collaborating hospital's Medical Research Ethics Committee (GEN/546/17). The lead researcher (GM), a PhD candidate in Psychology, conducted the interviews in person, either in the participant's home (n = 11) or in the outpatient department of the hospital (n = 3). The location of the interviews was dependent on participant preference. Before interview, a brief demographic questionnaire was used to collect detail on participants' age, gender, ethnicity, home setting (rural/ urban), length of diagnosis, and treatment status. The semistructured interview schedule was developed based on prior literature in accordance with qualitative research guidance (Braun

& Clarke, 2013; Robson & McCartan, 2016; Kirk, 2007), and was reviewed by an orthopaedic specialist and paediatric health psychologist. The schedule consisted of open-ended questions and related probes that encouraged participants to discuss their experiences of living with AIS in a flexible manner. The semistructured interview provided an ideal format for the collection of rich data as there was scope for the participant to raise topics important to them while also ensuring a level of consistency across interviews as participants were asked the same key questions (Braun & Clarke, 2013). Interview topics covered the diagnosis of scoliosis, an exploration of what areas of their lives were affected by scoliosis, concerns or challenges associated with scoliosis, body image, and their medical treatment (i.e., prospective surgery). For further detail on data collection procedure and a summary of the interview schedule procedures see Appendix C.

While the interviews were private and confidential, participants and their parents were made aware of the limits of confidentiality and the researcher's duty of care (i.e. if a participant disclosed information indicating they were at risk or in significant distress, the parent or relevant medical professional would be informed). Given the potential for sensitive issues to be raised in qualitative health research with young people, it is considered best practice to provide clarity on confidentiality in this way (Duncan et al., 2009). Interviews were audio-recorded and lasted between 15 and 48 minutes (*M* = 32 minutes). Participant recruitment and data collection ceased when GM and AF judged that the dataset contained sufficient information power to generate meaningful themes and no new topics of discussion were being introduced in interviews. Information power relates to whether the data demonstrates the required breadth and depth relative to factors including the homogeneity of the sample and the scope of the study (Morse, 2000; Malterud et al., 2016) and is a helpful concept in determining appropriate sample size in exploratory inductive qualitative studies. Previous qualitative studies conducted with this patient group that had a comparable scope and sample specificity reported samples of 12 (Donnelly et al., 2004) and 11 (MacCulloch et al., 2009). Our final sample (*N* = 14) slightly exceeded those previously reported, further supporting that adequate information power to address our research objectives had been obtained. Interview data were transcribed verbatim, and any potentially identifying information was anonymized at the stage of transcription.

3.2.3. Data Analysis

Data were analysed using reflexive thematic analysis (TA) with an inductive approach (Braun & Clarke, 2006; 2019). Reflexive TA is a method that involves a process of systematically coding patterns of meaning across the dataset and it was chosen for this study to develop a rich thematic account of participants' experiences of their AIS through analytic narrative and data extracts (Braun & Clarke, 2020). In line with our focus on participants' experiences, the qualitative analysis was underpinned by a critical realist or contextualist perspective, recognising that knowledge is accessible through the lens of participants' subjective experiences and perspectives (Braun & Clarke, 2013; 2021). Within this approach, knowledge is not independent from participants contextually situated unique realities (Braun & Clarke, 2021), and findings were therefore said to represent a reflection of reality based on the adolescents' perspectives. The inductive approach suited the exploratory and experiential nature of this research as we focused on developing themes grounded in the data rather than utilising predetermined codes (Braun & Clarke, 2019). Furthermore, the chosen analysis was particularly suited to analysing interview data, for providing implications for clinical practice, and for presenting findings in an accessible format suitable for multidisciplinary settings (Braun & Clarke, 2006; 2014; 2020).

Following the steps of reflexive TA, GM familiarised themselves with the data and conducted initial coding of the transcripts with an inductive approach (i.e., exploratory & data-driven). In this way, themes were to be generated using a bottom-up process. Coding (defined as the process of identifying units of meaning within the dataset) was an iterative process that involved revisiting transcripts and reviewing codes as the analysis progressed. GM and supervisor AF independently coded approximately one third of the dataset at the beginning of the coding phase and again midway through analyses when a more developed knowledge of the dataset had been established. They met regularly to collaboratively discuss interpretations of the data and theme development. This collaborative approach was intended to promote a rich and rigorous analysis and enhance researcher reflexivity, rather than as a reliability check. Codes were clustered into broader units of meaning which were refined to create themes and a narrative describing each theme was developed. A small number of subthemes were included in the thematic narrative to highlight specific notable

aspects of the themes' central organising concepts (Braun & Clarke, 2019). NVivo 12 software was used to facilitate data management.

Recommended strategies for the promotion of good quality reporting and rigour in qualitative research were incorporated throughout this study (O'Brien et al., 2014; Nowell et al., 2017; Braun & Clarke 2006). Originally proposed by Lincoln and Guba (1985), the dimensions of trustworthiness considered in this study included credibility, dependability, and transferability (Nowell et al., 2017). To promote credibility in line with reflexive TA (Braun & Clarke, 2019), critical self-evaluation of the researcher's influence (e.g., through keeping reflective notes, and supervisory debriefing) alongside the collaborative approach to data analysis (described above) facilitated engagement in researcher reflexivity and promoted a thorough analysis grounded in the data. Importantly, reflexive TA acknowledges the role of the researcher in the knowledge production process and engaging in reflection through these strategies aided the analytic process (Berger, 2015). Dependability was enhanced through transparent and detailed reporting of the study in line with standards for reporting qualitative research (SRQR; O'Brien et al., 2014). A clear account of participant recruitment and the characteristics of the sample were provided to contextualise the findings and provide guidance on transferability.

3.3. Results

Analysis of the interview data generated four key themes which reflected pertinent aspects of adolescents' lived experiences prior to surgery. Themes included *"Proceeding with Caution," "Am I Different?'," "An Emotional Journey,"* and *"No Pain, No Gain."* Thematic findings and illustrative data extracts are presented in Table 3.2. Quotation's representative of participants' experiences across the dataset were embedded throughout the thematic narratives, and participants are specified by participant numbers (e.g., P1) followed by M or F (male or female), and their age.

Table 3.2

Thematic Findings for Adolescents with Idiopathic Scoliosis

Themes	Sample Data Extracts
1. Proceeding with Caution	*"I used to be able to play hurling in P.E. and I can't do that anymore now because we'd be kind of rough with it." (M, 14). "I'm just making sure I'm not straining it too much just in case it might get worse." (F, 16).*
2. Am I Different?	
2a. Appearance Changes	*"it's twisted my ribs and I have a lump here [on my side]." (M, 16). "I was just like looking at myself in the mirror and I was like my body looks a little bit odd, like it kind of looks lopsided…" (F, 15).*
2b. A Hidden Condition	*"I asked a few of my friends [about my AIS] and some of them said that they actually never realised." (F, 15).*
3. An Emotional Journey	
3a. Emotional Rollercoaster	*"I was just a bit shocked [at diagnosis] 'cause like I didn't know what it was and I didn't know if it was real threatening or anything." (F, 16). "I think a lot about my back and kinda how scary the operation might be and sometimes I get into a panic and my back starts to hurt even more." (F, 16).*
3b. Not the Only One	*"It's just knowing that there's so many other people as well it's reassuring that you're not kind of… different I suppose." (F, 15).*
4. No Pain, No Gain	*"The surgery will make me less conscious of my back, and I can wear a normal top again instead of the big baggy ones." (F, 14). "I think the surgery kind of scares me a bit but I know it's going to help me and, in the end, I know it's the right thing to do." (F, 13).*

[a] In brackets F = female, M = male, followed by age.

3.3.1. Thematic Findings

3.3.1.1. Theme 1: Proceeding with Caution

The first theme captures how participants approached activities with increased caution as they managed the physical symptoms of their scoliosis and avoided straining their back in the presurgical period. It was evident that adolescents were cautious of activities that may aggravate their condition. Pain or discomfort was commonly discussed and could occur when

sitting for long periods at school, when carrying bags, or after physical activity. Some of the adolescents appeared to be aware of their own limits and avoided activities that could cause back pain and they rested when needed. This was demonstrated by an adolescent who recalled attending a school trip where she could not participate to the same level as her peers: *"they were doing emm, kind of like bungee jumping and I just chose not to do it because I went rock climbing with them before that and I was in bits after so I just didn't want to risk it… it looked fun though." (P5, Female, 16).* Despite avoidance of certain activities or sports, it was evident that most of the adolescents continued to remain active, albeit with increased caution; *"I do taekwondo and I can't do like kind of self-defence where you knock people to the ground anymore because obviously my back would get hurt." (P4, Female, 14).*

Some adolescents could no longer participate in sports and activities to the same level as before their scoliosis, which could lead to feelings that they were missing out. This was particularly evident for high impact or contact sports; *"…my friends started after me doing boxing, and then they got to go on, and like, go to all the competitions and win all the medals and stuff like that, and I found out [that I had scoliosis] just before I was going for the competition level, and I couldn't do it." (P9, Male, 14).* As well as avoiding specific activities, day to day life could take its' toll physically; *"you have to be careful, and you know I might say well I'm only going to go out for that long because I'll have to go lie down then for a while and just take it easy." (P2, Female, 13).*

Adolescents felt that it was important to follow advice from medical staff in relation to what to avoid, what activities could be helpful, as well as keeping healthy in advance of their surgery; *"I just try and do everything the doctors say like eat healthy, like vitamins… and the dietician was like you need to be like, perfect health and stuff, so listening to them and going swimming and everything, that like helps me feel better cause like how could it go wrong then." (P7, Female, 15).* This participant, like others, had taken up swimming since their diagnosis, a form of exercise often recommended for those with AIS as a noncontact activity that can increase fitness and mobility in advance of undergoing surgery.

Importantly, for adolescents who were participating in sports and exercise, they felt it was helpful to them in the presurgical period; *"Just the fact that I play sport kinda helps me like*

and just gets my mind off [my scoliosis], I would just totally forget about it there." (P3, Female, 12). As noted here, at a time when preferred activities may be on hold, taking part in exercise as recommended by the medical team may be just as beneficial for adolescents' mental health as it is for maintaining presurgical fitness.

3.3.1.2. Theme 2: Am I Different?

The second theme describes the experiences of adolescents as they process their changing bodies and question how their appearance may be perceived by others, including two subthemes. *Appearance Changes* focuses on adolescents' perceptions of their condition and how it has affected their appearance. Relatedly, *A Hidden Condition* captures how adolescents manage the visibility of their condition and how they may navigate this in the context of their peers.

Subtheme: Appearance Changes. All of the adolescents acknowledged changes in their physical appearance as a result of their scoliosis. Changes were typically associated with truncal asymmetries, as adolescents described their backs, shoulders, ribs, hips, and posture as uneven. Noticing irregularities in their appearance was often what led adolescents to seek medical advice and subsequently be diagnosed with AIS; *"When I was getting out of the shower one day I just kinda noticed in the mirror like, it was kinda odd like, a bump at the lower back and my shoulder blade was kind of gone out a bit […] I had no idea what it was, I thought maybe it could be a tumour or something." (P10, Male, 17).* Similar to this adolescent, many could remember the first time they noticed visible signs of their spinal condition and the confusion that surrounded it.

Throughout interviews, words were used which indicated that participants perceived their condition as 'abnormal', and they felt different to their peers; *"When we were all getting in our swimsuits and going to the beach and all that… I didn't want to get into my swimsuits because my body didn't look right." (P6, Female, 15).* For some participants, the impact that their scoliosis had on their appearance was particularly burdensome, as articulated by an adolescent who felt she could no longer wear clothing that she liked, and that corrective surgery was necessary in order for her to be happy with her body again: *"I wouldn't be able to live with it… because I just wouldn't be able to like my body." (P7, Female, 15).*

There was variation among adolescents in relation to appearance concerns, as half of the participants indicated that in general, they were content with their looks despite scoliosis-related appearance changes. It was apparent in some instances that there may have been a disconnect between how adolescents' felt about the appearance of their scoliosis and how they felt about their overall or general body appearance. For example, this was demonstrated by an adolescent who felt she was 'very different looking' from her peers and was concerned about how she looked in certain clothes, but also mentioned that she was a "confident person," and that she was reasonably happy with her looks: *"like it does bother me that I have it [scoliosis] but emm, I don't mind looking the way I do." (P5, Female, 16).* Another adolescent described a number of visual symptoms of her scoliosis (e.g., unlevel shoulders) but also perceived that her body image satisfaction was similar to other teenage girls: *"They [appearance changes] don't really bother me too much, like I wouldn't be 100% happy with how I look, but I think that's even what most girls are like really." (P13, Female, 16).*

Subtheme: A Hidden Condition. Although adolescents observed changes in their appearance, there was uncertainty as to whether others could see the physical changes in their bodies, or how visible their scoliosis was to others. Many adolescents believed their scoliosis was not very noticeable; *"I don't think they (peers) notice it, being honest, I never talk to them about it. I think it's just me because I see it every day." (P11, Female, 14).* They acknowledged that their scoliosis would be more visible in certain situations such as when swimming, wearing tighter or more revealing clothing, or while getting changed for P.E. at school.

Some adolescents made efforts to disguise or hide their condition from others: *"when I found out I was very insecure I didn't know what it looked like to other people, and I was trying to like, straighten it as much as I could…" (P8, Female, 15).* Different camouflage strategies discussed by adolescents included wearing bigger or 'baggy' clothing, being vigilant of their posture, and changing quickly at school. One adolescent reported actively trying to reduce her body asymmetry by losing weight and building muscle at the gym. The perception of scoliosis as 'abnormal' and a condition of limited awareness within the population seemed to exacerbate adolescents' concerns about others seeing their scoliosis; *"If someone just broke*

an ankle or broke a bone you know you wouldn't really second look it, I feel like [scoliosis] is definitely something that people would look at and look again at, and be kind of like, 'oh what's that?' and it's kind of different, it's a lot different to what friends or you know, what they would look like." (P2, Female, 13).

Some adolescents demonstrated a reluctance to talk with their peers about scoliosis, either because they felt they were unable to explain it or because speaking about it would make them uncomfortable. For example, a participant had asked a peer to be his date for an upcoming social event, but worried about the fact that he had not told her about his scoliosis and potential surgery: *"Honestly, I'd feel a bit relieved [if I told her], but honestly I just don't know how to say it like in the right way." (P10, Male, 17).* Another adolescent had started to avoid going to youth club as he felt his scoliosis was becoming more noticeable and did not want to have to explain it anyone: *"I was trying to get it off my mind, stop thinking about it the whole time, and I'm like, if people were bringing it up the whole time I didn't want that happening so […] I just didn't want the hassle of explaining it to them to be honest." (P9, Male, 14).* There appeared to be a desire to maintain normality for many adolescents as they did not want to dwell on their scoliosis and endeavoured to *'get on with it'*.

Telling friends about their scoliosis came across as a difficult step, and while some adolescents appeared to tell close friends as a way of seeking social support, others may have told their friends out of necessity. For example, explaining why they missed school for hospital appointments or why they were not taking part in particular sports anymore. For those who told friends about their scoliosis, some peers had initially been surprised at the diagnosis indicating that the adolescents' scoliosis may not have been noticeable at the time. Confiding in close friends appeared to be helpful to adolescents, as they felt their friends reacted well and were supportive: *"it's nice to be able like… them not judging you about it like it's nice to be able to talk to them about how sometimes it's painful or.. sometimes you can't do things, like that you don't want to mention to other people." (P14, Female, 12).*

3.3.1.3. Theme 3: An Emotional Journey

From the time of their scoliosis diagnosis to preparing for surgery, it was evident adolescents could experience an *Emotional Rollercoaster* as described by the first subtheme. However, knowing that they were not alone in their experience of AIS could provide adolescents with support along this emotional journey as captured by the second subtheme *Not the Only One.*

Subtheme: Emotional Rollercoaster. Many adolescents recalled their shock and distress around the time of their diagnosis; *"I was definitely, my head was spinning a bit, I was thinking about everything that could happen and, just because I wasn't really expecting it to look like that at all, especially seeing the S [curvature on the X-ray]." (P13, Female, 16).* The majority of adolescents recalled having limited or no previous knowledge of scoliosis. For the couple of adolescents who had some previous exposure to the condition, their prior knowledge of the condition contributed to fear at diagnosis, perhaps because they were aware of the potential severity of the condition: *"I remember like seeing it on the Late-Late show with that girl and I said to mam, aww that'd be like the worst thing ever to have, and then like a week later I found out I had it so I was really upset." (P11, Female, 14).*

While some adolescents were clearly emotionally distressed and apprehensive following diagnosis of their condition, it appeared that others went through a process of change in their emotions, as they realized the severity of their condition over time. Reaching the stage of surgical consideration could be interpreted as a turning point for many adolescents, who may have not realised the severity of their condition until surgery was suggested as a treatment option by their medical team; *"Well I… when the word was first thrown out I didn't have any idea what scoliosis was emm, but it was explained to me what it was, and, well I wasn't really that worried at the time, but, I think, now like, recently, like it's kinda dawned on me." (P10, Male, 17).*

Adolescents could feel particularly down when their scoliosis caused them to miss out on activities with friends or impeded their capabilities in sports and activities. The process of choosing clothes to wear was also challenging for some as this could highlight appearance-related issues. The prospect of undergoing spinal surgery understandably caused fear and anxiety, which was likely to be heightened as the surgery date neared. For example, an

adolescent whose surgery was scheduled for less than two weeks following the interview described how the thought of surgery could influence her mood: *"I was at my friend's house a while ago and we were all having great fun and all that and then it [surgery] came into my head... and I actually wouldn't talk to anyone for about 20 to 30 minutes because I started freaking myself out you know?" (P6, Female, 15).* Despite their fears, adolescents also demonstrated considerable optimism in relation to their prospective surgery as they looked forward to improvements in their condition postoperatively.

Subtheme: Not the Only One. While experiencing a rollercoaster of emotions throughout the presurgical period, adolescents could gain some reassurance through realising they were not alone. In coming to terms with their condition, many adolescents gained emotional support through meeting or hearing stories about other young people with scoliosis, and through this they felt less isolated and could relate to the challenges that each other were going through. This was reflected in a participant's experience at a swimming group for young people with scoliosis: *"when you're there you kind of feel like there's other people and there's always someone worse than you and you feel… it's going to be okay like I'm not the only one." (P2, Female, 13)*. Some adolescents thought it was particularly helpful to meet someone who had already been through surgery as they gained practical advice about what to expect at the hospital and during recovery. Seeing the results of surgery provided reassurance; *"I was actually talking to one girl and I think she got [surgery] when she was like seventeen and it makes you more relaxed when you talk to people, like knowing that you're actually going to be fine after surgery." (P5, Female, 16).*

3.3.1.4. Theme 4: No Pain, No Gain

This final theme centered on anticipation of post-surgical benefits despite fears or concerns about surgery. Although the prospect of undergoing major surgery was a stressor for many adolescents, the operation also represented a life-changing event with positive outcomes and an opportunity to regain normality. This balance was reflected in adolescents' beliefs about their upcoming surgery; *"I'm nervous to get it done, but I know I'll be glad when it's over, 'cause in the long term as I said it would just be really good and helpful, and make me feel good, better about myself as well." (P13, Female, 16).*

Surgical concerns related to possible complications and the subsequent recovery period. Concerns included the risk of paralysis (although also acknowledged as unlikely), the appearance of the surgical scar, and the possibility of damaging their spinal fusion postoperatively. A couple of adolescents doubted the permanency of the spinal fusion by questioning if their scoliosis could return in future. Concern about their lives being interrupted by the surgery was also evident, as they anticipated a challenging recovery and considered what they might fall behind on in school, sport, or social activities. Adolescents were often keen to put the surgery behind them so that they could get on with their lives; *"honestly I think I just want it to be done and over with. It's a case of going in, it's over and then the recovery..." (P6, Female, 15).*

Long-term, many adolescents felt that undergoing surgery was important to prevent their scoliosis from progressing further and to avoid a more debilitating curvature in the future. Many also anticipated an improvement in the appearance of their body following surgery. Other positive outcomes included regaining normality in their lives, the ability to play various sports, and improved self-esteem. It was apparent that for some adolescents having surgery was believed to be the solution to the problems they were experiencing; *"Once I get the surgery done I feel like I'll be able to get back to normal." (P4, Female, 14).*

Similar to how the pros and cons of surgery were acknowledged, the two adolescents who wore a torso brace as part of their treatment reflected on both negative and positive aspects of their brace. Despite challenges including discomfort and a reluctance to wear the orthosis in social settings, adolescents appreciated the prognostic benefits of their brace; *"Well I don't mind wearing it, like I know it's doing good for me so that's always like a kind of lift up because I know it's doing me better." (P14, Female, 12).* These adolescents looked forward to no longer wearing the brace postoperatively.

Adolescents anticipated that they would benefit from the surgery, and this provided reassurance. It appeared that this allowed adolescents to remain hopeful and focused on the future, which potentially giving less weight to the challenges they were experiencing in the presurgical period. This was articulated by an adolescent in relation to appearance: *"Once I get the surgery the whole look of it is going to change. Right now, I'm not a huge fan of the*

way it looks but you know you just kind of have to, that's just the way it is, so em, I am unhappy about that but em I know that will change so kind of have that comfort I suppose." (P2, Female, 13). Adopting an optimistic mindset and focusing on an ideal surgical outcome in this way appeared to facilitate coping throughout the presurgical period.

3.4. Discussion

This study explored the experiences of presurgical adolescents with AIS from a psychosocial perspective and generated four key themes with associated subthemes providing rich insight into the experiences of this patient group. As depicted in the first theme "Proceeding with Caution," many adolescents faced physical limitations and pain because of their scoliosis and were conscious of their own limits to avoid straining their condition. Despite their AIS, many continued to keep active, albeit with increased caution. Next, "Am I Different?" captured adolescents' perceptions of their changing appearance as well as their uncertainty surrounding visibility of their scoliosis. There was variation in how these changes could affect appearance satisfaction, some attempts to hide their condition and difficulties in communicating about AIS with their peers. Furthermore, within the "Emotional Journey" adolescents discussed the various feelings they experienced in relation to their AIS, which overall equated to a rollercoaster of emotions. This included their shock at diagnosis, the daunting realisation of the severity of their condition and a mix of fear and optimism in advance of their surgery. Meeting peers with scoliosis could be a source of emotional support and reassurance. Finally, "No Pain, No Gain" related to their prospective surgery, as adolescents voiced surgical concerns but also acknowledged positive surgical outcomes. Many were keen to put their surgery behind them and focusing on positives could provide reassurance.

Adolescents' physical limitations and caution to avoid pain or injury are common experiences among young people with physical health conditions, with similar reports of 'missing out' identified in research concerning juvenile idiopathic arthritis (Tong et al., 2012). Despite physical constraints, many adolescents in this study continued to participate in sport and exercise. This is positive given the benefits of physical activity for overall health, aerobic capacity, and wellbeing in patients with scoliosis (Kakar et al., 2017), and considering the importance of recreational activities for social skills and friendship development during

adolescence. However, adolescents with AIS may be at risk of reduced participation in physical activity due to factors such as back pain and fear of injury (Kakar et al., 2017). It is generally recommended that those with AIS remain active where possible, however there is no definitive guidance available on suitable levels or types of physical activity for AIS at various stages of treatment (Tarrant et al., 2014). Adolescents were keen to follow guidance from their medical team at the presurgical stage and can seek clarity on activity participation in the clinical setting. The medical team therefore play an important role in using their expertise to guide those with AIS on activity participation throughout treatment.

Adolescence is a time of heightened appearance-related pressure (Rumsey & Harcourt, 2012) and developing an appearance altering condition such as scoliosis can threaten a desire for conformity with peers and healthy body image development (Auerbach et al., 2014; Gallant et al., 2018). Our findings demonstrated that although some adolescents were unhappy with the appearance of their scoliosis, others indicated they were generally happy with their appearance. Possible reasons for variation in adjustment to appearance change could be due to individual differences in appearance-related cognitions, such as levels of appearance investment or perceived visibility of the condition. As highlighted in recent research concerning other appearance-altering paediatric conditions (e.g., skin & craniofacial conditions), there is limited evidence concerning factors or processes which may exacerbate or ameliorate appearance (dis)satisfaction among these paediatric populations (Gee et al., 2020). Given the importance of body image development in adolescence, appearance satisfaction is recognised as a key outcome in AIS care (Negrini et al., 2006) and future research could seek to identify factors which may buffer against negative body image in AIS.

While participants acknowledged a changing appearance, their condition could be kept private, and some were reluctant to explain their AIS to others. Explaining health conditions to peers has previously been identified as a challenge for adolescents receiving treatment within rheumatology, cardiology, and cystic fibrosis services (Secor-Turner et al., 2011; Kaushansky et al., 2017). As scoliosis can be concealed or of limited visibility, the decision to disclose is often left to the adolescent who may weigh up the threat to their desire for peer conformity and possible benefits such as social support from peers. Addressing adolescents' health-related communication needs through educational strategies is therefore worth

exploring in the context of AIS. Group-based psychoeducational programmes aimed at promoting psychosocial wellbeing among adolescents with various chronic conditions have previously incorporated strategies to enhance communication (Last et al., 2007; Douma et al., 2019). The programmes developed by these authors included online and in-person learning activities focused on how and what adolescents may want to tell others about their health condition and taking initiative in informing others (e.g., at school, in peer groups) about issues related to their condition (e.g., reasons for absences, limitations). Such strategies may be worthwhile to consider in development of supports for those with AIS.

The emotional burden of living with a serious health condition during adolescence is well recognised (Sawyer et al., 2007) and this was evident among participants as they came to terms with their AIS diagnosis and juggled fear and optimism in advance of surgery. Encouragingly, adolescents were able to gain emotional and practical support through communicating with others affected by AIS. They valued being able to share their experiences and were reassured by seeing others who had recovered from surgery. These findings build on those of MacCulloch et al. (2009) who reported that adolescents requested that their online scoliosis resource contain an interactive component to share personal stories. A peer support strategy may be of benefit to adolescents at the stage of surgical consideration and future research may seek to explore this further. Peer support for health conditions is understood to benefit participants by providing opportunity to learn coping techniques, acknowledge shared experiences, access encouragement and guidance, and reduce feelings of isolation (Olsson et al., 2005). Importantly, previous exploration of orthopaedic healthcare providers' recommendations for peer support highlight that such strategies should be professionally moderated (MacCulloch et al., 2010).

Surgical concerns voiced by the adolescents reiterate those reported in previous research related to surgery for AIS, including pain, surgical complications, and postsurgical limitations during the recovery period (Chan et al., 2017). However, the results of this qualitative investigation go beyond patients' concerns to shed light on their positive expectations of surgery. Adolescents looked forward to regaining normality in their lives, returning to sports they enjoyed, and anticipated improvements in the appearance of their scoliosis postoperatively. Focusing on positive expectations could be interpreted as a way of coping in

advance of surgery for some of the adolescents. Notably, a previous study reported that vigilant coping prior to surgery for AIS (defined as seeking information about surgery and recovery, acknowledging complications, and focusing on surgery benefits) was associated with higher levels of return to participation in recreational and social activities among adolescents in the 9 months after surgery (LaMontagne et al., 2004). Remaining cognisant of positive outcomes associated with AIS surgery may therefore be beneficial as part of preoperative preparation.

Although the optimism demonstrated by participants enabled them to view their upcoming surgery in a more positive light, it is also important to also consider management of realistic postoperative expectations. In particular, education about postsurgical appearance expectations is important for future satisfaction with surgical outcomes, as previous research has shown that higher presurgical expectations about postoperative appearance were associated with lower satisfaction two years after surgery (Sieberg et al., 2018). Spinal fusion surgery for AIS aims to prevent future progression and correct the existing curvature and while modern surgical techniques typically achieve curve correction of more than 60-70%, complete correction cannot be guaranteed (Imrie et al., 2011; Beauchamp et al., 2019).

Limitations and Future Directions

As is typically the case with qualitative approaches, the study sample was limited to a relatively small cohort of patients. Recommendations regarding clinical implications can be considered as tentative suggestions based on the experience of this sample, which require further research and evaluation of their suitability or efficacy. The homogeneity of our sample means that findings are most relevant to adolescents with moderate to severe AIS at the stage of surgical consideration or presurgical preparation, rather than all patients with AIS at various stages of treatment. Furthermore, participants all identified as White Irish and were predominantly female. The gender breakdown is consistent with the higher incidence of AIS among females (Konieczny et al., 2013), however lack of ethnic or racial diversity within the sample may fail to capture experiences reflective of the broader population of patients with AIS. Further research exploring experiences of those with AIS at different stages of treatment and among diverse samples is warranted. Another factor to consider in the interpretation of findings is that anticipated time to surgery may have impacted some participant perspectives

surrounding their treatment. However, it was not possible to estimate surgical wait times, nor did we collect information on expected waiting times among this sample.

Building on the findings of this research, additional studies are required to investigate the needs, goals, and the support preferences of presurgical adolescents with AIS, so that appropriate psychosocial interventions and supports can be developed. For example, preferred formats for strategies such as peer support could be considered (e.g., web-based, in-person). Future investigations should also seek to identify specific factors which may protect or buffer against negative psychosocial outcomes in AIS such as low body image so that supports or interventions can be tailored accordingly. Additionally, given the role of parents in their child's AIS care, exploration of parental experiences throughout the presurgical period represents another avenue for future research to inform patient and family-centered care in this area.

Conclusion

This study has provided valuable insight into the experiences of adolescents with idiopathic scoliosis, developing an understanding of the ways in which adolescents negotiate psychological and social aspects of their condition throughout the presurgical stage of treatment. Findings have led to the consideration of strategies which may benefit the psychosocial wellbeing and functioning of presurgical patients with AIS. Strategies include guidance and promotion of suitable activity participation at various stages of treatment, addressing health-related communication needs, the development of and providing access to peer support networks, and preoperative assessment and counselling focused on positive postsurgical expectation management to promote effective coping and surgical outcomes. These practical implications can be applied both clinically and by those working with adolescents with AIS in community support settings. As conservative and operative treatment for AIS continue to advance in technique and efficacy, a concurrent focus on addressing psychosocial wellbeing and support needs is key in optimizing care and promoting best outcomes for this patient group.

Chapter 4

The SCOLI Survey: An Investigation of Clinical Characteristics, Psychosocial Processes, Health-Related Quality of Life, and Body Image Among Adolescents with Idiopathic Scoliosis

Chapter Overview

This chapter details the development and conduct of the SCOLI survey, a cross-sectional, web-based quantitative survey which investigated clinical characteristics, psychosocial variables, health-related quality of life (HRQOL), and body image among a sample of 115 young people aged 12 – 21 years living with non-surgically corrected AIS. In particular, this study sought to identify psychosocial variables contributing to variation in key outcomes HRQOL and body image among those living with AIS and further to examine potential support needs. Firstly, the rationale and study framework are presented, followed by an account of the selection of variables for inclusion in the survey. After finalising the survey variables, existing standardised measures that mapped onto these variables were chosen and a small number of additional survey items were created. As this study was conducted during the COVID-19 pandemic, the implications of this unique research context are considered. This is followed by a description of the method including participant recruitment, detail of the survey content, and the study procedure. The results are then presented including a descriptive profile of the sample, results of a multiple hierarchical regression analysis, and further insights into support needs and COVID-19 findings. A follow-on detailed analysis of the coping strategy data through clustering techniques is subsequently presented in Chapter 5.

4.1. Introduction

In the previous chapter, an in-depth exploration into the experiences of adolescents living with AIS was provided, specifically those who were at the stage of surgical consideration. While some psychosocial concerns may be heightened among those with larger spinal curves reaching the surgical threshold, previous research demonstrates that associations between condition severity and HRQOL are small to moderate at best (Parent et al., 2010), meaning those with smaller curves are also vulnerable to difficulties. Indeed, previous research has

mainly focused on how clinical characteristics such as curve severity are associated with psychosocial wellbeing among adolescents with AIS and there is a need to identify other variables that contribute to the adjustment of this patient group (Berliner et al., 2013; Asher et al., 2004). Identifying contributory factors and processes would provide a more informed understanding of the impact that AIS can have on adolescents, in terms of why some adolescents may experience better or worse psychosocial outcomes. Taking a broadly biopsychosocial approach, this study sought to investigate whether psychosocial variables contributed to variation in key outcomes for AIS; health-related quality of life and body image (Asher et al., 2006; Auerbach et al., 2014). In line with this, a holistic framework was developed for this study to guide the examination of psychosocial variables among a sample of adolescents at various stages of treatment for non-surgically corrected AIS. A detailed account of the development of this study follows.

4.1.1. Study Framework

The design of the SCOLI survey was guided by the Appearance Research Collaboration (ARC) framework of adjustment to appearance-altering conditions (Rumsey et al., 2008). The ARC framework was established to inform research investigating psychosocial factors or processes that may contribute to variation in adjustment among populations with physical health conditions affecting appearance. The framework includes three main facets, which are "Predisposing Factors," "Intervening Processes," and "Outcomes." Predisposing factors typically include the clinical characteristics of the condition of interest, such as severity or general demographic characteristics, which can have an influence on outcomes. Intervening processes include psychosocial or cognitive variables such as cognitive representations of the self or coping which can have the potential to positively or negatively affect outcomes. These processes are considered potentially amenable to change. The third facet consists of outcomes which are intended to reflect the effects of the health condition in the individual (e.g. psychological wellbeing) (Rumsey et al., 2008). Figure 4.1 presents a working framework for this study based on the ARC framework. Through a well-considered process of research and refinement which is detailed in the subsequent survey development section, this initial framework was elaborated on to create the final SCOLI survey framework later presented in Figure 4.2.

Figure 4.1

Working Framework Guiding Design of the SCOLI Survey

(i) Predisposing Factors	(ii) Intervening Processes	(iii) Outcomes
Demographic & Clinical Variables	Psychosocial/ Cognitive Variables	Psychosocial Wellbeing
Demographics	Perception of Appearance	Quality of Life
Scoliosis Details	Appearance Investment	Body Image
Treatment Status	Coping Strategies	Mood & Anxiety

The impact of AIS on appearance is consistently recognised as a core aspect of the condition (Gallant et al., 2018; Weinstein, 2019) which prompted the use of this framework in the present study. This framework was also chosen to guide the selection of study variables as it aligned with the study aim to identify factors contributing to variation in key AIS outcomes health-related quality of life and body image. Moreover, it was designed to facilitate the identification of such factors or processes which are potentially amenable to change through psychosocial support or intervention. A further advantage of this framework was that it allows a focus on protective factors contributing to positive adjustment rather than focusing on difficulties alone (Rumsey et al., 2008; Clarke et al., 2014). While the ARC framework was created as part of a programme of research with participants over the age of 18 years, authors advised that the framework was also intended to inform future research including children and adolescents when variables relevant to the population of study and age-appropriate measures are utilised. Furthermore, recent qualitative research with health professionals working with young people who have chronic health conditions has identified the ARC framework as a suitable framework to guide investigation of the psychosocial adjustment of paediatric populations (Gee et al., 2020). Employing an adapted version of this framework for the AIS population represents a novel contribution to the literature.

4.1.2. Survey Development

Using the adapted framework presented in Figure 4.1 as a basis for survey development, previous literature informed the selection of variables to be included in the SCOLI survey. Findings from the qualitative interview study detailed in Chapter 3 also helped to inform survey development in some instances. Below, the inclusion of specific survey content is detailed under three headings corresponding with the aforementioned framework. Furthermore, consultations with relevant experts and stakeholders were conducted to ensure relevance and applicability of the survey (see section 4.1.2.1), and the implications of COVID-19 for the survey were considered (see section 4.1.2.2). Through this process of survey development, the working framework for the study was expanded and finalised. Please see Figure 4.2 for the final SCOLI survey framework, depicting the variables included in the survey. The selection of associated measures are detailed under Materials in the following section of this chapter (4.2.2).

(i) Predisposing Factors*:* In the context of AIS, predisposing factors of interest included the Cobb angle of the major scoliotic curve. This is the most common indicator of scoliosis severity (i.e. mild 10° – 24°; moderate 25° – 44°; severe 45°+) (Altaf et al., 2013), and more severe curves are associated with decreased health-related quality of life (Berliner et al., 2013). Standard demographic details were collected, including age which has been shown to potentially impact HRQOL outcomes (Daubs et al., 2014; Diarbakerli et al., 2019). Treatment details was another factor to be included. In particular, previous research indicates that bracing treatment may be associated with poorer quality of life, most notably in self-image and mental health domains (Wang et al., 2021). To profile the sample, additional detail regarding treatments accessed (e.g. physiotherapy) was of interest. Furthermore, AIS can impact functional ability or activity participation, and as identified in the qualitative study, adolescents may discontinue sporting activities. Therefore, it was decided to collect information on activity participation within the larger survey sample.

(ii) Intervening Processes: Coping strategies were included as a psychosocial process of interest for this study, which were hypothesized to contribute to variation in participants HRQOL. In the context of paediatric health conditions, coping can be described as the way the young person manages the internal and external demands of their condition, and it is

understood that these coping efforts can influence their HRQOL (Compas et al., 2012). However, there has been limited attention paid to coping strategies in the context of AIS (LaMontagne et al., 2004; Beka et al., 2006). Specifically, the ways in which adolescents with non-surgically corrected AIS cope with the stressors of AIS in their daily lives is yet to be investigated. A more detailed discussion of coping is provided in the following Chapter 5.

Next, given the impact of AIS on appearance and body image concerns in this population, three appearance-specific variables were included in order to investigate their potential contribution to body image outcomes. While curve size is considered a key indicator of condition severity, subjective perception of appearance-altering conditions may be a better indicator of psychosocial distress (Moss, 2005). To assess adolescents' subjective perception of their scoliosis appearance, pictorial scales were included in the survey in line with previous recommendations (Matamalas et al., 2014). The second appearance-specific variable of interest was appearance investment (i.e. the importance of appearance to ones' self-concept), identified as a key dimension of body image (Kling et al., 2019; Crerand et al., 2017). Greater investment in appearance is suggested to increase the likelihood of body image disturbance and is valuable to consider in the context of appearance-altering conditions, as those with lower investment may be less concerned about their condition (Crerand et al., 2017). Third, perceived visibility is another variable considered in the ARC framework, the inclusion of which in this study was further supported by the findings of the previous qualitative study (Chapter 3) surrounding the sometimes hidden nature of AIS and uncertainty regarding visibility. To our knowledge, neither appearance investment nor perceived visibility have previously been examined in relation to AIS.

(iii) Outcomes: Previous literature related to the AIS population was consulted to identify key outcome variables for assessment (Tones et al., 2006; Gallant et al., 2018; Bago et al., 2013; Matamalas et al., 2014). The most widely used outcome measure to capture psychosocial wellbeing of adolescents with AIS is the Scoliosis Research Society (SRS) measure of HRQOL and this was selected as a key outcome variable. The SRS measure (SRS-22r) is a comprehensive assessment of key domains of quality of life affected by scoliosis, namely function/activity, pain, self-image, and mental health (Asher et al., 2006). General appearance satisfaction was included as a variable of interest given that appearance satisfaction is

recognised as a key outcome in AIS care (Negrini et al., 2006). In addition, body image disturbance was selected as an outcome variable. Body image disturbance goes beyond a measure of (dis)satisfaction, and is described as the extent of appearance-related distress and its' impact upon psychosocial functioning (Cash et al., 2004). Research indicates that psychosocial functioning can be hindered by AIS-specific appearance concerns (Auerbach et al., 2014), and therefore this outcome was important for inclusion.

Figure 4.2

Final Framework for the SCOLI Survey Including Key Variables of Interest and the Associated Measures Selected for Inclusion in the Survey

(i) Predisposing Factors	(ii) Intervening Processes	(iii) Outcomes	
Demographic & Clinical Variables	Psychosocial/ Cognitive Variables	Psychosocial Wellbeing	+ (iv) Support Needs & Preferences
Demographics • Age, gender, etc. Scoliosis Details • Curve severity • Length diagnosis Treatment Status • Orthotic bracing	Coping Strategies • **(CSI & CODI)** Perception of Appearance • Perception of scoliosis appearance **(TAPS)** • Perceived visibility of scoliosis Appearance Investment • **(MBSRQ-AO)**	Quality of Life • Health related quality of life **(SRS-22r):** Function Pain Self-Image Mental Health Body Image • Scoliosis-related body image disturbance **(BIDQ-S)** • Appearance satisfaction **(BESAA-A)**	
+ (v) Context: Impact of COVID-19 pandemic **(CASPE)**			

(iv) Support Needs & Preferences: As a secondary objective to this study, and to further inform support of the AIS patient group, an additional element "support needs and preferences" was added to the study framework. While previous authors have suggested that psychosocial supports may be helpful in ameliorating negative effects of AIS on patients (Tones et al., 2006), there is a dearth of research on this topic meaning little is known about specific types of supports that would be beneficial or welcomed by the AIS patient group. One previous study had developed guidance for online information needs of patients with AIS who were undergoing surgery (MacCulloch et al., 2009), however, more research is required to expand on the wider psychosocial support of this group. The development of the items for

this section was informed by qualitative data from Chapter 3. In particular, qualitative insights pointed to the importance of information sources (for scoliosis education/support), the potential of peer support strategies, value of sport/exercise for wellbeing, and possible health-related communication needs. In consultation with members of the spinal team, these insights were translated into questions assessing adolescents' views on support strategies that had potential for future development.

4.1.2.1. Consultations on Survey Development

Members of the spinal team at the orthopaedic department of Children's Health Ireland (CHI) Crumlin, the national centre for paediatric scoliosis care, were consulted on the development of the SCOLI survey. The purpose of this consultation was to obtain feedback from the spinal team on the applicability and usefulness of the survey content, considering their experience with the AIS population and expertise in scoliosis care. Attendees included spinal nurse specialists, a clinical psychologist, physiotherapist, and consultant orthopaedic surgeon. Discussion of the survey content indicated agreement on selection of survey variables and potential clinical utility of the survey findings. Detailed guidance was received to inform the items collecting clinical information such as how to assess curve size (i.e. with an open-ended item). Items assessing treatment and physiotherapy access were also developed in consultation with the team to ensure collection of clinically relevant data. Feedback was also obtained to aid creation of the support needs items. A brief consultation was also held with a professor from the Centre for Appearance Research, Bristol, UK, with expertise in conducting research with young people with appearance-altering health conditions. The aim of this consultation was to discuss suitability of appearance-related variables and confirm suitability of the relevant measures selected for the survey.

Public & Patient Involvement (PPI). PPI describes patients contributing to the research process as advisors and sometimes as co-researchers (Bate et al., 2016). In line with guidance on PPI in health-related research, an adolescent with AIS was consulted on the suitability of the SCOLI survey (Bate et al., 2016; Coupe & Mathieson, 2020). Invited to partake in this process through one of the support group recruitment channels, the adolescent and their parent received the information materials and the adolescent completed the survey before providing feedback. The PhD researcher discussed via phone the purpose of the study,

and the type of feedback that would inform the research (i.e. relevance and comprehensibility of the survey content & information materials, whether anything was missing from the survey, and overall experience). The adolescent was provided with a shopping voucher to acknowledge their contribution. Feedback was positive indicating no concerns regarding ease of survey completion and clarity of materials. The survey content was relevant and interesting to complete. Based on feedback, minor clarifications were added to a small number of survey items. This consultation can be considered a lower level of PPI, situated within multiple levels of engagement where the highest level could include co-leadership and production of the research project (Irish Health Research Forum, 2015; Bates, 2021). The level adopted was suitable for this doctoral level study to fulfil the aim of determining suitability and comprehensiveness of study materials (Coupe & Mathieson, 2020).

4.1.2.2. Implications of COVID-19 Pandemic for the SCOLI Survey

The SCOLI survey was conducted during the COVID-19 pandemic, and in line with guidance for ongoing research studies with paediatric populations there were implications to consider (Stiles-Shields et al., 2020). COVID-19 was declared a pandemic by the World Health Organization on 11th March 2020 and changes to daily life were evident from this time including lockdowns, school closures, and implementation of social distancing measures. Please see Appendix D for an outline of the COVID-19 context and how it aligned with the survey timelines. These effects were present throughout data collection for the SCOLI survey which took place from September 2020 to February 2021. As outlined by Serlachius et al. (2020), young people with health conditions will have faced many challenges during COVID-19, and while many challenges are likely to be similar to those faced by all young people, some may relate to their health issues. Importantly for those with AIS, many elective procedures in paediatric orthopaedics such as spinal fusion surgery and outpatient appointments were postponed due to the increased demand on hospital systems and safety precautions during the pandemic (Peiro-Garcia et al., 2020). Two main aspects of the study which were affected by the COVID-19 pandemic were the survey content and participant recruitment.

COVID-19 & Survey Content. It was necessary to consider the potential impact of the pandemic on participants and how this may have influenced survey responses. COVID-specific survey items were therefore included to aid interpretation of the research findings by

capturing the research context (Stiles-Shields et al., 2020). The National Institutes of Health compilation of COVID-19 data collection instruments (Office of Behavioral and Social Sciences Research, 2020) was consulted and the COVID-19 Adolescent Symptom & Psychological Experience Questionnaire was identified (CASPE; Ladouceur 2020). To avoid over-burdening of participants, it was necessary to be selective in the number of additional items to be included in the survey. In line with this, nine COVID-19 related items were included (see Appendix E for copy of survey). Impact of the COVID-19 pandemic was therefore added into the study framework presented in Figure 4.2. Furthermore, mood and anxiety were not pursued as key outcome variables. Initially the inclusion of the Depression Anxiety Stress Scale (DASS; Loviboland & Loviboland, 1995) as a measure of negative emotional symptoms was proposed. However, the nature of this measure (i.e., general anxiety and mood symptoms over previous week period) was deemed particularly susceptible to influence by the COVID-19 context and in line with this, recent evidence has demonstrated that COVID-19 has been associated with increased depression and anxiety in adolescents (Nearchou et al., 2020).

COVID-19 & Participant Recruitment. It was initially anticipated that in-person recruitment through hospital clinics and support group meetings would be possible over the course of this study. However, factors such as prioritising safety of participants, adhering to social distancing, and the pausing of non-essential hospital activity meant that this recruitment approach could not be pursued. To proceed with the study, an online, remote recruitment strategy was considered most suitable in line with restrictions that precluded in-person interaction. Notably, AIS is a relatively rare condition and therefore the potential pool of participants for this study was small, with the pandemic representing an additional barrier to obtaining adequate sample size. To combat this, recruitment was extended beyond Ireland to include the UK, USA and Canada. These locations were chosen as treatment for AIS is similar in the healthcare systems of these countries, and for pragmatic reasons including that the survey was available in English only and that the researcher was able to establish links with recognised scoliosis organisations in these countries through collaborations in Ireland.

4.1.3. The Present Study

To summarize, this cross-sectional survey study adopted a comprehensive framework comprising clinical and demographic variables, psychosocial process variables, and key

patient-reported outcome variables health-related quality of life (HRQOL) and body image. Using this framework, we sought to investigate the contribution of coping to variation in HRQOL and the contribution of appearance-related cognitions to variation in body image outcomes among young people with AIS. In this way, the study sought to better understand the impact that AIS can have on affected adolescents, in terms of why some adolescents may demonstrate better adjustment (i.e. higher self-reported HRQOL and body image satisfaction). Complementary to this, we sought to examine support needs and preferences from the perspective of adolescents with AIS.

Aims and Objectives

This study aimed to identify variables contributing to health-related quality of life and body image outcomes among adolescents with AIS. Secondary to this, we sought to examine the support needs and preferences of this patient group. Specific objectives were:

(i) To examine the contribution of clinical and demographic characteristics to variation in health-related quality of life and body image outcomes in AIS.

(ii) To examine the contribution of coping strategies to variation in health-related quality of life in AIS (Regression 1).

(iii) To examine the contribution of appearance-related cognitions to variation in body image outcomes in AIS (Regression 2).

(iv) To assess the patient perspective on their support needs and preferences for potential support provision.

4.2. Method

A cross-sectional, web-based survey study was designed to investigate clinical characteristics, psychosocial processes, HRQOL, and body image outcomes among a sample of adolescents with idiopathic scoliosis.

4.2.1. Participants and Recruitment Procedures

4.2.1.1. Eligibility Criteria

Potential participants aged 12 – 21 years, diagnosed with AIS, with no previous scoliosis corrective surgery, were invited to take part. A participant age range of 12 – 21 was determined within the limits of the World Health Organisation definitions of adolescents and

young people (Sawyer et al., 2018), and in line with the age appropriateness of the survey measures as well as guidance from scoliosis clinicians. While both males and females were invited to take part, it was anticipated that the majority of the sample would be female due to the higher incidence of AIS among females (Konieczny et al., 2013). Participants were recruited from Ireland, the United Kingdom, Canada, and the United States of America.

4.2.1.2. Participant Recruitment

Recruitment occurred from September 2020 to February 2021 through an online, remote recruitment strategy in collaboration with scoliosis community and support networks. Connections were established by the researcher with Scoliosis Awareness & Support Ireland (Ireland), Straight Ahead (Ireland), Scoliosis Advocacy Network (Ireland), Straight2Swimming (S2S) (Ireland), Scoliosis Association UK (SAUK) (UK), and Curvy Girls Scoliosis (CGS) Support Network (Ireland/ UK/ USA/ Canada). Scoliosis Awareness and Support Ireland and the Scoliosis Advocacy Network are support groups consisting of parents and young people diagnosed with scoliosis. Straight Ahead is an orthopaedic charity aiming to improve scoliosis care, S2S is a swimming group for people with scoliosis, SAUK is a charity for people with scoliosis and families which provides information and advice, and CGS is an international scoliosis network founded in the US consisting of peer support groups. Leaders within each of these groups circulated details of the study and eligibility criteria through social media (i.e., private Facebook groups, Instagram groups, Twitter pages) or through email correspondence sharing the study web link, inviting participants on a voluntary opt-in basis.

Parents and potential participants with AIS were directed to follow the hyperlink included on the study invitation or to contact the PhD researcher using the details provided for further information. The hyperlink was supported by Qualtrics software (Qualtrics, Provo, UT) and led to a webpage including the study information sheet and sign up instructions. Sign-up and subsequent informed consent procedures varied depending on the age of the participant. For potential participants aged 12 to 17 years, a parent/guardian was required to read the study information sheet and complete the parental consent form. If the parent consented for their child to participate, they were prompted to enter an email address for the participant who would be completing the SCOLI survey. A secure survey link was then sent to the provided email address through the email distribution function on Qualtrics. Upon clicking the survey

link, the participant was required to read and complete an age-appropriate study information sheet and assent form before accessing the survey questions. Potential participants aged 18 to 21 years were required to read and complete the study information sheet and consent form before entering their own email address to receive the survey link. Survey links were sent within three working days of sign up with instructions. Most participants signed up to complete the survey online, however contact details were provided on the information sheet for requesting hard copy materials via post (participants in Ireland) and selected by five participants.

Over the course of recruitment, a total of 136 potential participants were signed up for the survey. A reminder email was sent after approximately one week and again after three weeks if the survey was incomplete. In total, 115 out of 136 completed the survey, with a response rate of 85% (111 online surveys, 4 posted surveys). Due to the remote nature of recruitment, reasons for non-completion were unknown. Within the Qualtrics system, it was evident that all survey emails were delivered to recipients, however it is possible that some emails may have reached junk folders as this is a recognised issue within the Qualtrics email distribution system. It could also have been the case that some parents or guardians may have signed up a young person who was subsequently not interested in taking part.

4.2.2. Materials

The survey totalled 142 items, which included a mixture of pre-existing standardised measures and items developed for the purpose of this study. Permissions were obtained for the use of measures where required. To aid coherence and ease of completion, the SCOLI survey was structured into four sections. These included (1) General & Scoliosis Information, (2) Appearance, (3) Health and Wellbeing, and (4) Coping and Support. An additional fifth section was included which related to the impact of the pandemic. For a copy of the survey, please refer to Appendix E.

4.2.2.1. Section 1: General & Scoliosis Information

Demographics and Characteristics of the Sample. Demographic data included age, gender, ethnicity, educational status, country of residence, living location (i.e. urban/rural). Cobb angle of the major curve, curve type, length of time since scoliosis diagnosis, types of

treatment accessed for scoliosis, health complications, and other diagnosed medical conditions were collected. Information on sports/ extracurricular activities and whether scoliosis had impacted these activities was collected.

Perceived Visibility of Scoliosis. Perceived visibility was measured by one item ("Do you think your scoliosis is visible to other people?") with responses ranging from 0 (Never visible) to 3 (Visible all of the time).

Trunk Appearance Perception Scale (TAPS). The TAPS is a pictorial scale that measures an individual's subjective perception of their scoliosis appearance (Bago et al., 2010) and is a valid and reliable instrument recommended for research with AIS populations from age 10 years (Thielsch et al., 2018). It includes three sets of figures depicting the body from three views (to the back, front, and bending), each at five levels of scoliosis severity for males and females. For each set of figures, participants selected the body they perceived to look the most like their own body. A mean scale score was calculated by summing the items and dividing by the number of items. Possible scores ranged from 1 (largest degree of asymmetry/ severity) to 5 (smallest asymmetry/ severity).

4.2.2.2. Section 2: Appearance

Body Esteem Scale for Adolescents and Adults – Appearance Subscale (BESAA-A). The BESAA appearance subscale includes 10 items which measure overall feelings about one's appearance (Mendelson et al., 2001). This measure has previously been used with clinical and non-clinical populations including young people with chronic health conditions (e.g. Barke et al., 2016). The BESAA demonstrates good psychometric properties (Kling et al., 2019), and has been demonstrated as reliable when used with adolescent populations (Dooley et al., 2019). Participants were required to rate each statement (e.g. "I'm satisfied with how I look," "I wish I looked like someone else") in terms of how often they agreed with it on a 5-point Likert scale ranging from 0 (Never) to 4 (Always). Mean scale scores were calculated for each participant with higher scores indicating greater appearance satisfaction.

Multidimensional Body-Self Relations Questionnaire - Appearance Orientation Subscale (MBSRQ-AO). The appearance orientation subscale of the MBSRQ measures the

extent of cognitive and behavioural investment in one's appearance (i.e. the importance of appearance to self-worth) (Cash, 2000). This 12-item measure is psychometrically sound and appropriate for use with adolescents above 12 years of age (Banasiak et al., 2001; Marco et al., 2017). Participants were required to read 12 statements relating to appearance investment (e.g. "Before going out, I usually spend a lot of time getting ready") and rate how much they agreed with each statement on a 5-point Likert scale ranging from 1 (Definitely disagree) to 5 (Definitely agree). The mean of the items was computed to form a scale score for each participant, with possible scores ranging from 1 – 5. Higher scores indicate higher appearance investment.

Body Image Disturbance Questionnaire – Scoliosis (BIDQ-S). The BIDQ-S is a 7-item, valid and reliable instrument adapted from the original BIDQ, that assesses scoliosis-specific appearance concerns and how these concerns impact upon psychosocial functioning (Auerbach et al., 2014). The BIDQ-S was developed specifically for use in AIS, is identified as a key outcome measure for this population (Gallant et al., 2018), and has been validated with adolescents from age 11 years (Auerbach et al., 2014; Wetterkamp et al., 2017). Participants answered each question (e.g. "Has your back shape caused problems with your friends, family members, or dating?") on a 5-point Likert scale from 1 (Never) to 5 (All the time), with higher scores indicative of greater body image disturbance. Total mean scores were derived by averaging the scores of the 7 primary questions. Items 2, 5, 6, and 7 included sub-questions inviting a free-text response.

4.2.2.3. Section 3: Health and Wellbeing

Scoliosis Research Society 22-Item Revised Measure (SRS-22r). The SRS is the most widely used, valid and reliable instrument to assess health-related quality of life in AIS (Asher et al, 2006). The 22-item revised measure (SRS-22r) is the most recent version and has been identified as the most metrically sound version for use with adolescents (Bago et al., 2013). The SRS-22r contains 20 items that assess four key dimensions of HRQOL in scoliosis, each with five items (Function, Pain, Self-image, and Mental Health), and two additional items which measure Satisfaction with Treatment. Participants responded to each item by scoring a 5-point Likert scale (e.g. "Does your back limit your ability to do things around the house?"), with answers from 1 (Very Often) to 5 (Never). Scoring followed general practice by

calculating a mean for each subscale and an overall HRQOL score (i.e., mean of the four HRQOL dimensions). Higher scores indicate greater quality of life.

Visual Analog Scales (VAS) for Anxiety & Mood. Visual Analog Scales (VAS) are single item measures typically used to measure subjective psychological states or levels of distress with a visual scale. VAS carry the advantages of being quick and straightforward to complete, and can be as efficient as more complex self-report scales (Kontou et al., 2012; Amend et al., 2014). Participants completed two VAS related to their feelings at the present time. One related to feelings of anxiety and the other related to low mood in relation to their scoliosis. Possible responses ranged from 0 (Not at all) to 10 (Very much), with higher scores indicating increasing anxiety or lower mood.

4.2.2.4. Section 4: Coping and Support

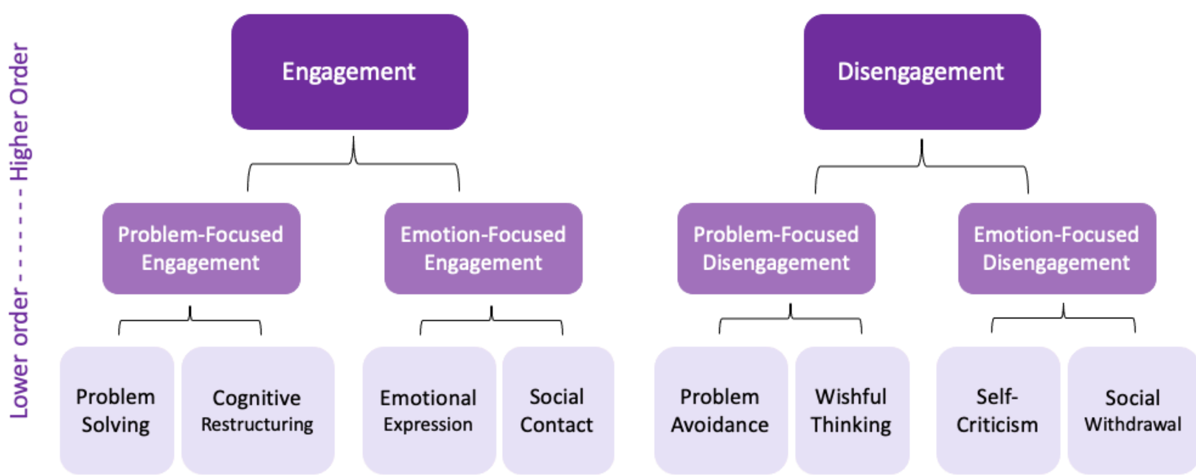
Coping Strategies Inventory – 32 Item Measure (CSI). The 32-item CSI (Tobin 1991) assesses eight different types of engagement or disengagement coping strategies that may be utilised in response to a specific stressor (in this case, scoliosis). Subscales include Problem Solving, Cognitive Restructuring, Emotional Expression, Social Contact, Problem Avoidance, Wishful Thinking, Self-Criticism, and Social Withdrawal. These 8 primary subscales can be grouped into four secondary subscales (Problem-Focused Engagement, Emotion-Focused Engagement, Problem-Focused Disengagement, and Emotion-Focused Disengagement), and further into two tertiary subscales (Engagement and Disengagement). See Figure 4.3 for a graphic representation of the CSI scale structure (Tobin et al., 1991). In the instructions for this measure, participants were asked to think about times when their scoliosis may cause them stress to prompt them to consider coping in relation to their condition. Participants rated each coping item (e.g. "I try to look on the bright side of things") by how often they used each strategy on a 5-point Likert scale ranging from 1 (Never) to 5 (Very often). Subscale scores were calculated as the mean for each subscale, with higher scores indicating greater use of the strategy.

Coping with Disease Questionnaire (CODI). The CODI was developed to assess a smaller number of coping strategies among young people with paediatric health conditions (Petersen et al., 2004). The 6-item Acceptance subscale (acceptance of condition) and the 4-

item Distance subscale (appraising condition as less serious) were included in the SCOLI survey on an exploratory basis. These subscales were chosen as they represented concepts not covered within the CSI and demonstrated good reliability (Oppenheimer et al., 2018). Participants were asked to read each statement (e.g. "I think my scoliosis is no big deal") and rate how often they thought in these ways on a 5-point Likert scale from 1 (Never) to 5 (Very often). The CODI subscales were not included in the regression model but were included in the cluster analysis described in Chapter 5 (see page 96, paragraph 2 for details).

Figure 4.3

Structure and Subscales of the Coping Strategies Inventory



Support Needs and Preferences. Five items collected details regarding participants' access to scoliosis information and satisfaction with information available. Two items assessed whether participants had previously met another young person with scoliosis and whether this experience had been positive or negative. Participants were then asked to rate how helpful seven types of supports or services would be to them and other young people with scoliosis (relating to increased awareness, communication skills, peer support, exercise group, professional guidance) on a 5-point Likert scale from 1 (Definitely Helpful) to 5 (Definitely not helpful).

4.2.2.5. COVID-19 Section: Pandemic Related Questions

COVID-19 Adolescent Symptom & Psychological Experience Questionnaire (CASPE). The CASPE was developed by Ladouceur (2020) to provide a timely research tool assessing

COVID-related experiences among adolescents. Four primary items from this measure were included in the survey which assessed the impacts of COVID-19 on participants' lives. Participants rated on a 5-point Likert scale ranging from 1 (Not at all) to 5 (A great deal) how much they thought their lives had been impacted both negatively and positively. For each response, participants were also asked to select up to three events or changes to daily life which were the most negative (e.g. "Having to stay at home," "Worried about someone who has or has had the virus") or positive (e.g. "Reduced amount of schoolwork," "More time to exercise or go outside").

COVID-19 and Treatment. Participants indicated whether their medical treatment had been impacted as a result of the pandemic and if so, they were prompted to enter free-text detail. This question was included in Section 1 of the survey alongside the other questions regarding medical treatment.

COVID-19 and Scoliosis Questions. Six items were developed which mapped broadly onto key variables that had been included in the survey. Participants were asked to indicate whether the following experiences were the same, better, or worse as a result of the COVID-19 pandemic: "The way you feel about your general appearance," "The way you feel about the appearance of your scoliosis," "The way you cope with your scoliosis," "Your level of pain," "Your ability to function/ do activities," and "Your happiness/ mental wellbeing."

4.2.3. Procedure

Prior ethical approval for this study was granted by the University College Dublin Humanities Research Ethics Committee (HS-20-Motyer-Fitzgerald). Data collection took place between September 2020 and February 2021. The survey was predominantly completed online (*n* = 111) supported by Qualtrics software (Qualtrics, Provo, UT) and on paper by fewer participants (*n* = 4), taking approximately 30 minutes to complete. Online, up until survey submission, participants could leave the survey session if desired and return to the point they had last completed. The survey was set up in a user friendly format with full instructions. For posted surveys, the survey content matched the online version. At the end of the survey, participants were presented with information about seeking help or advice if they were concerned about their scoliosis (e.g. from a parent, GP, member of their medical team) and links to official support networks and information pages relevant to scoliosis and wellbeing. After taking part in the survey, participants were eligible to receive an amazon voucher to the

value of 15€/£/$ by email or post depending on the survey format selected. Previous research indicates that it is appropriate to compensate older children and adolescents (>9 years of age) in recognition of their research participation. This age group are shown to understand the meaning and value of fair compensation for research participation, while also making a voluntary decision to participate (Bagley et al., 2007). Survey data were downloaded from Qualtrics and imported into SPSS. Data collected using hard copy surveys (n = 4) were manually input to the SPSS datafile. The dataset was checked for quality (e.g. absence of straight lining) and completeness of responses.

4.2.4. Analysis

All analyses were conducted using IBM SPSS (Statistical Package for the Social Sciences) Version 24. In a preliminary analysis of the dataset, descriptive statistics and chi-square analyses were conducted to profile the sample and examine potential differences between groups sampled from the different geographical locations (i.e., Ireland, UK, USA & Canada). Exploratory independent sample t-tests were also used to probe for possible gender differences. Descriptive statistics and frequencies were reported to provide a detailed profile of the sample and the data collected for key variables.

Using the framework for the SCOLI survey presented in Figure 4.2 (page 51) as a basis, two hierarchical linear regression models were conducted to assess the relationships between clinical and demographic variables, psychosocial variables, and key outcomes of health-related quality of life (HRQOL) and body image. For the regression analyses, in line with guidelines for required sample size ($N \geq 50 + 8m$, where m is the number of predictor variables; Tabachnick & Fidell, 2014), a sample of 114 was required to run a regression model with eight predictor variables. A-priori power analysis conducted using G*Power 3.1 (Faul et al., 2007) indicated that a sample of 109 was required for multiple regression with eight predictors for a moderate effect size of 0.15, alpha level of .05, and power of 0.8. Initially, Pearson product moment correlations (two-tailed) were conducted to examine relationships among intended regression predictor and outcome variables. For regression model 1 (predicting HRQOL), age, curve size, length of diagnosis, and brace wear were entered in block 1, and the four CSI secondary types of coping were entered in block 2. For regression model 2 (predicting body image disturbance and appearance satisfaction), age, curve size, length of

diagnosis, and brace wear were entered in block 1, and perceived visibility, subjective perception of appearance, and body image investment were entered into block 2. This approach allowed examination of the combined amount of variance contributed by the predictor variables as well as the relative contribution of the psychosocial variables when controlling for demographic and clinical characteristics.

4.3. Results

4.3.1. Description of the Dataset

4.3.1.1. Missing Data

The frequency of missing data was minimal, indicating a high level of engagement with the survey. For variables measured using standardised instruments (i.e. TAPS, BESAA-A, MBSRQ-AO, BIDQ-S, SRS-22r, CSI, CODI), the range of missing data was between 0 and 0.8%. For the remaining single item variables on the survey, the level of missingness ranged from 0 to 2.6% (three participants did not report their curve size). Based on the low level of missing values (i.e. <5%) which were missing at random due to a small number of incomplete surveys, listwise deletion was applied to handle missing data for statistical analysis (Bennett, 2001; Dong & Peng, 2013).

4.3.1.2. Overview of the Sample

Of the 115 participants who took part, 26 (22.6%) were from Ireland, 62 (53.9%) were from the United Kingdom, and 27 (23.5%) were from the United States & Canada. The United States and Canada were grouped as one geographical area due to low participant numbers from Canada (n = 3). Details of demographic and scoliosis characteristics for the overall sample and by participant location are presented in Table 4.1. To check for differences between groups recruited from each of the three geographical areas, chi-square tests were conducted for Gender, Education, Curve Severity, Curve Type, Length of Diagnosis, Brace wear, and Physiotherapy access by Geographic Area (results included in Table 4.1). After applying the Holm Sequential Bonferroni correction for multiple tests (Holm, 1979), the chi-square for education stage was significant. The groups were broadly similar on key characteristics and for the purpose of further analyses, were analysed as one sample group. Detailed characteristics of the sample are described in the following section 4.3.2.

Table 4.1

Demographic and Clinical Characteristics of the SCOLI Survey Sample (N = 115)

	Full Sample (N = 115)	Ireland (n = 26)	UK (n = 62)	US/CAN (n = 27)
Age				
Range	12 – 21	12 – 19	12 – 21	13 – 21
Mean (*SD*)	15.5 (2.50)	14.6 (2.10)	16.02 (2.59)	15 (2.40)
Gender,[a] $\chi^2(2, N = 113) = 6.27, p = .044^{\dagger}$				
Female	98 (85.2%)	20 (76.9%)	52 (83.9%)	26 (96.2%)
Male	15 (13%)	6 (23.1%)	9 (14.5%)	0
Other	2 (1.7%)	0	1 (1.6%)	1 (3.7%)
Ethnicity				
White	108 (93.9%)	26 (100%)	58 (93.5%)	24 (88.9%)
Asian	4 (3.5%)	0	2 (3.2%)	2 (7.4%)
Multiple	1 (.9%)	0	0	1 (3.7%)
Black	1 (.9%)	0	1 (1.6%)	0
Other	1 (.9%)	0	1 (1.6%)	0
Education Stage, $\chi^2(2, N = 107) = 10.54, p = .005$*				
School	81 (70.4%)	23 (88.4%)	36 (58.1%)	22 (81.5%)
College/ University	26 (22.6%)	2 (7.7%)	21 (33.9%)	3 (11.1%)
Other	8 (7%)	1 (3.8%)	5 (8.1%)	2 (7.4%)
Curve Size (degrees)				
Range	11 – 110°	11 – 90°	13 – 110°	12 – 56°
Mean (SD)	43° (20.5)	40.2° (21.7)	49° (21)	32.67° (12.51)
Curve Severity,[b] $\chi^2(4, N = 112) = 12.72, p = .013^{\dagger}$				
Mild (<25°)	19 (17%)	6 (23.1%)	4 (6.5%)	$9^{\dagger}$ (33.3%)
Moderate (25°-44°)	46 (41%)	10 (38.5%)	24 (38.7%)	12 (44.4%)
Severe (45°+)	47 (42%)	10 (38.5%)	31 (50%)	6 (22.2%)
Curve Type,[c] $\chi^2(4, N = 106) = 1.94, p = .746$				
Thoracic	28 (24.3%)	7 (26.9%)	17 (27.4%)	4 (14.8%)
Thoracolumbar	68 (59.1%)	15 (57.7%)	35 (56.5%)	18 (66.7%)
Lumbar	10 (8.7%)	3 (11.5%)	5 (8.1%)	2 (7.4%)
Length Diagnosis, $\chi^2(4, N = 115) = 8.43, p = .077$				
0 – 2 years	48 (41.7%)	13 (50%)	28 (45.2%)	7 (25.9%)
2 – 4 years	33 (28.7%)	10 (38.5%)	15 (24.2%)	8 (29.6%)
4 + years	34 (29.6%)	3 (11.5%)	19 (30.6%)	12 (44.4%)

(contd.)	Full Sample (N = 115)	Ireland (n = 26)	UK (n = 62)	US/CAN (n = 27)
Wearing Orthotic Brace, $\chi^2(2, N = 115) = 9.52, p = .009^{\dagger}$				
Yes	40 (34.8%)	8 (30.8%)	16 (25.8%)	$16^{\dagger}$ (59.3%)
No	75 (65.2%)	18 (69.2%)	46 (74.2%)	11 (40.7%)
Accessing Physiotherapy, $\chi^2(2, N = 115) = 1.11, p = .574$				
Yes	70 (60.9%)	17 (65.4%)	35 (56.5%)	18 (66.7%)
No	45 (39.1%)	9 (34.6%)	27 (43.5%)	9 (33.3%)

[a]Two participants selected other gender, including nonbinary and questioning. [b]Three participants did not report a curve size. [c]Nine participants were unsure of their curve type.
Note. For chi-square tests, to reduce cells with expected counts <5, the following categories were not included; Gender: (Other) and Education Stage: (Other).
*Holm's sequential Bonferroni procedure was applied to correct for multiple testing with alpha .05 (Holm, 1979). Significant Chi-Squares are indicated by an Asterix, standardised residuals ±2 also marked.
†As tests were exploratory, tests significant at $p < .05$ are marked for information purposes.

4.3.2. Profile of the Sample

4.3.2.1. Demographic and Clinical Characteristics

Demographic Information

As shown in Table 4.1, the majority of the sample identified as White (93.9%). In line with incidence rates of AIS, there was a higher proportion of females than males in the sample (n = 98; 85% female). The mean age of the sample was 15.5 years (Range 12 – 21; SD = 2.5), with 83% of the sample aged 18 years or younger. Most of the sample were in formal education, with 7 (6%) attending primary school, 74 (64%) attending secondary school, and 26 (23%) attending College or University. Eight participants (7%) specified another occupation including professional skills training or gap year between school and college. In terms of their living location, most participants identified their locality as a city/ suburb (n = 36, 31%) or a town (n = 42, 37%), while 20 (17%) lived in a village, 13 (11%) lived in the countryside, and 3 (3%) lived on a farm.

Gender Differences

Disproportionate numbers precluded formal gender comparisons and exploratory analyses were therefore conducted. A comparative sample of females was extracted from the dataset (n = 15), purposively matched to the male sample in terms of geographical location and age, and as close as possible for spinal curve magnitude and length of diagnosis. Independent

sample *t*-tests (two-tailed) were used to probe for potential differences between males and females on key variables (SRS-22r, BESAA, BIDQ-S, MBSRQ, Coping). While tests were conducted on exploratory basis, to minimise Type I error rate for multiple testing, the Holm's sequential Bonferroni procedure was applied (Holm, 1979). Findings indicated no statistically significant differences between males and females, however it was notable from the mean differences that males reported lower levels of appearance investment (M = 2.82) than females (M = 3.69). Results of *t*-tests are tabulated in Appendix F.

Clinical Characteristics

AIS Diagnosis. The average Cobb angle of participants' major scoliotic curve was self-reported as 43° (Range 11° - 110°, SD = 20.5). Most participants reported moderate curves ranging from 25° - 44° (41%) or severe curves exceeding 45° (42%). Fewer reported mild curvatures less that 25° (17%). Scoliotic curves exceeding 70° are considered of relatively higher severity due to increased rates of complications and surgical complexity (Tarrant et al., 2016). In this sample, 10 participants reported curvatures >70°. For discussion of the use of Cobb angle measurements, please see Appendix G. The most commonly reported curve type was thoracolumbar (59%), followed by thoracic (24%), and lumbar (9%) in line with the most common types (Konieczny et al., 2013). 41% of participants reported a length of diagnosis under 2 years, 29% from 2 – 4 years, and 30% were diagnosed for over 4 years. Participants were asked to rate how often their scoliosis was a "problem in their life." Only 5 participants (4%) indicated that their scoliosis was never a problem. 54% (n = 62) indicated their scoliosis was sometimes a problem, 30% (n = 34) reported it was a problem a lot of the time, and 12% (n = 14) indicated it was a problem all of the time.

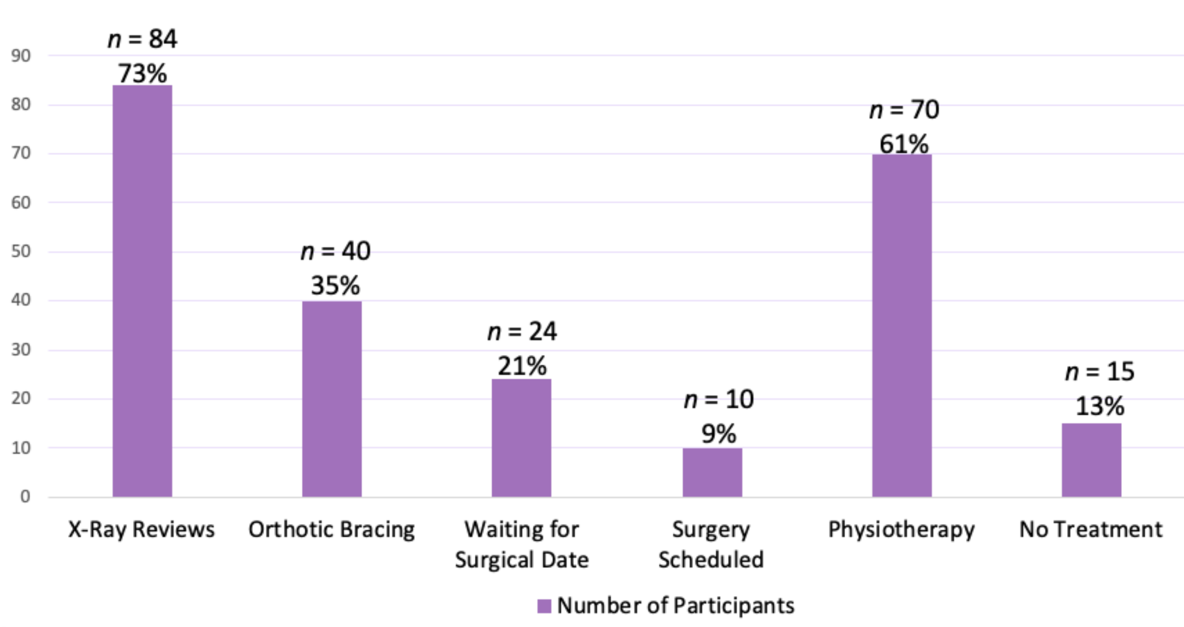
Health Complications. A third of participants (n = 39; 34%) indicated health complications related to their scoliosis, including shortness of breath/ difficulty breathing (n = 27), pain (n = 21) and headaches (n = 2). Pain reports were varied and included descriptions such as "back ache," "shoulder and rib pain," and "stabbing pain." Some participants also reported having another diagnosed medical condition (n = 31; 27%). The most common was asthma (n = 13), with other conditions reported by one or two including anaemia, bowel issues, depression, anxiety, endometriosis, hypermobility, cardiac issues, eosinophilic esophagitis, hyperthyroidism, gender dysphoria, and ADHD.

Treatment Details

The medical treatment that participants were receiving for their scoliosis at the time of survey completion is summarised in Figure 4.4.[1] The majority of participants were being routinely reviewed with radiographic imaging (*n* = 84; 73%). Just over one third of participants were being treated with an orthotic brace (n = 40; 35% of overall sample). Of those undergoing bracing, 60% (n = 24) also reported attending physiotherapy, while 14% (n = 6) also reported waiting for a surgery date or that surgery was scheduled. Among the 75 participants who were not undergoing bracing (65% of overall sample), 61% (n = 46) attended physiotherapy and 37% (n = 28) reported waiting for a surgery date or that surgery was scheduled. Looking to the specific types of physiotherapy that were accessed by the overall sample, 30 (26%) had attended physiotherapy at a local hospital or practice, 31 (27%) had accessed scoliosis-specific physiotherapy (e.g. Schroth) and 25 (22%) participants reported doing a daily exercise programme for their scoliosis.

Figure 4.4

Treatment Details Self-Reported by Participants



Note. Percentages do not equate to a total of 100 as participants selected as many that applied.

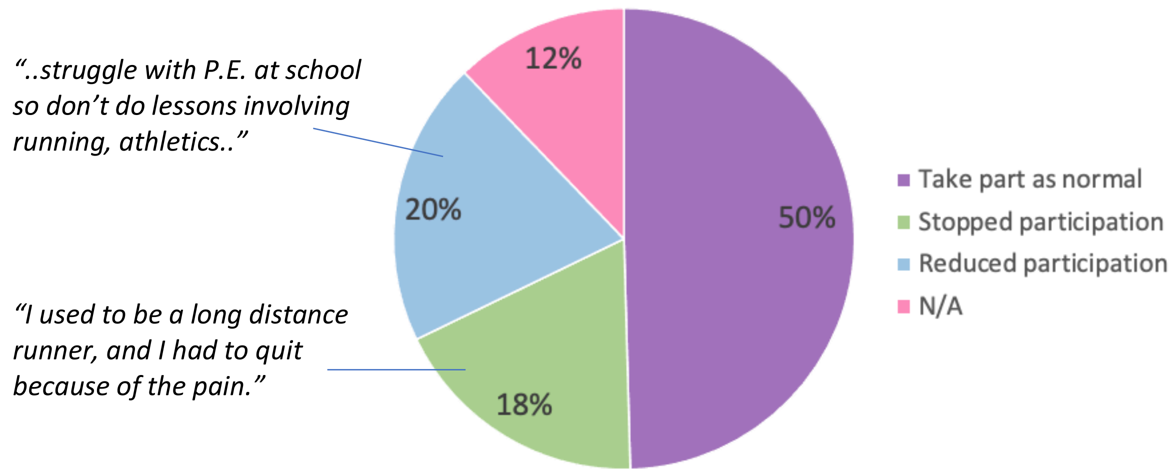
[1] While numerous descriptors were collected on the sample, not all were included in further statistical analyses. Permission was obtained to retain the dataset for future analysis. Descriptive statistics are presented to provide a detailed profile of the sample.

Activity Participation

82 participants (72%) indicated that they normally take part in sport or extra-curricular activities (prior to COVID restrictions), while the remainder indicated that they did not. Participants listed a broad range of 36 different activities with the most common being dance (*n* = 26), swimming (*n* = 19), football (*n* = 11), going to the gym (*n* = 8), running (*n* = 8), basketball (*n* = 8), gymnastics (*n* = 7), and yoga or Pilates (*n* = 7). Of these participants, 80% spent 7 hours or less per week on their activities (*M* = 3.6, *SD* = 1.8), and the remaining 20% reported spending over 7 hours per week. The effect of AIS on activity participation is shown in Figure 4.5. The 44 participants who reporting stopping or reducing activity participation had significantly larger curvatures (*M* = 49°, *SD* = 21) than the 57 participants who reported activity as normal (*M* = 37°, *SD* = 17), *t*(96) = 2.868, p = .005.

Figure 4.5

Participant Responses Regarding Activity Participation



4.3.2.2. Psychosocial Processes

Descriptive statistics and reliabilities for coping strategies (CSI and CODI), trunk appearance perception (TAPS), and appearance investment (MBSRQ-AO) are shown in Table 4.2. Cronbach's alpha values revealed acceptable to excellent reliability (0.64 – 0.90). Results for perceived visibility of scoliosis, measured with a one item indicator, are illustrated in Figure 4.6. The majority of participants reported that they believed their scoliosis to be visible "sometimes" (*n* = 88, 76%), while 15% of participants felt that their scoliosis was visible

"most" or "all" of the time (*n* = 17). Only 9% of participants felt their scoliosis was "never visible" (*n* = 10).

Table 4.2

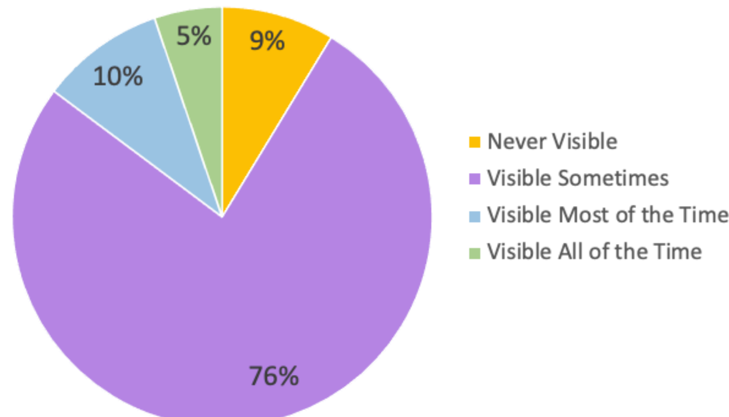
Psychometric Properties for Study Variables Assessed by Standardized Measures

Scale	*M*	*SD*	Range	Cronbach's α
TAPS	3.18	0.89	1 – 5	.84
MBSRQ-AO	3.42	0.80	1 – 4.8	.90
BIDQ-S	2.20	0.77	1 – 4.6	.87
CSI				
PF[a] Engagement	3.25	0.73	1 – 5	.85
EF[b] Engagement	3.26	0.80	1.5 – 5	.90
PF Disengagement	3.06	0.62	1.6 – 4.4	.64
EF Disengagement	2.35	0.88	1 – 4.9	.90
CODI				
Acceptance	3.76	0.83	1 – 5	.86
Distance	2.29	0.99	1 – 5	.85

Note: For the all measures, data presented are based on participants' mean scale scores as per scoring instructions. For the CSI, data for the four secondary subscales are presented.
[a]PF = Problem Focused, [b]EF = Emotion Focused.

Figure 4.6

Participant Responses on Visibility of Their Scoliosis to Others



4.3.2.3. Outcomes

Descriptive statistics and reliabilities for HRQOL (SRS-22r), appearance satisfaction (BESAA-A), and body image disturbance (BIDQ-S) are shown in Table 4.3. Good to excellent reliability was observed for each of the measures (α = 0.80 = 0.93). For the two single-item visual analogue scales, mean score for anxiety was 4.59 (Range 0 – 10, *SD* = 2.88) and mean score for low mood was 4.05 (Range 0 – 10, *SD* = 2.92). Both the anxiety and low mood VAS correlated with the mental health subscale of the SRS-22r ($r = -.57$ for both, $p < .001$). Further detail on data collected with the BIDQ-S measure is presented below.

Table 4.3

Psychometric Properties for Outcome Variables Assessed by Standardized Measures

Scale	*M*	*SD*	Range	Cronbach's α
BESAA-A	2.21	0.90	0 – 3.9	.93
BIDQ-S	2.20	0.77	1 – 4.6	.87
SRS-22r Total	3.58	0.68	1.6 – 4.9	.93
Function/Activity	4.12	0.74	1.6 – 5	.80
Pain	3.54	0.90	1 – 5	.85
Self-Image	3.35	0.81	1 – 5	.81
Mental Health	3.23	0.86	1.2 – 4.8	.89
Satisfaction	3.80	1.00	1 – 5	.87

Note: For the all measures, data presented are based on participants' mean scale scores as per scoring instructions. For BESAA-A, higher scores indicate higher satisfaction with appearance. For BIDQ-S, higher scores indicate higher levels of body image disturbance. For the SRS-22r, higher scores indicate better outcomes in each HRQOL domain.

Body Image Disturbance (BIDQ-S). 13% (*n* = 15) of participants were "not at all'" worried about the appearance of their back. 37% (*n* = 43) were somewhat worried, 31% (*n* = 36) were moderately worried, and 19% (*n* = 21) were very or extremely worried. Specific concerns about appearance most commonly selected were uneven shoulders (*n* = 70), followed by shoulder blades sticking out (*n* = 69), a rib bump (*n* = 61), hip asymmetry (*n* = 54), and lastly frontal chest asymmetry (*n* = 37). Free-text response data from the BIDQS provided detail on how participants body image concerns impacted upon their psychosocial functioning. This data is summarised in Table 4.4.

Table 4.4

BIDQS Sample Free-Text Responses

Item	Sample Free-Text Detail
2(b). Effect of back shape on life	*"I feel I can't wear things I would like to wear and I feel constantly worried about being judged." (female, age 14). "I have to always wear jumpers because otherwise you can see it through my t-shirts." (male, age 17).*
5(b). Back shape caused issues with friends, family members, dating	*"Some people tease me." (female, age 14). "It concerns me when it comes to dating, if the person would mind that I have scoliosis." (female, age 21).*
6(b). Back shape caused issues with school, job, or doing things important to you	*"Not being able to sit and write for extended periods." (female, age 15). "I'm cautious of what I do, I avoid things that I know will make my back hurt." (female, age 18). "I'm not able to row with the lads anymore." (male, age 15).*
7(b). Avoidance of activities due to back shape	*"Swimming, so I don't have to show my back." (female, age 18); "Would never post a picture on social media that would show my back shape." (female, age 19).*

4.3.3. Hierarchical Regression Analyses

Hierarchical linear regression models were used to investigate the relative contribution of psychosocial variables in explaining key outcomes among adolescents with AIS (i.e. health-related quality of life and body image). Initial correlational analyses demonstrated significant relationships between the independent and dependent variables chosen for inclusion in the regression models. Demographic and clinical characteristics including age, curve size, length of diagnosis, and brace wear were controlled for in block 1 and relevant psychosocial variables were entered into block 2 of the regression models. Variables included in the regression models are shown in Table 4.5. In preparation for the regression analyses, data were screened and preliminary analyses were conducted to ensure no violation of the required assumptions including normality, linearity, multicollinearity, and homoscedasticity. A square root transformation was applied to the Perceived Visibility variable to obtain a nearer-normal distribution. All tolerance and variance inflation factor values fell within an acceptable range (tolerance range .424 to .935; VIF range 1.07 to 2.36). For a detailed account of data screening and the steps taken, see Appendix H.

Table 4.5

SCOLI Survey Multiple Hierarchical Regression Analyses

Independent Variables Step 1	Independent Variables Step 2	Dependent Variables
Regression Model 1		
Age Curve Size Length Diagnosis Brace Wear	Problem Focused Engagement (CSI) Emotion Focused Engagement (CSI) Problem Focused Disengagement (CSI) Emotion Focused Disengagement (CSI)	Health-Related Quality of Life (SRS-22r)
Regression Model 2		
Age Curve Size Length Diagnosis Brace Wear	Trunk Appearance Perception (TAPS) Perceived Visibility Appearance Investment (MBSRQ-AO)	Body Image Disturbance (BIDQ-S) Appearance Satisfaction (BESAA-A)

Note. Gender, geographical location, and physiotherapy (yes/no) were also tested as predictor variables for block 1 however were not included in the final regression models due to non-significant associations with the dependent variables via initial correlation analyses/ omission did not alter results.

4.3.3.1. Regression Model 1: Health Related Quality of Life

Correlations

Correlations among the predictor (demographic and clinical variables, coping strategies) and dependent variable (HRQOL) to be included in the regression model are presented in Table 4.6. To reduce Type 1 error rate when performing multiple tests, the criterion for statistical significance was reduced from .05 to .01 and therefore only correlations with $p < .01$ will be reported on. In terms of relationships between demographic/ clinical variables and coping strategies, larger curve size was associated with lower use of problem-focused engagement (r = -.278). A longer length of diagnosis was also associated with higher usage of emotion-focused disengagement (r = .272), while wearing a brace was associated with greater use of problem-focused engagement (r = -.262). These correlations were small to moderate in nature. In terms of HRQOL, correlation analyses demonstrated that increased age, curve size, and length of diagnosis were associated with decreased HRQOL (r = -.270, -.368, -.284, respectively), while wearing a brace was associated with increased HRQOL (r = -.331), with moderate effects.

Table 4.6

Pearson Correlations Among Demographic & Clinical Variables, Coping Strategies, and HRQOL

Measure	1	2	3	4	5	6	7	8	9
1. Age	-								
2. Curve Size	-.048	-							
3. Length Diagnosis	.538***	-.024	-						
4. Brace (Yes/No)[a]	.380***	.206*	.031	-					
5. CSI PF[b] Engagement	-.046	-.278**	-.008	-.262**	-				
6. CSI EF[c] Engagement	.048	-.155	.022	-.075	.363***	-			
7. CSI PF Disengagement	.038	.242*	.019	-.007	-.243**	-.195*	-		
8. CSI EF Disengagement	.199*	.080	.272**	.213*	-.343***	-.365***	.369***	-	
9. SRS Quality of Life	-.270**	-.368***	-.284**	-.331**	.490***	.287**	-.446***	-.616***	-

[a]Point biserial correlation reported for dichotomous variable. [b]PF = Problem Focused, [c]EF = Emotion Focused. *** $p < .001$, ** $p < .01$; * $p < .05$ (two-tailed). *Note.* Cohen's Pearson *r* effect size guidelines: small = 0.10, medium = 0.30, large = 0.50.

Examining coping, the two engagement domains, problem and emotion-focused were positively correlated (r = .363). Similarly, the two disengagement domains, problem and emotion-focused were positively correlated (r = .369). Problem-focused engagement was also negatively correlated with the two disengagement domains (r = -.243, -.343), while emotion-focused engagement was negatively correlated with emotion-focused disengagement (r = -.365). Again, these associations between coping domains were considered small to moderate. Finally, the coping strategy domains were all significantly correlated with HRQOL. Higher use of the engagement strategies was associated with higher HRQOL (problem-focused r = .490, emotion-focused r = .287), while conversely, higher use of the disengagement strategies was associated with lower HRQOL (problem-focused r = -.446, emotion-focused r = -.616).

With regards to the subscales of the SRS-22r (function, pain, self-image, and mental health), small to moderate correlations were observed between these subscales and the clinical/demographic factors age, curve size, length of diagnosis, and brace wear. Increasing age, curve size, and length of diagnosis were associated with lower scores (reduced HRQOL) in these domains, while wearing a brace was associated with higher scores. As observed for

overall HRQOL scores, greater use of engagement coping strategies was associated with better scores in function, pain, self-image, and mental health domains, while disengagement strategies were associated with lower scores across these domains. An expanded correlation table including the SRS-22r subscales is presented in Appendix I.

Hierarchical Regression

The demographic and clinical variables entered in step 1 explained 29% of the variance in HRQOL, $F(4, 107) = 10.639$, $p < .001$. In step 2, coping strategies brought the total variance in HRQOL explained by the model to 61%, $F(8, 103) = 19.827$, $p < .001$. In this final stage of the model, significant predictors of HRQOL were curve size ($p = .002$), problem-focused engagement ($p = .003$), problem-focused disengagement ($p = .004$), and emotion-focused disengagement ($p = .000$). Results for overall HRQOL are presented in Table 4.7.

Table 4.7

Hierarchical Regression Analysis Predicting Health Related Quality of Life (SRS-22r)

Variable	*B*	*SE B*	95% CI	β	ΔR^2	R^2
Step 1					.29***	.29
Age	-.02	.03	-.08, .04	-.07		
Curve size	-.01	.00	-.02, -.01	-.33***		
Length diagnosis	-.09	.04	-.16, -.01	-.23*		
Brace	-.34	.14	-.61, -.06	-.23*		
Step 2					.32***	.61
Age	-.03	.02	-.08, .02	-.10		
Curve size	-.01	.00	-.01, -.00	-.21**		
Length diagnosis	-.04	.03	-.10, .02	-.11		
Brace	-.17	.11	-.38, .05	-.11		
PF Engagement	.21	.07	.07, .34	.22**		
EF Engagement	.01	.06	-.12, .13	.01		
PF Disengagement	-.23	.08	-.38, -.07	.20**		
EF Disengagement	-.30	.06	-.42, -.18	-.38***		

*** $p < .001$, ** $p < .01$, * $p < .05$.

Note. Adjusted R^2 for Step 1 =.26, Step 2 = .58. CI = confidence interval (lower limit, upper limit).

A summary of the regression analysis with the SRS-22r subscales is presented in Table 4.8. Full regression output tables for the subscales are presented in Appendix J. In step 2 of the models, disengagement coping was a significant predictor of poorer functional outcomes, while higher curve size, length of diagnosis, and emotion-focused disengagement were predictors of higher pain. For self-image and mental health domains, problem-focused engagement predicted better outcomes while disengagement strategies were predictive of poorer outcomes, and more severe curves predicted lower self-image scores.

Table 4.8

Hierarchical Regression Analyses for SRS-22r Subscales

	Function		Pain		Self-Image		Mental Health	
	β	ΔR^2	β	ΔR^2	β	ΔR^2	β	ΔR^2
Step 1		.16**		.25***		.25***		.18***
Age	-.01		-.02		-.05		-.15	
Curve size	-.23*		-.29**		-.38***		-.21*	
Length diagnosis	-.19		-.31**		-.16		-.12	
Brace	-.21*		-.19		-.18		-.19	
Step 2		.21***		.15***		.30***		.30***
Age	-.02		-.05		-.07		-.17	
Curve size	-.12		-.24**		-.26**		-.10	
Length diagnosis	-.11		-.20*		-.05		-.01	
Brace	-.15		-.10		-.08		-.07	
PFE	.12		.12		.21**		.29**	
EFE	.01		.06		.03		-.08	
PFD	-.28**		-.07		-.22**		-.16*	
EFD	-.27**		-.26**		-.34***		-.37***	
Total R^2	.37		.40		.55		.48	
Adjusted R^2	.33		.36		.51		.44	

*** $p < .001$, ** $p < .01$, * $p < .05$.

4.3.3.2. Regression Model 2: Body Image Outcomes

Correlations

Correlations among the predictor variables (demographic and clinical variables, appearance-related variables) and the dependent variables (body image disturbance and appearance satisfaction) are shown in Table 4.9. Both age and length of diagnosis were significantly positively correlated with levels of appearance investment (r = .355, r = .308, respectively). Increasing curve size was associated with lower scores on the TAPS (indicating increased body asymmetry) (r = -.617) and increased perceived visibility scores (r = .470). Bracing treatment was associated with reduced perceived asymmetry on the TAPS measure (r = -.242).

Table 4.9

Pearson Correlations Among Demographic & Clinical Variables, Appearance Variables, and Body Image Outcomes

Measure	1	2	3	4	5	6	7	8	9
1. Age	-								
2. Curve Size	-.048	-							
3. Length Diagnosis	.538***	-.024	-						
4. Brace (Yes/No)[a]	.380***	.206*	.031	-					
5. TAPS	-.013	-.617***	-.149	-.242**	-				
6. Perceived Visibility	.046	.470***	.175	.128	-.662***	-			
7. Appearance Investment MBSRQ	.355***	-.165	.308**	.155	.063	.048	-		
8. Body Image Disturbance BIDQ-S	.277**	.321**	.292**	.319**	-.492***	.494***	.318**	-	
9. Appearance Satisfaction BESAA-A	-.277**	-.347***	-.226*	-.333***	.353***	-.428***	-.486***	-.699***	-

*** $p < .001$, ** $p < .01$; * $p < .05$ (two-tailed). [a] Point biserial correlation reported for dichotomous variable.
Note. Cohen's Pearson r effect size guidelines: small = 0.10, medium = 0.30, large = 0.50.

In relation to the body image outcome variables, increasing age and curve size were associated with lower levels of appearance satisfaction (r = -.277, -.347, respectively) and increasing age, curve size, and length of diagnosis were associated with higher levels of body image disturbance (r = .277, .321, .292, respectively). Wearing a brace was also associated with higher appearance satisfaction (r = -.333) and lower body image disturbance (r = .319). Lower scores on the TAPS (i.e. increased body asymmetry) was related to lower levels of appearance satisfaction (r = .353) and higher levels of body image disturbance (r = -.492).

Similarly, participants who felt their scoliosis was more visible experienced lower appearance satisfaction (*r* = -.428) and more body image disturbance (*r* = .494). Finally, higher levels of appearance investment were associated with lower appearance satisfaction (*r* = -.428) and higher levels of body image disturbance (*r* = .494).

Hierarchical Regression

In testing the models predictive value for appearance satisfaction, the demographic and clinical variables entered in step one contributed to 25% of the variance, $F(4, 106) = 8.784$, $p < .001$. In step 2, addition of subjective perception of appearance (TAPS), appearance investment, and perceived visibility resulted in the model accounting for 51% of the variance in appearance satisfaction, $F(7, 103) = 15.468$, $p < .001$. Curve size ($p = .007$), brace wear ($p = .029$), perceived visibility ($p = .009$), and appearance investment ($p = .000$) were significant predictors of appearance satisfaction. Results are presented in Table 4.10.

Table 4.10

Hierarchical Regression Analysis Predicting Appearance Satisfaction (BESAA)

Variable	*B*	*SE B*	95% CI	β	ΔR^2	R^2
Step 1					.25***	.25
Age	-.05	.04	-.14, .03	-.14		
Curve size	-.01	.00	-.02, -.01	-.32***		
Length diagnosis	-.06	.05	-.16, .05	-.12		
Brace	-.41	.18	-.78, -.05	-.22*		
Step 2					.26***	.51
Age	-.02	.04	-.10, .06	-.05		
Curve size	-.01	.00	-.02, -.00	-.25**		
Length diagnosis	.01	.04	-.07, .10	.03		
Brace	-.34	.15	-.64, -.04	-.18*		
Perceived visibility	-1.10	.41	-1.92, -.28	-.25**		
TAPS	.04	.11	-.17, .26	.04		
MBSRQ-AO	-.55	.09	-.72, -.38	-.49***		

*** $p < .001$, ** $p < .01$, * $p < .05$.

Note. Adjusted R^2 for Step 1 = .22, Step 2 = .48. CI = confidence interval (lower limit, upper limit).

In testing the models predictive value for scoliosis-related body image disturbance (as shown in Table 4.11), the clinical and demographic variables entered in step 1 explained 30% of the variance, $F(4, 105) = 11.137$, $p < .001$. The addition of TAPS, perceived visibility, and appearance investment in step 2 increased the model's predictive value to 53%, and these three variables were the only significant predictors of body image disturbance in step 2 (p = .013, .001, .000, respectively).

Table 4.11

Hierarchical Regression Analysis Predicting Body Image Disturbance (BIDQ-S)

Variable	*B*	*SE B*	95% CI	β	ΔR^2	R^2
Step 1					.30***	.30
Age	.06	.04	-.02, .13	.18		
Curve size	.01	.00	.01, .02	.34***		
Length diagnosis	.07	.04	-.02, .15	.17		
Brace	.31	.15	.01, .61	.15*		
Step 2					.24***	.53
Age	.04	.03	-.02, .10	.14		
Curve size	.00	.00	-.00, .01	.08		
Length diagnosis	.005	.04	-.07, .08	.01		
Brace	.19	.13	-.06, .44	.12		
Perceived visibility	1.15	.35	.46, 1.8	.31**		
TAPS	-.22	.09	-.40, -.05	-.26*		
MBSRQ-AO	.30	.07	.16, .45	.31***		

*** $p < .001$, ** $p < .01$, * $p < .05$. TAPS = Trunk Appearance Perception Scale.
Note. Adjusted R^2 for Step 1 = .27, Step 2 = .50. CI = confidence interval (lower limit, upper limit).

4.3.4. Support Needs & Preferences Data

4.3.4.1. Information Needs

89% (n = 102) of participants reported accessing scoliosis information from a medical professional, 73% (n = 84) accessed information on the internet, and 52% (n = 60) indicated that they got information from parents. 13% specified another information source, including

friends with scoliosis (*n* = 8), support groups (*n* = 4), physiotherapist (*n* = 2), and Instagram (*n* = 1). Nearly half of participants (*n* = 56, 49%) were happy with the amount of information they had, and the other half reported requiring more information about scoliosis (*n* = 58, 51%). Topics that participants wanted more information about are summarised below in Table 4.12. Of the 58 participants who specified where they would like to access more information about scoliosis, 32 (55%) selected from a website, 16 (28%) selected from their medical team, and 3 (5%) selected a leaflet. Most participants reported talking with their family members about scoliosis, with 4% (*n* = 4) reporting 'never', 13% (*n* = 15) reporting 'rarely', 47% (*n* = 54) reporting 'sometimes', 28% (*n* = 32) reporting 'often', and 8% (*n* = 9) reporting 'very often'.

Table 4.12

Scoliosis Topics That Participants Required More Information About (n = 58)

Information Topic	*n*
What is scoliosis?	7
How to look after/ manage my scoliosis	44
Patient stories	30
Bracing treatment for scoliosis	18
Surgery for scoliosis	27
Other topic[1]	15

[1]Other topics listed by fewer participants included scoliosis-related pain, what to expect post-surgery/ post-bracing, living with scoliosis as an adult, and causes of scoliosis.

4.3.4.2. Peer Support

In terms of previous contact with others with scoliosis, 21% (*n* = 24) of participants reported that they had never met another young person with scoliosis. 61% (*n* = 70) had previously met another young person with scoliosis in person, while the remaining 18% (*n* = 20) had been in contact online. Of the 90 participants who had previously met another young person with scoliosis either online or in person, 97% reported that this was a positive experience, while just 3% reported this as a negative experience. Descriptions of the negative experiences included that they were not affected by scoliosis in the same way, or that the other person's

post-surgical experience had worried them. Reasons described by participants for the encounter being a positive experience are summarised in Table 4.13.

Table 4.13

Participants' Positive Experiences of Meeting Others with Scoliosis (n = 87)

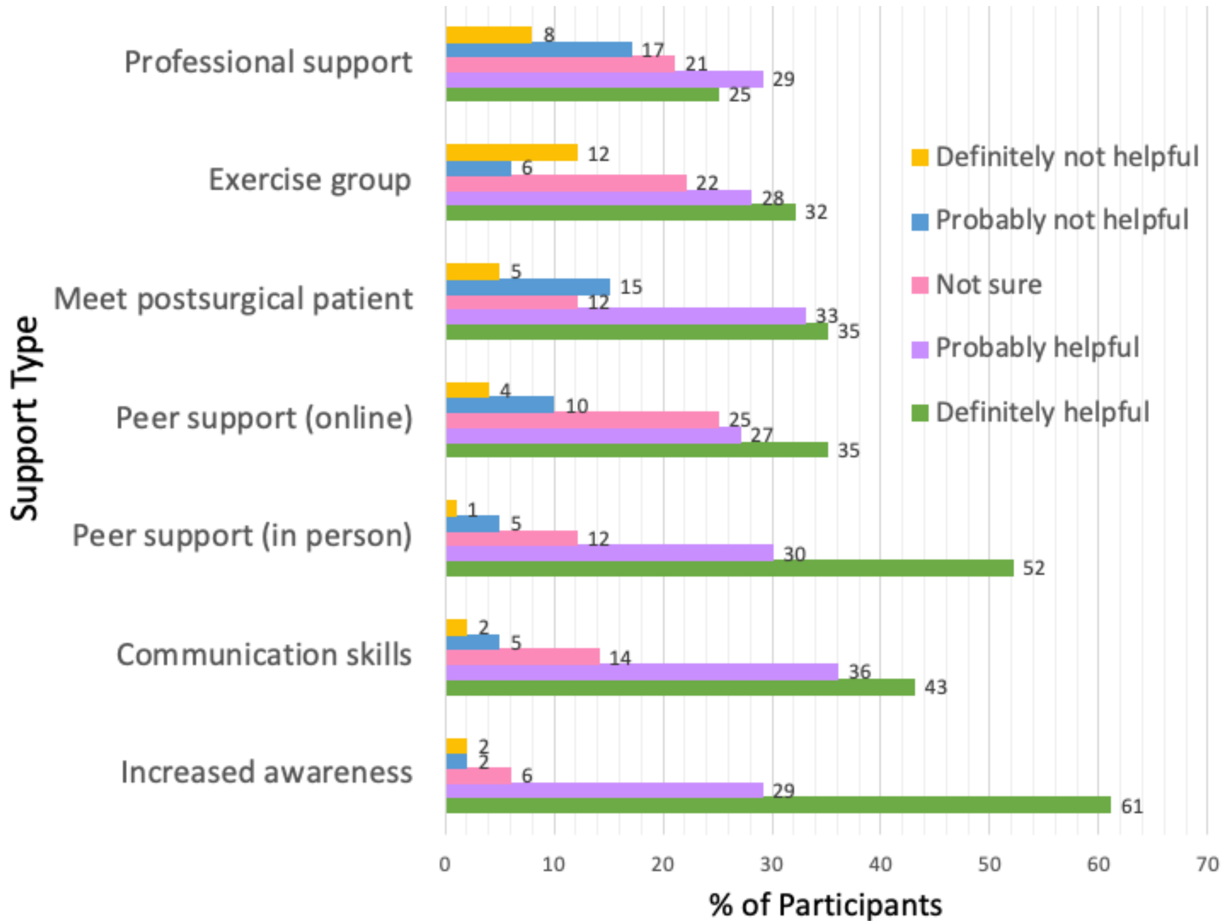
Benefit	*n*	Sample Quotations
Someone who understands scoliosis	30	*"It's nice to connect with someone who knows what I'm going through in a way other people just can't."*
Gaining information and tips about scoliosis	12	*"She knew a lot more about what was going on than I did so she helped me learn about it."*
General positivity	13	*"Amazing experience (can say the BEST)."*
Feeling less alone	12	*"It made me feel a lot more comfortable with my scoliosis and it made me accept it better once I saw that I wasn't alone."*
Someone to talk to about scoliosis	11	*"It is very helpful to meet others with scoliosis and talk about our struggles."*
Reassurance from someone at later stage of treatment	9	*"See others at the different stages, see others post- surgery bouncing back."*
Growth of a friendship	6	*"I was nervous at first but after getting to know each other we became really good friends."*

4.3.4.3. Support Preferences

Participants' usefulness ratings of potential support strategies are presented in Figure 4.7. Top rated supports were increased community awareness of scoliosis as 90% (*n* = 103) rated this as probably or definitely helpful. This was followed by in-person peer support (rated by 82% (*n* = 93) as probably or definitely helpful) and learning how to explain their scoliosis to others (79%; *n* = 90).

Figure 4.7

Participants' Usefulness Ratings of Potential Supports



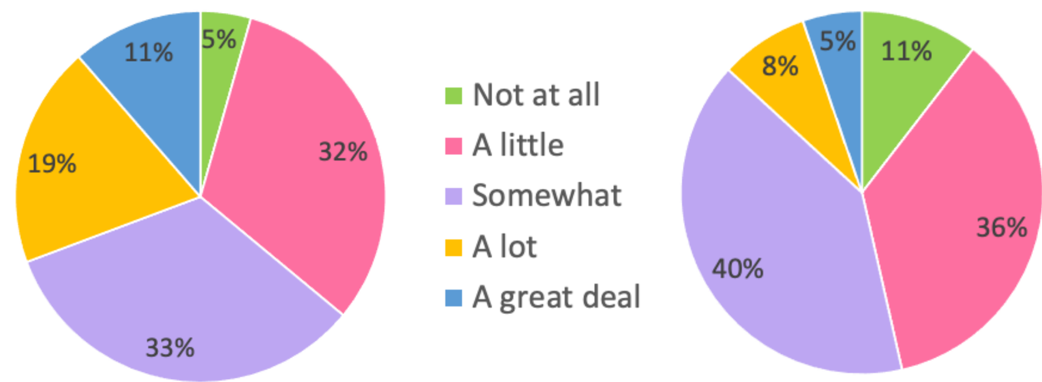
4.3.5. COVID-19 Pandemic Data

4.3.5.1. Negative & Positive Impact of COVID-19

The extent to which participants reported that their lives had been affected in a negative and in a positive way as a result of the COVID-19 pandemic is presented in Figure 4.8. The majority of participants endorsed a negative impact on their lives (M_{score} = 3.02, *SD* = 1.07, Range 1 - 5), and conversely, most also acknowledged some degree of positive impact on their lives, with only 11% indicating no positive impact at all (M_{score} = 2.61, *SD* = 0.96, Range 1 - 5).

Figure 4.8

Negative (Left) and Positive (Right) Impact of the COVID-19 Pandemic on Participants' Lives



In terms of the negative impact of COVID-19 on participants' lives, the top concerns selected by participants were *"Not seeing friends in person"* (64%), *"Having to stay at home"* (44%), *"Increased stress or disorientation from not having a schedule"'* (44%), and *"Not going to school"* (43%). The top four selected positive changes to daily life resulting from the COVID-19 pandemic were *"Spending more time with family"* (34%), *"More time to relax"* (34%), *"Getting to do things I don't usually have time for like music, art, cooking"* (29%), and *"Getting more sleep"* (26%).

4.3.5.2. COVID-19 and Healthcare

Considering the impact of the COVID-19 pandemic on participants' healthcare, 71 (62%) reported that their medical treatment had been impacted, 36 (31%) reported that it had not been impacted, while 8 (7%) were unsure. Detail on how their treatment had been disrupted by COVID-19 was provided by 69 participants and these data are summarised in Table 4.14.

Table 4.14

Summary of COVID-19 Related Treatment Disruptions Described by Participants

Treatment Disruption	*n*[a]	Sample Participant Quote
Cancelled/delayed hospital appointments	25	*"Had to wait longer to see the spinal consultant."*
Unable to attend physiotherapy	16	*"Was due to start scoliosis specific physiotherapy in March but will now start in October."*
Delayed X-Ray access	14	*"My hospital is a COVID hospital so I can't get any X Rays done."*
Postponed/cancelled surgery	12	*"Surgery delayed as hospital stopped doing non-critical ops."*
Delayed brace fittings	5	*"I was supposed to get a new brace in March of 2020 but had to wait until end of June for the clinic to reopen."*

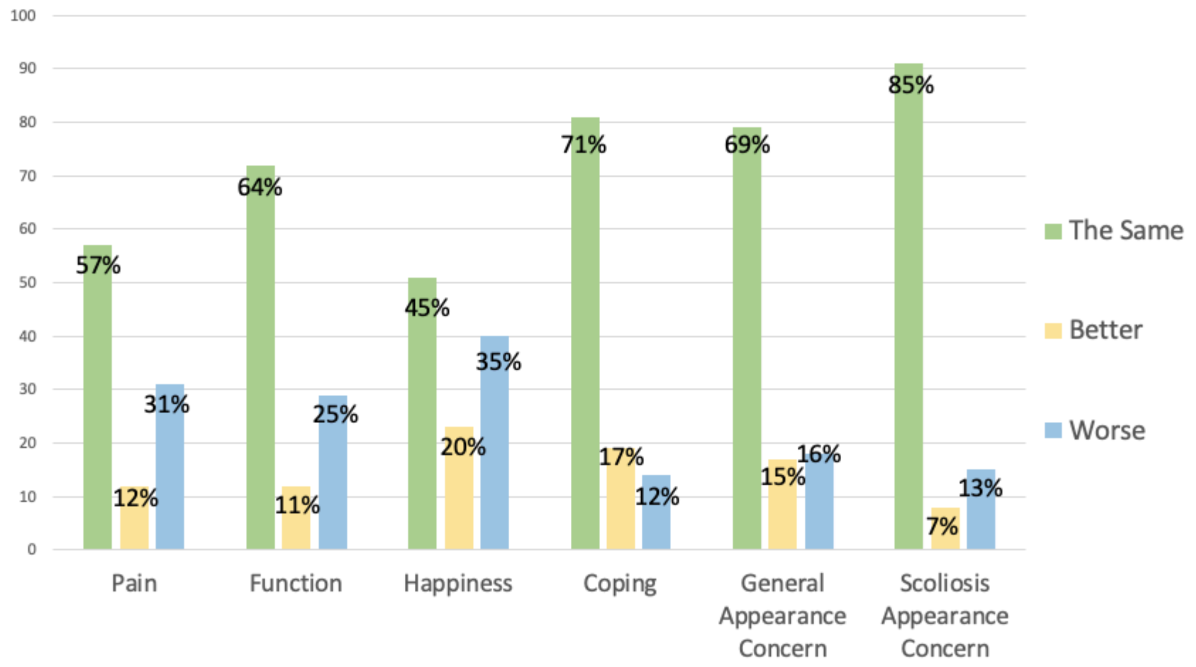
Note. [a]No. of participants does not add up to 69 as some participants mentioned more than one disruption.

4.3.5.3. COVID-19 and HRQOL/Body Image Domains

Whether participants perceived that (a) their pain levels, (b) their functional ability, (c) their happiness/ mental wellbeing, (d) the way they coped with their scoliosis, (e) their general appearance concern, and (f) their scoliosis-related appearance concerns had changed during the COVID-19 pandemic is presented in Figure 4.9. For each of these domains, most participants felt that they were the same as non-pandemic times, with the exception of happiness/ mental wellbeing whereby only 45% felt this was the same as non-pandemic times.

Figure 4.9

Participants' Perceived Changes in Quality of Life Domains as a Result of COVID-19



4.4. Discussion

This cross-sectional online survey examined clinical and demographic characteristics, psychosocial processes, and how they relate to key patient outcomes health-related quality of life (HRQOL) and body image among adolescents with AIS. Considering the findings related to clinical characteristics, higher curve magnitude was moderately associated with less favourable HRQOL and body image outcomes, which is in line with previous research (Asher et al., 2003; Cheung et al., 2019; Auerbach et al., 2014). Of interest, brace wear was associated with better function scores, lower levels of body image disturbance, and higher satisfaction with appearance in this study (in Step 1 of the regression models). While previous research investigating the impact of brace wear on psychosocial wellbeing is mixed (Schwieger et al., 2016a; Cheung et al., 2019; Zhang & Li, 2019), our findings indicated that brace wear did not have a negative effect on participants in terms of their HRQOL or body image. This may be reflective of ongoing advances in the acceptability and relative discreteness of modern orthotic braces for the treatment of AIS (Rahimi et al., 2019). Beyond these clinical characteristics, the research findings provided important insights into the role of psychosocial variables.

Coping Strategies

In terms of HRQOL, as measured by scores on SRS-22r (Regression 1), clinical and demographic characteristics accounted for 29% of the variance in HRQOL, and addition of coping strategies increased this to 61%. It was demonstrated that higher curve size and longer length of diagnosis were significant contributors to lower HRQOL, and wearing a brace was associated with better HRQOL scores. When these variables were controlled for, the types of coping that adolescents used were shown to significantly influence their HRQOL. In particular, greater use of problem-focused engagement (e.g., problem solving and cognitive restructuring) contributed to higher HRQOL, while both problem and emotion-focused disengagement (e.g., wishful thinking, social withdrawal) were associated with lower HRQOL. These findings broadly align with previous literature which demonstrates that engagement coping is associated with more positive quality of life outcomes and that disengagement coping is associated with more negative outcomes, among adolescents in general and those with chronic health conditions (Compas et al., 2001; 2012; Jaser & White, 2010).

Identification of effective coping strategies in the context of AIS has positive clinical implications, as research with other paediatric conditions demonstrates that adaptive coping strategies can be taught to patients with the aim of promoting wellbeing (Blount et al., 2008; Grey et al., 2000; Serlachius et al., 2012). However, while this insight into adaptive and non-adaptive coping is helpful in guiding the promotion of certain strategies over others, further research is needed to understand patterns of coping strategy use among adolescents with AIS and how they may use different combinations of coping strategies. Examining individual dimensions of coping as predictors of HRQOL may not capture the dynamic process of coping, and previous researchers have recommended identifying different patterns or combinations of strategies used by adolescents, and how these may relate to HRQOL (Russell et al., 2015; Oppenheimer et al., 2018). Building on the findings within this chapter, the following Chapter 5 takes these recommendations on board, utilising cluster analysis to study patterns of coping among adolescents with idiopathic scoliosis.

In terms of the subscales of the SRS-22r, it was notable that coping strategies did not demonstrate as much explanatory power for pain as they did for other HRQOL domains. Pain in AIS is caused by the musculoskeletal changes resulting from the spinal curve that can lead

to overstretching or compression of muscles and connective tissues on either side of the curvature, and it is recognised that physical factors such as rigidity of the spinal curve can be associated with higher levels of pain (Smorgick et al., 2013). It is plausible that coping strategies alone may not improve this. For those with significantly progressed spinal curvatures, surgery may be required to aid in reducing pain. It was also demonstrated that almost one third of participants reported worse pain during the COVID-19 pandemic, which may suggest that access to specific treatments, supports, or exercise play an important role in pain management. Furthermore, there could be other pain-specific coping strategies relevant for scoliosis (e.g., resting, stretching) that were not assessed in the present study.

Appearance-Related Cognitions

For Regression 2, which investigated the body image outcomes (i.e., appearance satisfaction and body image disturbance), clinical and demographic characteristics accounted for 25% and 30% of variance in these outcomes, respectively. Increasing curve size was associated with lower appearance satisfaction and higher levels of body image disturbance, while wearing a brace was associated with better outcomes in these domains. Appearance-related cognitions including subjective perception of appearance (TAPS), perceived visibility, and appearance investment made a significant contribution to the explanatory power of the model, increasing the variance accounted for to 51% and 53%, respectively. Greater perceived body asymmetry on the TAPS was related to higher levels of body image disturbance. Furthermore, higher perceived visibility of AIS and higher investment in appearance significantly contributed to lower appearance satisfaction and higher body image disturbance. These findings highlight the importance of the adolescents' appearance-related cognitions in determining the impact that the appearance-altering nature of AIS can have on their body image.

Of note is the finding that subjective perception of appearance was significantly associated with body image disturbance when controlling for curve size. This finding is in line with previous reports on other appearance-altering conditions emphasizing that an individuals' subjective perceptions can be more indicative of body image disturbance and associated impairment in functioning than objective indicators of condition severity (Moss, 2005; Feragen & Stock, 2018). This also supports the proposal of authors who created the BIDQ-S that adolescents' subjective perception of their scoliosis would be an important predictor of

this outcome, rather than curve size (Auerbach et al., 2014). To the best of our knowledge, this is the first study to test this prediction using the BIDQ-S with an AIS population.

Higher investment in appearance, whereby appearance is more central to one's self-worth, was associated with worse body image outcomes among those with AIS. While this form of "evaluative" appearance investment can be maladaptive, it is worth noting that authors have previously highlighted an alternative form of adaptive appearance investment that can be beneficial to those who undergo changes to their appearance (Tylka & Wood-Barcalow, 2015). This is characterised by a benign "motivational" investment, that can involve engaging in appearance-related self-care, or grooming behaviours that project individuality or personal style. More research is required to understand adaptive appearance investment among young people (Holmqvist-Gattario & Frisén, 2019), and how this could be relevant or helpful to adolescents with chronic health conditions such as AIS. Interestingly, previous researchers have also suggested that young people with appearance-altering conditions may endorse lower levels of evaluative appearance investment in comparison to non-affected peers, as an adaptive effort to emphasize other aspects of their self-concept and reduce importance placed on appearance (Crerand et al., 2017; Stock & Feragen, 2017). Future work may seek to investigate this concept with a comparison sample.

Research on the concept of perceived visibility among populations with health conditions affecting appearance would benefit from a comprehensive and reliable, validated measure of this construct. Previous research on the visibility/ invisibility dimension has typically focused on individuals with conditions assumed predominantly 'invisible' (e.g. a psychiatric condition) or 'visible' to others (e.g. conditions requiring mobility aids) (Shpigelman & HaGani, 2019). However, a condition like AIS is not possible to dichotomise in terms of visibility, as there are varying degrees of physical severity and contexts when the condition may be more visible (e.g. depending on clothing). While our research has focused solely on those with AIS, it is plausible that this may also be the case for other paediatric conditions of varying visibility such as burn injuries. Perceived visibility has also been recently highlighted by Gee et al. (2020), as a relevant factor which could contribute to adjustment in various paediatric conditions, based on interviews with health professionals. In the SCOLI survey, it was found that 76% of participants felt their scoliosis was visible "sometimes." A more comprehensive

measure of this construct could include aspects such as effort expended in trying to reduce visibility of the condition, perceived awareness among others about the condition, and confidence in telling others about the condition, all of which were described in the subtheme *"A Hidden Condition"* presented in Chapter 3 (page 36).

Support Preferences

Overall, findings related to adolescents' support needs and preferences provide implications for practice, and inform future development of resources. It was notable that 90% of participants felt increased awareness would be probably or definitely helpful, and 79% felt this way about increasing their health-related communication skills. This highlights the potential "hidden" nature of AIS that was discussed in the qualitative findings in Chapter 3, and of some paediatric conditions in general. Other previous qualitative research has reported that paediatric patients with diabetes or cancer felt more awareness of their conditions was needed in the school setting to enhance acceptance and help with the issue of peer questioning (Gannoni & Shute, 2010). Endorsement of a peer support approach was apparent, which builds on previous research providing support for online peer support in relation to surgical treatment of AIS (MacCulloch et al., 2009; 2010). In particular, 82% reported that it would be helpful to interact with other people their age with scoliosis in person, while 62% reported that interacting online would be helpful. Of the 90 participants who reported previously interacting with another young person with scoliosis, almost all reported that this was a positive experience. Positive aspects included shared understanding and feeling less alone, having someone to talk to about scoliosis, and gaining information/ tips or reassurance from someone at a later stage of treatment. These benefits are in line with previous literature describing the key psychosocial mechanisms by which peer support strategies for adolescents with chronic conditions can act (Olsson et al., 2005). Adolescence is also considered an opportune time for peer support strategies given the importance of peer relationships as a source of support during this developmental period, and the potential for chronic health conditions to interfere with usual social activities (Ahola Kohut et al., 2018).

COVID-19

The pandemic had both negative and positive impacts on participants, with 5% reporting no negative impact, and 11% reporting no positive impact. This is comparable to findings of a national COVID-19 survey in Ireland, whereby only 1% of young people reported no challenges, and 10% reported no positives (SpunOut, 2020). Moreover, in the SCOLI survey, top indicated negative changes (i.e., not seeing friends in person, not going to school) and positive changes (i.e., spending more time with family, getting more sleep) were comparable to previous research utilising the CASPE among adolescent females in the US who rated similar top positives and negatives (Silk et al., 2021). In terms of how COVID-19 may have impacted the HRQOL and body image domains included in this survey, it was shown that most participants felt the same within these domains as non-pandemic times. For example, 69% reported that their feelings about their appearance had not changed, and 71% felt they were coping the same. This suggests that in general, survey responses are likely to be reasonably comparable to non-pandemic times. The domain most affected by the pandemic was mental wellbeing, with 45% feeling they were the same in this regard, 20% feeling better off, and 35% feeling worse. This provides an indication as to how survey responses may have been influenced. For example, as 35% reported their mental wellbeing to be worse off due to the pandemic, it is possible that this could have translated into lower scores for some participants on the SRS-22r mental health domain, and this should therefore be considered in interpretation of study findings.

COVID-19 data also indicated that over half of participants experienced disruption to their medical care during the pandemic. This is important to consider as treatment and appointment delays may lead to worse outcomes such as more complex curvatures or decreased HRQOL. In the case of major surgery delay and severe progression, this could lead to increased surgical complexity and cost and delayed return to function for patients, as these effects are demonstrated for curvatures exceeding 70° (Tarrant et al., 2016). Future research is needed to examine the extent of the impact that postponement of such surgeries can have on patients with surgical AIS in terms of their quality of life and long-term outcomes.

Although the COVID-19 pandemic represented a potential barrier to this study, aspects of the study design were adapted to facilitate successful completion. Among these were the use of

the online survey platform and a remote recruitment strategy. The high study completion rate (85%) and low level of missing data (<5%) demonstrate a high level of engagement in the study. Informed selection of study variables and measures, aided by stakeholder consultations to ensure relevance of the survey is likely to have contributed to this outcome. Furthermore, distribution of vouchers may have aided completion rates as research indicates that the quality of online survey responses are enhanced through this approach (David & Ware, 2014). Furthermore, previous research in a paediatric orthopaedic department demonstrated that 84% of adolescents with AIS reported a preference for internet-based research (Nitikman et al., 2017). As suggested by Stiles-Shields et al. (2020), the online approach may have increased the reach of the study to those who may not usually engage during face-to-face research interactions. However, it must also be acknowledged that those without stable internet access or not linked in with the selected recruitment channels were less likely to be represented.

Limitations and Future Directions

Although this study has identified a number of potentially important variables contributing to quality of life and body image among adolescents with idiopathic scoliosis, it must be emphasised that this survey was cross sectional in nature and as such, causal inferences cannot be made. Due to the remote online nature of this study, we relied on self-reported scoliosis Cobb angles and it is possible that reports may have been inaccurate in some instances. In Appendix G, an exploratory comparison of self-reported Cobb angles and corresponding clinician assessed measurements was conducted using data collected for the study in Chapter 3. This demonstrated a positive correlation between the two sources of Cobb angle measurement (r = 0.83), indicating that self-reported Cobb angles appear to be reasonably reliable although not the typical approach. Other recent research has also described using self-reported Cobb angles (Wetterkamp et al., 2017). Future research could seek to further examine the reliability of this approach through comparison of clinical and self-report data, as this would be useful to inform future research using a remote, online approach with the AIS population. Furthermore, recruitment was directed through community/ support group channels and therefore our sample represents those who may be more likely to seek support in relation to their scoliosis. As participants may have already valued obtaining support for AIS, this could be partially responsible for the high endorsement

of the suggested support strategies in the support needs and preferences section of the survey. While these groups represent a valued recruitment channel, future research should seek to also recruit through the clinical setting to capture the views of those who may not be linked in with community groups.

In this study, coping strategies and appearance-related cognitions including perceived visibility, appearance investment, and subjective perception of appearance, were studied in terms of their contribution to HRQOL and body image outcomes in AIS. However, there are likely to be many other variables contributing to HRQOL and body image that were not included in this study. Drawing specifically on the appearance aspect of chronic conditions, factors such as the extent to which young people compare themselves to others, sociocultural pressures and expectations in relation to appearance, and parental appearance-related attitudes and values have been described as variables that may play a role in young people's adjustment to appearance-altering conditions (Gee et al., 2021; Clarke et al., 2014). In their qualitative study with health professionals, Gee et al. (2021) suggest that high appearance investment among parents may influence the child's adjustment, and such interrelations between parent and child represent an avenue for future studies in AIS.

In general, the framework used in this study provided a useful approach on how to group different types of variables or factors that can contribute to outcomes among the population under study. However, the terminology originally included in the working framework proposed by the ARC collaboration is worth considering, specifically the heading of "intervening processes" (Rumsey et al., 2008). "Processes" can be understood as a set of interpersonal, affective, cognitive, or behavioural factors or variables, the study of which assesses how they may contribute to, and influence particular outcomes of interest (American Psychological Association, n.d.). However, the term "intervening" is suggestive of a potential mediating influence, which was not investigated in this study. Rather, the framework developed for this study was conceptualised as a preliminary framework for investigating the relative contribution of specific variables to key outcomes among adolescents with AIS. On reflection, this was a suitable step in identifying psychosocial variables contributing to the wellbeing of adolescents with AIS, given the dearth of research on this topic. To build on this, future work may seek to further develop the preliminary framework presented in this study,

through investigation of other variables as suggested above, and potential mediator and moderator variables.

Conclusion

The findings of this study demonstrate the importance of coping strategies and appearance-related cognitions in contributing to key outcomes in AIS (i.e. HRQOL and body image), relative to the influence of clinical characteristics which have dominated previous research on AIS. The coping strategies used by adolescents with AIS and how they relate to HRQOL represents a promising avenue for future research, and the following Chapter 5 seeks to investigate this relationship further. The impact of AIS on body image is recognised as a key aspect of the condition, and this study has demonstrated the subjective nature of body image through the examination of perceived visibility and perception of appearance (TAPS), as well as the potential role of investment in appearance. This represents progress toward understanding the multidimensional nature of body image in AIS. Findings also highlight the need to consider how the COVID-19 pandemic has impacted upon those living with paediatric health conditions, as well as how it has impacted upon paediatric research, in terms of both conducting and interpreting research. Furthermore, this study has provided important insight into support preferences among the AIS patient group, which may encourage future research and development in relation to these topics.

Chapter 5

Examining the Relationship Between Coping and Health-Related Quality of Life Among Adolescents with Idiopathic Scoliosis

Chapter Overview

This chapter supplements Chapter 4 by presenting a cluster analysis of the coping data collected as part of the SCOLI survey in order to identify common patterns of coping used by adolescents with AIS. In Chapter 4, findings from the regression model indicated that coping strategies were significant predictors in explaining variance in health-related quality of life (HRQOL), when controlling for relevant clinical and demographic characteristics. This demonstrated that coping strategies used by adolescents in response to their scoliosis can play an important role in their HRQOL. Given this finding and the range of coping strategies measured within the SCOLI survey, cluster analysis was deemed appropriate to obtain further understanding of the relationship between coping and HRQOL. This chapter begins with an overview of coping in the paediatric literature, followed by coping in adolescent idiopathic scoliosis, and the assessment of coping strategies. Next, the specific aims of this follow-on analysis and a description of the methodology are presented. The resulting insights into participants' coping patterns and their relationship with HRQOL are detailed, along with a discussion of these findings.

5.1. Introduction

5.1.1. Coping

It is well recognised that the coping strategies used by a young person to manage the demands or stressors associated with having a paediatric health condition can influence their adjustment and wellbeing (Blount et al., 2008; Compas et al., 2012). Lazarus and Folkman (1984) classically defined coping as cognitive and behavioural efforts to manage external and internal demands that are appraised as taxing, and coping is understood to be a multidimensional and dynamic process among young people (Compas et al., 2001). The specific dimensions or structure of coping responses have been conceptualised in multiple ways in previous decades, but there is a common recognition of higher order coping

dimensions under which lower order ways of coping are nested (Skinner et al., 2003). See Figure 4.3 (Chapter 4, page 61) for the hierarchical structure of the Coping Strategies Inventory, a widely used measure of coping (Tobin, 1991) recommended for use in paediatric research (Blount et al., 2008).

Higher order coping dimensions reflect “families” or general domains of coping, typically distinguished by problem-focused vs. emotion-focused coping, and engagement vs. disengagement or approach vs. avoidance coping (Skinner et al., 2003; Blount et al., 2008). Problem-focused coping traditionally refers to attempts to change or manage the stressor, while emotion-focused coping refers to regulating emotions related to the stressor (Lazarus & Folkman, 1984). Both problem- and emotion-focused strategies can be grouped into the dimensions of engagement (or approach) coping and disengagement (or avoidance) coping. While engagement coping encompasses responses directed towards the stressor, disengagement coping involves responses that allow the individual to withdraw or direct attention away from the stressor (Compas, 2001).

A review by Compas et al. (2012) has considered that among youth living with paediatric health conditions, engagement coping efforts can be conceptualised as active (primary control) versus accommodative (secondary control), while disengagement coping efforts are described as passive. In terms of engagement, active coping describes efforts to change the source of stress through problem solving, or one’s response to the stressor through emotional expression. In contrast, accommodative coping includes efforts to adapt to the source of stress through reappraisal, positive thinking, acceptance, or distraction. Finally, passive or disengagement coping includes efforts to avoid the stressor or one’s reactions to it through avoidance, denial, and wishful thinking. Generally, engagement strategies are shown to be positively related to better outcomes including quality of life and depression or anxiety symptoms, while disengagement strategies are associated with worse scores on such outcomes (Jaser & White, 2010; Compas et al., 2012). Lower order categories which represent specific coping actions are nested underneath the higher order coping dimensions (Skinner et al., 2003). Assessment of these coping actions or “strategies” is discussed in the following section.

5.1.2. Assessment of Coping Strategies

Measurement of coping strategies varies across the paediatric literature. A commonly used approach is to use a suitable 'generic' measure of coping (that assesses the various higher and lower order coping domains) and direct the participant to rate the items in terms of how they cope with their specific health condition (Blount et al., 2008). In the SCOLI survey (Chapter 4), this approach was employed with the 32-item Coping Strategies Inventory (CSI; Tobin, 1991). The CSI was chosen as it is a comprehensive measure of coping that encompasses multiple coping dimensions from an engagement/ disengagement and problem/ emotion-focused perspective. Specifically, the measure assesses four engagement strategies including two problem-focused: *Problem Solving* (e.g. I work on solving the problems in the situation), *Cognitive Restructuring* (e.g. I try to look on the bright side of things), and two emotion-focused: *Emotional Expression* (e.g. I let out my feelings to reduce the stress), and *Social Contact* (e.g. I ask a friend or relative I respect for advice). Four disengagement strategies are also assessed including two problem-focused: *Problem Avoidance* (e.g. I avoid thinking or doing anything about the situation), *Wishful Thinking* (e.g. I hope a miracle will happen), and two emotion-focused: *Self-Criticism* (e.g. I blame myself), and *Social Withdrawal* (e.g. I avoid my family and friends). Participants were directed to think of times when their scoliosis might bother them or cause them stress and answer each coping item based on how they coped with their scoliosis.

Alternatively, Petersen et al. (2004) developed the Coping and Disease questionnaire (CODI) to evaluate coping responses specifically related to coping with chronic conditions in childhood and adolescence. The CODI includes six standalone domains of coping, namely Acceptance, Avoidance, Cognitive-Palliative, Distance, Emotional Reaction, and Wishful Thinking. On an exploratory basis, two CODI coping domains, Acceptance and Distance, were included in the SCOLI survey as they represented coping concepts not covered by the CSI measure that could be relevant in the context of coping with a paediatric health condition such as AIS. *Acceptance* refers to the extent to which an individual has accepted and adjusted to the presence of their condition (e.g. I have got used to my scoliosis), while *Distance* refers to appraising one's condition as less serious or minimizing its' significance (e.g. I don't care about my scoliosis). Both acceptance and distance coping have been shown to be positively

correlated with HRQOL among paediatric patients with various conditions including diabetes, asthma, and arthritis (Petersen et al., 2006; Quitmann et al., 2015).

5.1.3. Coping and Adolescent Idiopathic Scoliosis

There is limited previous research examining coping strategies in the context of adolescent idiopathic scoliosis (AIS). Lysenko et al. (2016) reported multiple coping strategies that were used by their presurgical AIS cohort, which included problem avoidance, being humorous, developing self-reliance and optimism, developing social support, and ventilating feelings, however relationships between coping and patient wellbeing were not studied. In their study focused on how coping related to surgical recovery among 113 patients with AIS, LaMontagne et al. (2004) administered a structured preoperative coping interview that rated participants along an avoidant – vigilant (i.e. engaged) dimension. More vigilant preoperative and postoperative coping (described as seeking information, focusing on positives) was a significant predictor of positive functional outcomes and academic performance 9-months postoperatively. In another study using a brief coping screening measure (KidCope), Beka et al. (2006) reported that strategies including wishful thinking, resignation, or blaming others were negatively correlated with general self-esteem among 82 children and adolescents with idiopathic scoliosis. For brace-wearers, distraction, social withdrawal, and self-criticism were also negatively correlated with general self-esteem.

While these studies provided preliminary insight into the role of coping in specific circumstances including surgical recovery and bracing treatment, previous literature has not sought to understand coping more generally among patients with AIS, in terms of how they cope with AIS-related stressors in their daily lives. Furthermore, to our knowledge no previous studies have investigated how coping responses in AIS relate to health-related quality of life (HRQOL). Our coping findings detailed in Chapter 4 represented a first step by demonstrating that disengagement coping predicted lower HRQOL among adolescents with AIS, while problem-focused engagement predicted higher HRQOL. Further investigation of how adolescents use these coping strategies is needed. Importantly, understanding coping in paediatric populations is an important step in uncovering adaptive processes and informing future development of effective support strategies and interventions (Blount et al., 2008; Compas et al., 2012; Wysocki et al., 2017, p.261).

5.1.4. Analytic Approach to Coping

While past research has examined individual dimensions of coping and their relationships with health outcomes, the value of moving toward exploring combinations of coping strategies has been highlighted (Russell et al., 2015; Oppenheimer et al., 2018). Coping is a dynamic process, and it is likely that adolescents use more than one isolated coping strategy in response to their condition, and that different strategies may be more suitable in different situations. Therefore, exploring combinations of coping strategies used by adolescents with AIS would provide a more informed understanding of how this group cope with their condition in their daily lives.

Previous authors have utilised statistical clustering techniques in order to investigate combinations of coping strategies used by paediatric populations (e.g., Claar et al., 2008; Russell et al., 2015; Oppenheimer et al., 2018). Clustering is an exploratory technique useful for identifying subgroups of participants within a dataset who are similar to each other based on a set of specific criteria (Henry et al., 2005). In a sample of adolescents with spinal cord injury, Russell et al. (2015) identified four coping profiles and found that those described as "cognitive copers" (26% of sample) reported significantly better quality of life than those with other coping profiles. The cognitive coping cluster reported higher levels of cognitive restructuring and acceptance and relatively lower use of other strategies in comparison to the other clusters which were defined as "active" (e.g. problem solving, seeking social support, cognitive restructuring), "avoidance" (e.g. social withdrawal, distraction) and "ineffective" (high levels in most coping domains, particularly blame). Another study by Oppenheimer et al. (2018) including children and adolescents with asthma, diabetes, or coeliac disease identified two clusters they described as "effective" that had higher HRQOL compared to one "non-effective" cluster. The two effective clusters were similar in that they used higher levels of both acceptance and distancing compared to the non-effective cluster, alongside varying levels of other strategies, demonstrating the benefit of these two adaptive strategies.

5.1.5. The Present Study

The present study sought to build on the findings presented in Chapter 4 by further enhancing our understanding of how adolescents use coping in response to their AIS. Rather than

focusing on discrete categories of coping alone, there is growing recognition that examining common patterns of coping strategy usage and how this relates to HRQOL is a valuable approach. Limited previous research has focused on coping among those with AIS and to our knowledge, no study has previously examined combinations of coping strategies used by this patient group or how combinations of coping strategies relate to HRQOL. The present study therefore employed clustering techniques to examine the presence of common combinations of coping strategies and following this, we sought to assess the contribution of the identified coping combinations to participants' HRQOL.

Aim and Objectives

The aim of this study was to investigate the relationship between self-reported coping strategies and health-related quality of life (HRQOL) among adolescents with AIS. Specific objectives were:

(i) To identify whether there are common patterns of coping strategy usage reported by adolescents with AIS in response to their condition.

(ii) To investigate whether specific coping strategy patterns are associated with differences in HRQOL among adolescents with AIS.

5.2. Method[2]

5.2.1. Data Analysis

Clustering is a form of exploratory data analysis that produces homogenous subgroups of data points (i.e. clusters) in a given dataset. Clusters are intended to have lower within-group variance and higher between-group variance on specific criteria relevant to the research aims (Henry et al., 2005; Steinley et al., 2006). For the current study, clustering was used to identify clusters of adolescents using similar coping strategies and the cluster groups were then compared for differences in their HRQOL. Variables used for clustering were the four subscales of the CSI: Problem Focused Engagement (PFE); Emotion Focused Engagement (EFE); Problem Focused Disengagement (PFD); and Emotion Focused Disengagement (EFD), and the two CODI subscales (Acceptance and Distance). See Chapter 4 pages 60 - 61 for a

[2] The data used in this study were collected as part of the SCOLI Survey detailed in Chapter 4. Please refer to Chapter 4 pages 55 - 63 for a description of recruitment, data collection, and the sample characteristics.

description of these measures. The CSI secondary subscale variables were used rather than primary variables due to high correlations observed between the primary subscales (i.e. $r > .7$). It is advised to avoid substantial multicollinearity among clustering variables to prevent undue influence on the analysis through implicit weighting (Hair et al., 2010, p.527). Data were checked to ensure no concerns regarding missing data, outliers, or violation of assumptions as outlined in Chapter 4. Descriptive statistics and correlations for the study variables are presented in Table 5.1.

Table 5.1

Descriptive Statistics and Correlations for Main Variables used in Clustering Analysis

Measure	*M* (*SD*)	1	2	3	4	5	6
1. CSI PF[a] Engagement	3.25 (0.73)	-					
2. CSI EF[b] Engagement	3.26 (0.79)	.36**	-				
3. CSI PF Disengagement	3.06 (0.62)	-.24**	-.19*	-			
4. CSI EF Disengagement	2.35 (0.88)	-.34**	-.37**	.37**	-		
5. CODI Acceptance	3.76 (0.83)	.63**	.27**	-.44**	-.31**	-	
6. CODI Distance	2.29 (0.99)	.28**	.11	-.24*	-.11	.54**	-
7. SRS Quality of Life	3.56 (0.69)	.49**	.29**	-.45**	-.62**	.57**	.40**

[a]PF = Problem Focused, [b]EF = Emotion Focused. $^{**}p < .01$; $^{*}p < .05$.

5.2.1.1. *Clustering Procedures*

A two-step clustering approach was used. Firstly, hierarchical clustering was conducted to identify the appropriate number of clusters followed by *k*-means clustering to confirm cluster structure and identify optimal cluster membership. This approach to obtaining clusters is intended to capitalise on the strengths of both methods (Henry et al., 2005; Hair et al., 2010, p.536) and has been used in previous research investigating coping strategies among paediatric populations (Russell et al., 2015).

For step one, using standardised *z*-score responses on the six coping strategy variables, hierarchical clustering was performed with Ward's linkage method (1963) and the Euclidean

squared distance metric, which are suitable for and commonly used in identifying homogenous clusters with scale data (Hair et al., 2010; Henry et al., 2005). Hierarchical (agglomerative) clustering begins with singleton clusters (i.e. one data object) that continue merging two clusters at a time until the final maximal cluster (i.e. all data objects) is obtained, with the advantage of allowing examination of a range of cluster solutions (Aggarwal & Reddy, 2014, p.101). Initial analyses indicated a solution in the range of 2 – 4 clusters was the most appropriate. In step two, the *k*-means clustering procedure was run with the number of clusters specified from the previous hierarchical phase, and with the SPSS-generated random selection of initial cluster centre values which is shown to be an effective and robust initialization method (Peña et al., 1999). The *k*-means algorithm groups data objects closest to the cluster centres leading to an iterative process of data objects being assigned to clusters and cluster centres being updated. This continues until convergence is achieved and no more data objects change cluster membership (Aggarwal & Reddy, 2014, p.89). The advantage of this optimizing procedure is that it allows for reassignment of data objects to obtain a more accurate cluster membership (Hair et al., 2010). Cluster membership from the *k*-means output was saved as a new variable.

The optimal cluster solution was determined using commonly accepted guidelines which included inspection of the dendrogram plot and agglomeration schedule produced in the hierarchical output and examination of cluster interpretability through ANOVA testing for significant differences between clusters on coping strategy usage (Hair et al., 2010). Further information on cluster quality was obtained through silhouette statistics which indicate a measure of cluster cohesion compared to separation. Values range between -1 and 1 with values closer to one indicating better cluster solution (Rousseeuw, 1987; Rodriguez et al., 2019). Finally, as the *k*-means algorithm may be sensitive to the specified starting conditions (i.e. the initial cluster centroid values), further validation of our clusters was conducted by running *k*-means with multiple randomly selected starting conditions and comparing solutions to assess stability (Aggarwal & Reddy, 2014; Rodriguez et al., 2019).

5.2.1.2. *ANCOVA*

When the optimal cluster solution was determined, One-Way ANOVAs and post-hoc comparisons were used to test for differences between the clusters on relevant demographic

and clinical variables including age, curve size, and length of diagnosis. Chi-Square analyses were conducted to test for differences on gender, geographical location, and brace wear. Controlling for any significant differences on these variables, ANCOVA was performed to investigate whether the coping clusters differed in relation to their HRQOL on the Scoliosis Research Society (SRS22r) measure with four subscales; function, pain, self-image, and mental health. All analyses were generated using SPSS 24.

5.3. Results

5.3.1. Cluster Solution

The hierarchical output suggested that a solution of 2 – 4 potential clusters was appropriate and solutions of two, three, and four clusters were examined in detail. The agglomeration schedule was inspected for relatively large increases in the coefficient as clusters were combined. As shown in Table 5.2, there was a relatively large coefficient increase when three clusters reduced to two (compared to previous stages), indicating that dissimilar clusters were combined. It is recommended to select the cluster solution occurring before a relatively large increase which would suggest that a three cluster solution was appropriate (Hair et al., 2010). The dendrogram in Figure 5.1 graphically displays the clustering process, with larger distances between combinations indicating a more optimal solution with greater heterogeneity between clusters. The red horizontal dotted line indicates where the dendrogram is cut to produce a three cluster solution.

Table 5.2

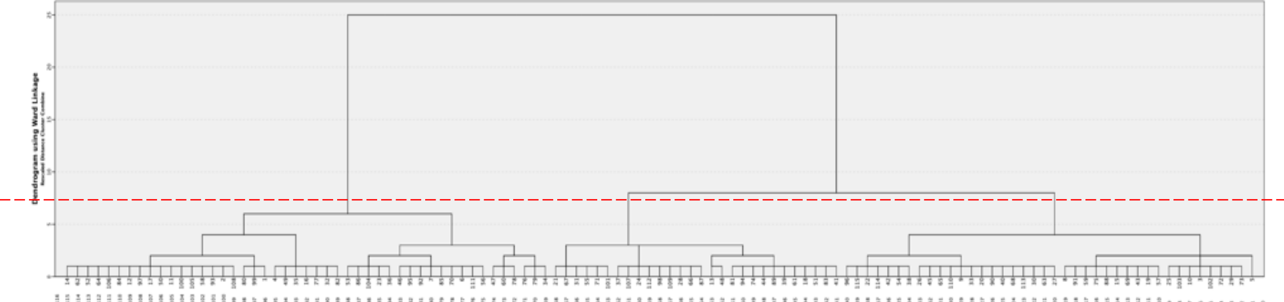
Agglomeration Schedule for Last Seven Stages of the Hierarchical Clustering Procedure

Number of Clusters	Coefficient
7	301.527
6	325.301
5	351.214
4	380.541
3	421.968
2	482.738
1	684.00

Note. Seven stages included to provide context to observed increases in coefficients as clusters are combined.

Figure 5.1

Dendrogram Produced in the Hierarchical Clustering Output



Note. Horizontal dotted line indicates where the dendrogram is cut to produce a three cluster solution.

Using *k*-means clustering with three clusters specified, participants were assigned to cluster groups.[3] On consideration of the various indices utilised within the cluster analysis, the three cluster solution was identified as the most meaningful and robust. In the three cluster solution, each cluster demonstrated a unique coping profile and there were significant differences observed between clusters on all coping strategies (using ANOVA). While there was some evidence for a two cluster solution (e.g. in agglomeration schedule), the resulting clusters did not provide meaningful insight into the various coping patterns that participants can employ. It is recognised that although a two cluster solution typically demonstrates the largest coefficient increase, it can be of limited value (Hair et al., 2010, p.551). A four cluster solution provided less distinct clusters when examined for significant between-group differences on coping strategies which hindered interpretability. This was also reflected in silhouette statistics that demonstrated lower cluster quality for the four cluster solution in comparison to three. Further detail on the indicators for the three cluster solution is provided below and description of the three clusters follows.

ANOVA. To assess whether meaningful differences were observed between the three clusters, One-Way Non-Repeated Measures ANOVAs were conducted to examine significant between-group differences on the coping strategy variables. Significant between-group differences were found as shown in Table 5.3. All three clusters differed significantly on

[3] Prior to finalisation of the three cluster solution, *k*-means was also used to obtain two and four cluster solutions for examination.

Emotion Focused Engagement, Disengagement, and Acceptance, while two groups differed significantly for Problem Focused Engagement, Disengagement, and Distance.

Table 5.3

Means, Standard Deviations, and One-Way ANOVA Statistics for Coping Strategy Use Between the Three Coping Clusters

Coping Strategy	*M (SD)*			ANOVA		
	Cluster 1 *n* = 44	Cluster 2 *n* = 30	Cluster 3 *n* = 41	*F*(2,112)	η^2	PCP[a]
PF Engagement	3.75 (0.64)	2.93 (0.65)	2.97 (0.58)	22.21***	.28	1>2, 1>3
EF Engagement	3.71 (0.75)	2.71 (0.57)	3.17 (0.72)	18.72***	.25	1>2, 1>3, 3>2
PF Disengagement	2.59 (0.57)	3.47 (0.39)	3.26 (0.49)	33.29***	.37	1<2, 1<3
EF Disengagement	1.79 (0.61)	3.45 (0.57)	2.13 (0.54)	78.35***	.58	1<2, 1<3, 2>3
Acceptance	4.55 (0.39)	3.47 (0.59)	3.13 (0.64)	79.00***	.59	1>2, 1>3, 2>3
Distance	2.91(1.01)	2.40 (0.81)	1.55 (0.50)	30.61***	.35	1>3, 2>3
Total	3.18 (0.32)	3.11 (0.23)	2.78 (0.31)	21.89***	.28	1>3, 2>3

****p* < .001. [a]PCP = Pairwise comparison procedure. Mean differences listed (with indicators > or <) are significant at *p* < .05 (Scheffé or Dunnett's C). PF = problem-focused, EF = emotion-focused.

Silhouette Statistic. The average silhouette statistic for the three cluster solution was .225. This value was higher than that achieved for other possible cluster solutions (e.g. four clusters) providing further support for the appropriateness of the three cluster solution.

Cluster Stability. *K*-means clustering for the three cluster solution was repeated five times with different initial centroid values randomly selected from the dataset. The stability of the cluster solution was then tested using crosstabs to assess the number of cases that were assigned to the same cluster across the solutions obtained with the different starting conditions. The proportion of cases that did not retain the same cluster membership ranged from 5.2% to 10.4% across the comparisons, which indicated a stable solution. It is recommended that a very stable solution is produced when there is less than 10% cluster

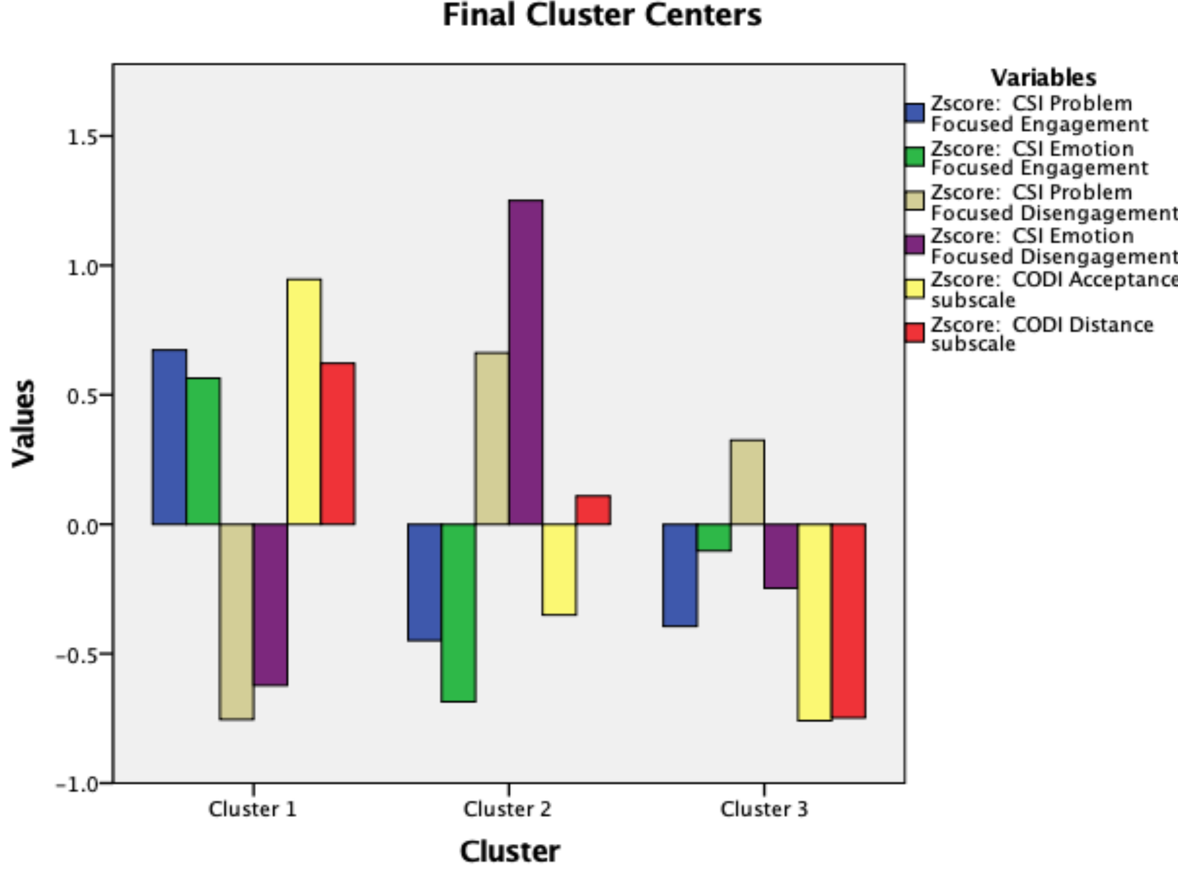
membership change, a stable solution with 10 – 20%, and somewhat stable with 20 – 25% (Hair et al., p. 540).

5.3.1.1. Description of Clusters

A bar chart depicting the final cluster centres for the three cluster solution is presented in Figure 5.2. Clusters were named 'highly engaged', 'emotionally disengaged', and 'limited strategy' to capture defining features of their coping profiles. A detailed description of each cluster follows.

Figure 5.2

Bar Chart Illustrating the Final Cluster Centres (Z Scores) Obtained with K-means Clustering



Cluster 1 'Highly Engaged' (n = 44, 38%). The highly engaged cluster were characterised by their significantly higher use of engagement coping strategies relative to the other groups. They were likely to use problem-focused engagement strategies such as

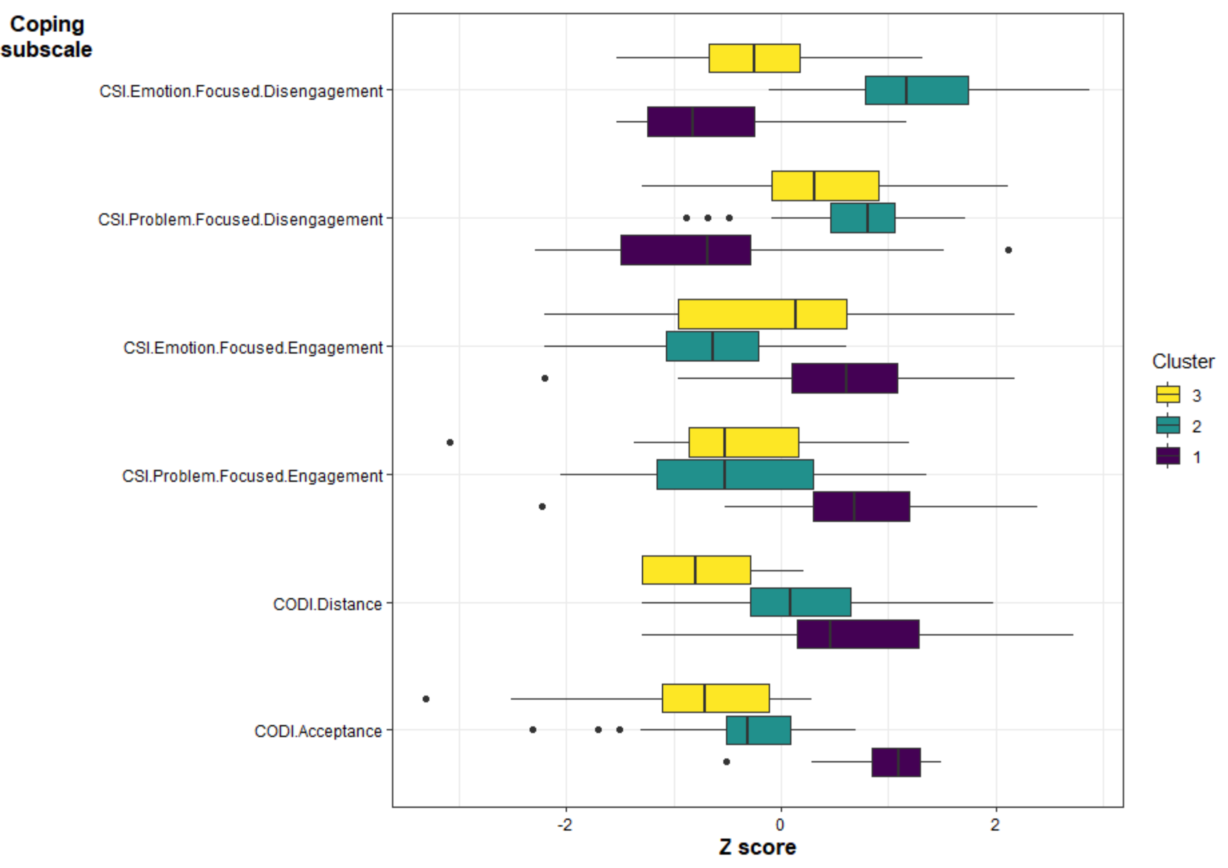
cognitive restructuring (e.g. looking on the bright side of things) and working on solving issues as well as emotion-focused strategies such as seeking social support from others and expressing their emotions. They reported significantly lower use of the disengagement strategies compared to the other clusters. They also endorsed the highest rates of both Acceptance (i.e. adjusting to and accepting condition) and Distance (i.e. appraising condition as less serious) in comparison to the other clusters. Overall this group were defined by their higher use of most coping strategies with the exception of disengagement strategies.

Cluster 2 'Emotionally Disengaged' (n = 30, 26%). Participants in the emotionally disengaged cluster were defined by their significantly higher use of emotion-focused disengagement strategies including self-criticism (e.g. blaming oneself) and social withdrawal (e.g. avoiding others). Also notable was that this cluster reported significantly lower levels of emotion-focused engagement in comparison to the other clusters. They also reported use of significantly higher levels of problem-focused disengagement and significantly lower levels of problem-focused engagement than the 'highly engaged' Cluster 1. In comparison to Cluster 3, they reported significantly higher levels of both Acceptance and Distance but they did not use these strategies to the same extent as Cluster 1. In summary, this cluster was characterised by their use of disengagement strategies, particularly their endorsement of emotion-focused disengagement and relatedly their lower use of emotion-focused engagement.

Cluster 3 'Limited Strategy' (n = 41, 36%). The final cluster reported the lowest levels of overall coping strategy usage. This cluster reported similar levels of problem-focused engagement and problem-focused disengagement to the 'Emotionally Disengaged' Cluster 2, however they reported significantly less frequent use of emotion-focused disengagement and more frequent use of emotion-focused engagement. They also did not utilise engagement strategies as often as the highly engaged Cluster 1. A distinguishable feature of this cluster was their significantly lower levels of Distancing (i.e. minimising significance of condition) and Acceptance (i.e. ability to manage/ accept situation) compared to the other two clusters. A box plot Illustrating the coping profiles of each cluster is shown in Figure 5.3.

Figure 5.3

Box Plot Illustrating the Coping Profiles of the Three Clusters



5.3.2. ANCOVA

Prior to the ANCOVA, a significant difference between clusters was observed for curve size using One-Way ANOVA, $F(2, 109) = 7.16$, $p = .001$. Participants in the highly engaged Cluster 1 had significantly smaller curves than those in the limited strategy Cluster 3 (Cluster 1 M = 35°, SD = 18; Cluster 3 M = 51°, SD = 21). The remainder of the ANOVA and Chi Square analyses for demographic and clinical variables demonstrated no significant differences between clusters with regard to age, length of diagnosis, gender, geographical location, and brace wear (all p values > .05). Curve size was therefore included as a covariate in the ANCOVA.

One-Way between-groups ANCOVA was conducted to compare the effect of the different coping profiles on participants' HRQOL, with estimated marginal means pairwise comparisons (Bonferroni corrected). The independent variable was cluster group membership and the dependent variable was scores on the SRS-22r measure, while curve size was included as a covariate. Using G*Power post-hoc power analysis (Faul et al., 2007) the sample of 112

participants provided sufficient power (.81) to detect a moderate effect of f = .3 (η_p^2 = .08) with an alpha level of .05 for an ANCOVA test with three groups and one covariate. Results are presented in Table 5.4 demonstrating that HRQOL varied significantly with cluster membership after controlling for curve size, $F(2, 108) = 22.49$, $p < .001$. The results indicated that coping cluster explained 29% of the variance in HRQOL (η_p^2 = .29). Examining pairwise comparisons, Cluster 1, the highly engaged group, reported significantly higher HRQOL than Cluster 2 and Cluster 3, while Clusters 2 and 3 did not differ significantly from each other in terms of HRQOL scores. This finding held across each of the SRS subscales including function, pain, self-image, and mental health.

Table 5.4

Adjusted Means, Standard Deviations, and ANCOVA Statistics for Health-Related Quality of Life (SRS-22r) by Cluster Group

Outcome	Cluster 1 Highly Engaged	Cluster 2 Emotionally Disengaged	Cluster 3 Limited Strategy	ANCOVA F(2,108)	η_p^2
SRS HRQOL	4.02 (0.42)	3.19 (0.69)	3.32 (0.62)	22.49***	.29
Function	4.54 (0.40)	3.79 (0.82)	3.88 (0.74)	13.78***	.20
Pain	3.91 (0.59)	3.20 (1.08)	3.38 (0.89)	6.94**	.11
Self-Image	3.89 (0.52)	3.01 (0.79)	3.05 (0.71)	21.39***	.28
Mental Health	3.75 (0.75)	2.78 (0.74)	2.96 (0.74)	16.85***	.24

Note. Means presented are estimated marginal means, SD's are not adjusted. Pairwise comparisons showed that Cluster 1 had higher quality of life than Clusters 2 & 3 in all domains ($p < .01$). SRS Range = 1 – 5 with higher scores indicating better quality of life. Covariate (curve size): $F(1,108) = 7.13$, p = .009, η_p^2 = .062.
***$p < .001$ **$p = .001$

5.4. Discussion

The present study sought to identify common patterns of coping among a sample of adolescents with idiopathic scoliosis and investigate whether these patterns of coping played a role in their health-related quality of life (HRQOL). Three coping clusters were identified, representing three subgroups within the sample who used different patterns of coping strategies. Cluster 1, the "highly engaged" group (38%) were characterised by their more

frequent used of both problem and emotion-focused engagement strategies as well as acceptance and distance. The highly engaged cluster demonstrated significantly better self-reported HRQOL in terms of function, pain, self-image, and mental health in comparison to the other two cluster groups. Cluster 2, the "emotionally disengaged" copers (26%) were defined by their frequent use of emotion-focused disengagement, while Cluster 3, the "limited strategy" group (36%) were less distinguishable, but characterised by lower use of strategies in general and significantly lower use of distance and acceptance. Notably, clusters 2 and 3 demonstrated no differences in HRQOL.

Results indicated that while curve size explained 6% of the unique variance in HRQOL, coping patterns explained 29% of the variance. This result aligns with a previous clustering study which indicated that coping patterns accounted for 20.5%, while type of disease explained 3.4% of variance in HRQOL in a paediatric population (Oppenheimer et al., 2018). Specifically, our findings demonstrated that using a range of coping strategies with lower use of disengagement strategies was associated with improved HRQOL among adolescents with AIS. In particular, those with higher HRQOL in the highly engaged cluster tended to more frequently use problem-focused engagement such as problem solving and cognitive restructuring, emotion-focused engagement including emotional expression and social support, as well as more frequent use of acceptance and distancing.

When examined closely, some of the strategies used by the highly engaged Cluster 1 may seem contradictory to each other. For example, distancing (i.e. minimising seriousness of one's condition) would appear to be a very different coping approach to the engagement strategy of problem solving (i.e. tackling issues head on). While these strategies may appear inconsistent, it is plausible that having various coping options for different contexts is advantageous. Importantly, it is acknowledged that the utility of a coping strategy is dependent on the context it is used in (Lazarus & Folkman, 1984). For example, an adolescent may want to focus on problem-solving some of the time (e.g. engaging in physiotherapy exercise or researching information about their condition/treatment), while at other times (e.g. when socialising) they may wish to detach from the seriousness of the situation or minimise the role of their scoliosis in their lives through distancing. Therefore, rather than focusing on the promotion of one specific coping strategy, it may be important for

adolescents to have a "toolkit" of coping strategies available to them, with different strategies proving useful in different contexts.

Findings can be compared to a similar cluster analysis by Russell et al. (2015) who reported that adolescents with spinal cord injuries who relied on acceptance and cognitive restructuring (and infrequent use of other strategies) had higher quality of life. In our study however, the "highly engaged" group who frequently used most coping strategies (including acceptance and cognitive restructuring as well as more active or problem-focused strategies) demonstrated higher quality of life. In considering this contrast, the more permanent nature of a spinal cord injury is notable compared to the course of AIS which is dominated by efforts to reduce severity of the condition. In their study, Russell et al. (2015) discuss that reliance on acceptance and cognitive restructuring among their "cognitive" copers may reflect a lack of need to use other strategies as they had adapted to a situation outside of their control. This is in line with accommodative coping described by Compas et al. (2012) characterised by adapting to the source of stress as opposed to a more active approach. The stressors associated with AIS could be considered more changeable due to uncertain and often rapid progression of the condition and available treatment such as bracing and surgery. Therefore, adolescents with AIS may need to be able to employ different strategies (both active and accommodative) to cope adaptively with the more changeable nature of their condition. However, while such comparisons can be made based on the conceptual dimensions of coping, more direct comparisons are hindered by the use of a variety of different coping measures across studies.

The comprehensive measurement of coping strategies was notable in this study, using the 32-item CSI measure which included engagement and disengagement strategies from both problem and emotion-focused perspectives (Tobin, 1991). This measure is recommended for use in paediatric research as a well-established measure of coping behaviours (Blount et al., 2008). The inclusion of two subscales from the CODI (Acceptance and Distance) captured two more coping concepts not captured by the CSI that were considered potentially important in the context of living with a paediatric health condition (Petersen et al., 2004). These two coping subscales were added to the survey as additional coping domains on an exploratory basis. Given the exploratory nature of the cluster analysis and the flexibility to identify

subgroups based on multiple different criteria (i.e. coping strategies), they represented a valuable addition to the analysis. Furthermore, in analysing the data a rigorous approach was taken by implementing the two-step clustering procedure in order to combine strengths of the hierarchical and *k*-means methods to obtain the most appropriate solution in terms of validity and stability. In terms of practical implications, screening of coping strategy usage among patients with AIS within orthopaedic outpatient departments may be a suitable step for future development. Through screening, those who exhibit lower levels of effective coping strategy usage and higher levels of disengagement strategies may be at greater risk of poorer HRQOL outcomes and could be referred to appropriate supports. To increase feasibility, a short, easily administered screening tool would be required. Shorter coping screening measures are available such as the KidCope (Spirito et al., 1988), an 11-item measure of generic coping among youth.

Limitations and Future Directions

There are some limitations of the clustering analysis to consider. There was some overlap observed between clusters and a small number of cluster outliers as shown in the box plot in Figure 5.3. The observed silhouette score was not very high, indicating that while the clusters were evident in the dataset they were not overly distinct. However, it is acknowledged when using real-world multivariate data that clusters commonly do not appear overly well separated even though they represent distinct groups in the dataset (Dalmaijer et al., 2020). Importantly, the three clusters identified in this study were defined by their own unique coping profiles with significant differences observed between clusters on coping strategy usage (via ANOVA), which indicated meaningful groups. Future studies seeking to identify coping patterns among other AIS samples would be useful to compare whether similar or new coping patterns are identified, as this is another way to validate the cluster solution and investigate potential generalisability of the results (Hair et al., 2010, p. 539).

Notably, the k-means algorithm used to partition the dataset is a hard-clustering procedure, whereby each data point or participant exclusively belongs to one cluster. An extension or alternative to the current analysis could involve obtaining “soft cluster” membership as this would provide more information on the fit of participants within their cluster groupings. Soft clustering techniques such as the fuzzy c-means algorithm (Dunn, 1973) indicate the degree

to which each data point begins to a cluster, so that participants can be profiled as belonging to a cluster to a greater or lesser extent (Aggarwal & Reddy, 2014). While a clustering approach was suited to the exploratory nature of this study and the sample size, latent profile analysis is another approach to identifying subgroups that could be considered in future work with larger datasets. Our findings, along with previous clustering studies on coping in paediatric populations, provide important insights into underlying structures and subgroups present in the data that such model-based approaches may build upon.

Qualitative enquiry into coping with AIS represents another avenue for future research to improve understanding of coping behaviours in this patient group. Specifically, exploring the contexts and stages of treatment in which various strategies may be considered effective, and whether certain strategies may be used interchangeably or simultaneously would add more clarity to this topic. Furthermore, it is possible that there are many other coping strategies used by adolescents with AIS that were not captured in this study. Exploring the patient perspective may help to validate the measurement of coping among this group and provide guidance for improvement (e.g. are we missing out on any other relevant coping strategies? are some strategies not deemed relevant?). For example, the inclusion of the Distance subscale in this study was in-part informed by qualitative interview data (collected as part of the study presented in Chapter 3) as a few adolescents appeared to describe instances where they had minimised the significance of their condition. This demonstrates the potential value of utilising qualitative insight particularly as coping is a relatively under researched topic in relation to AIS.

Furthermore, when more in-depth understanding is achieved about coping among adolescents with AIS and the comprehensive measurement of coping in this group, future lines of research should seek to investigate coping trajectories over the course of AIS. From the point of diagnosis, to progression of the condition over time, to undergoing relevant treatment, coping strategies used by adolescents may change or evolve. This research was limited by way of its' cross-sectional nature. Longitudinal studies would provide insight into the process of coping over time. This has been demonstrated by Fisher et al., (2021) who analysed coping trajectories among patients who had experienced childhood cancer over five years. A recent protocol by Warschburger et al. (2021) described a new longitudinal

investigation of coping and psychosocial health among adolescents with a variety of health conditions and highlights the value of moving toward this approach.

Conclusion

This study has provided insight into the combinations of coping strategies that may be common among adolescents with AIS, and demonstrated that the use of a range of coping strategies, with lower use of disengagement strategies, is associated with better HRQOL outcomes, in comparison to those who use other coping combinations. Notably, only 38% of participants in this sample were grouped in the "highly engaged" Cluster 1 with higher self-reported HRQOL. Adolescents with AIS may benefit from psychoeducation about different coping strategies that could be adaptive or non-adaptive for them. However, while the findings from this study have advanced our understanding of coping among patients living with AIS, it must be acknowledged that research on this topic remains relatively limited and further research as proposed above is required. Building on the present study, researchers who study HRQOL in AIS may be encouraged to continue the investigation of coping processes among other AIS samples. A body of findings will be key in informing the development of clinical interventions such as psychoeducational support programmes with the goal of promoting improved HRQOL among this patient group.

Chapter 6

A Systematic Review of Parents' Experiences of Adolescent Idiopathic Scoliosis: Information Needs, Treatment Concerns, and Psychological Wellbeing

Overview

This chapter presents a systematic literature review, the first of two studies pertaining to parents of children with adolescent idiopathic scoliosis (AIS). The aim of this review was to provide the first critical evaluation and synthesis of research studies examining the experiences of parents in the context of their child's AIS. The review was conducted in line with PRISMA guidelines and the final synthesis included a total of 18 eligible studies. This chapter begins with a brief background to the purpose of the review, followed by a detailed account of the search strategy and review process. Next, the included studies are narratively synthesized under three categories, namely "information needs," "treatment concerns," and "psychological wellbeing." The literature is then discussed in relation to practical implications and future research. Taking the same format, this study has been published in *Patient Education and Counseling* (Appendix B).

6.1. Introduction

Examining parental experiences is recognised as a key step to informing paediatric care, in terms of their involvement in, and adaptation to paediatric health conditions, and understanding their interactions with the health care system (Brown et al., 2002). This approach is in line with the patient and family-centered care perspective, which acknowledges that parents are personally affected when their child has a medical condition and recognises their key role in the patient's life and medical care (Eichner et al., 2012; Alderfer, 2017). This holistic approach emphasises important elements of paediatric care such as information communication between provider and the patient/family, and the provision of social-emotional support to families as well as patients to meet families' needs and even improve patient outcomes (Gallo et al., 2016, Alderfer, 2017). To inform these aspects of care in the context of AIS, an understanding of parents' experiences and perspectives is required.

Paediatric health conditions can introduce additional challenges to the parenting role, related to the emotional impact of the diagnosis, integrating the child's needs into family life, and parents' primary role in managing healthcare treatment (Pinquart, 2018; Smith et al., 2013; Golics et al., 2013). Parents of children with AIS are faced with specific stressors including unpredictable progression of the spinal curve and the possibility of invasive spinal surgery. Parental reactions vary, and while parents of children with health complications often cope well, some research reports increased levels of stress, depression, and adverse health compared to parents of non-affected children (Pinquart, 2018; 2019; Miodrag et al., 2015). It is also acknowledged that the way parents respond to their child's condition has potential to promote or confound the child's health outcomes (Kazak et al., 2017), therefore, supporting them in their parenting role is a primary concern for both their child's and their own wellbeing.

Given the growing recognition of the importance of understanding parental experiences in relation to paediatric health conditions (Kratz et al., 2009), systematic literature reviews relevant to parents of children with various health conditions are increasingly evident (e.g., Smith et al., 2013; Tong et al., 2008; Gabriel et al., 2018; Gates et al., 2019). However, to our knowledge no review specific to parents of children diagnosed with AIS is available. A systematic review is timely to provide a clear picture of existing knowledge on this topic, providing implications for practice and guidance for future research (Moynihan, 2004; Moher et al., 2009; Hong & Pluye, 2019).

6.1.1. The Present Study

Understanding parental experiences is recognised as an important topic in the field of paediatric healthcare in order to support parents in their role and promote provision of patient and family-centered care. The current study was therefore undertaken to provide the first systematic review focused on parents' experiences of their child's AIS. As such, this study sought to comprehensively capture the extent of available research focusing on parents, in terms of the impact that AIS can have on the parents of those affected, their key concerns, stressors, and associated needs. By synthesizing the body of evidence, this knowledge will assist health professionals in supporting families and guide future research on this topic.

Aim and Objective

The aim of this study was to examine what is known about parents' experiences and needs related to their child's AIS. The specific objective of this study was:

(i) To systematically review and synthesize previous research that has studied the experiences of parents in relation to their child's AIS.

6.2. Method

This systematic review was conducted in line with the Preferred Reporting Items for Systematic Reviews and Meta-analyses guidelines (Moher et al., 2009) and included research studies that reported on parents' experiences of their child's AIS. A mixed studies systematic review was conducted, whereby quantitative, qualitative, and mixed methods study designs were eligible for inclusion (Pluye & Hong, 2014). This mixed studies approach is increasingly used in health-related research and was adopted for this study in order to comprehensively capture the available research on this topic (Pluye & Hong, 2014; 2018).

6.2.1. Search Strategy

A systematic search, developed in consultation with a research librarian, was performed on the MEDLINE, CINAHL Plus, PsycINFO, SCOPUS, and EMBASE databases. These databases were selected as they provided comprehensive coverage of the health and social sciences literature. The search was originally conducted in October 2018 and updated in July 2019 and finally in October 2020. The search strategy combined key terms 'parent', 'scoliosis', 'child or adolescent', 'experience', and their variations to identify relevant studies published in the English language (see Appendix K for the search strategy). The search dates were from January 2000 to September 2020 in order to reflect current paediatric healthcare. Additionally, the reference lists of included studies were manually searched. Furthermore, in line with guidance for improving completeness of reviews (Mahood et al., 2014; Paez, 2017), ProQuest Digital Dissertations, OpenGrey.eu, Google, and Google Scholar were searched for further eligible studies and grey literature. For both Google and Google Scholar the first 15 pages of results were screened to review the most relevant records and maintain feasibility.

The search results were imported into Covidence Systematic Review software (www.covidence.org), where any duplicates were removed. A total of 302 unique records were identified through the database searches, and an additional 5 relevant records were identified by handsearching reference lists and by searching the other grey literature sources. Using the eligibility criteria detailed in Table 6.1, titles and abstracts of 307 studies were screened by two independent reviewers and 49 were chosen for full-text examination. There was strong inter-rater reliability during screening (kappa = 0.93). Following full-text examination by the lead researcher, 31 studies were excluded. Main reasons for exclusion were (1) children had conditions other than idiopathic scoliosis or, (2) study focus was not on parents' personal experiences, did not collect data from parents, or was predominantly focused on the child. A total of 18 studies were eligible for inclusion.

Table 6.1

Inclusion and Exclusion Criteria for Study Selection

Characteristic	Inclusion criteria	Exclusion criteria
Study Design	Primary studies (Quantitative, qualitative or mixed-method designs)	Not applicable
Population	Parents who have a child with adolescent idiopathic scoliosis	Parents of children with scoliosis of a non-idiopathic cause or other significant co-morbid medical conditions
Outcome	Studies investigating an aspect of parents' personal experiences over the course of their child's scoliosis	Family history or genetics studies

6.2.2. Quality Appraisal

The Mixed Methods Appraisal Tool (MMAT, 2018 version, Hong et al., 2018), developed to allow concurrent assessment of qualitative, quantitative, and mixed-method studies, was used. The MMAT criteria reflect quality of reporting within studies as well as quality of study design. Included studies were assessed against two universal screening criteria ("Are there clear research questions?" and "Do the collected data allow the research questions to be addressed?"), and five subsequent criteria specific to the research design. Specific appraisal

criteria related to issues such as selection bias, measurement validity, and integrity of results. This assessment was conducted by the lead researcher. The MMAT was initially developed in 2006 to address the need for a comprehensive appraisal tool for mixed studies reviews and has been updated several times in line with critical appraisal best practices (Hong et al., 2018). The tool demonstrates good interrater reliability (Pace et al., 2012) and content validity (Hong & Pluye, 2019). No studies were excluded based on this assessment.

6.2.3. Data Synthesis

For all included studies, details of the research objectives, study context, sample, research design, methodologies, and main findings were extracted from the original papers and tabulated. As both quantitative and qualitative research designs were included and studies explored a broad range of parental experiences, a narrative synthesis was chosen as an appropriate method of synthesising study findings. Guidance on conducting a narrative synthesis in systematic reviews developed by Popay et al. (2006) informed the process of identifying patterns across the literature, clustering studies into overarching themes, and producing a synthesis that “tells the story” of the included studies.

6.3. Results

A total of 18 studies were included. A PRISMA flow chart (Moher et al., 2009) illustrating the review process is shown in Figure 6.1.

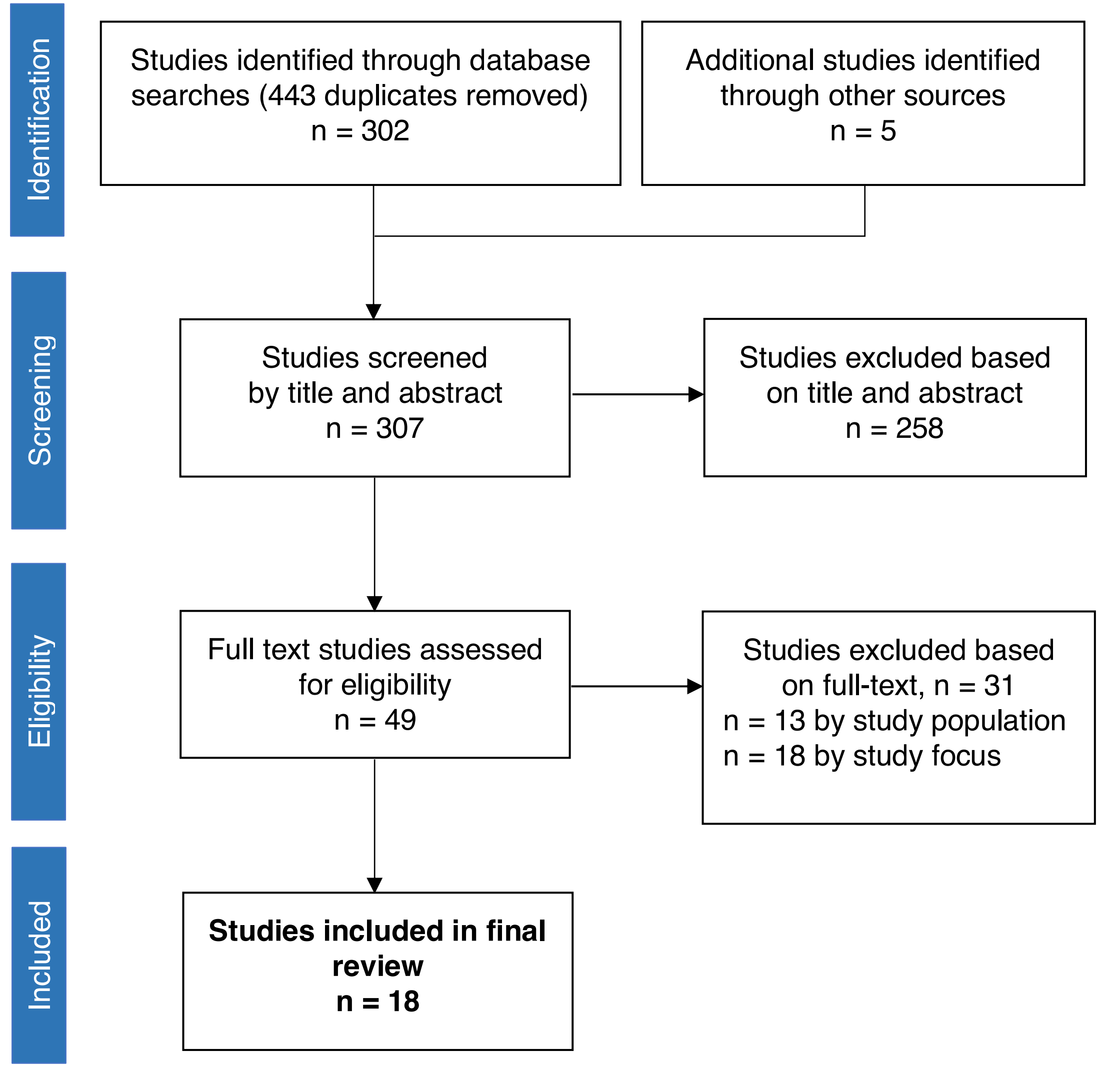
6.3.1. Study Characteristics

Studies were categorised as qualitative (*n* = 2), mixed methods (*n* = 1), and quantitative (n = 15). A variety of quantitative research designs were represented, including non-randomized controlled studies, cross-sectional surveys, and case series. Most studies recruited parents from clinical settings, while three collected data via support groups (Bull & Grogan, 2010; Schwieger et al., 2016b, Flynn et al., 2007). Of the twelve studies reporting parent gender, mothers made up an average of 85% of the samples. Studies spanned various stages of scoliosis treatment, but there was a predominant focus on parents whose children were undergoing surgical intervention. The mean age of patients reported in studies ranged from 11.9 to 17.3 years. Two studies included a small number of early onset scoliosis cases in their analyses (Bull & Grogan 2010; Baker et al., 2012), however these studies were included as the

majority of the samples were AIS. Most of the included studies were reported in academic journals while two were presented in theses available on repositories (Flynn, 2007; Narayanan, 2008). A summary table describing each of the included studies is presented in Appendix L.

Figure 6.1

PRISMA Flow Chart Illustrating Study Selection



6.3.2. Study Quality

All studies met the initial MMAT screening criteria. Within their respective study design categories, two thirds of the included studies (n = 12) did not meet or reported unclear information in relation to at least one of the 5 criteria. As shown in the MMAT assessment

summary presented in Appendix M, six studies met 5 criteria, nine studies met 4 criteria, and three studies met 3 criteria. This indicated that in general, the studies demonstrated moderate to high quality in reporting and design, but that caution is required in interpretation of some results. The two qualitative and one mixed-methods studies demonstrated no major methodological or reporting bias concerns, whereas several issues arose within the quantitative studies. Six studies did not report basic demographics including the gender of their parent samples, making it difficult to generalise findings to mothers and fathers. Another issue was risk of non-response bias in five studies, whereby missing or incomplete data was problematic or not explained, while one of the two non-randomized controlled studies reported they did not account for potential confounding factors in their design. The MMAT assessment summary is presented in Appendix M.

6.3.3. Narrative Synthesis

The included studies were heterogenous with respect to design, research questions, and children's stage in treatment. Despite the variability observed, we identified three main study categories that were evident across the literature: "Information Needs" (*n* = 4), "Treatment Concerns" (*n* = 7), and "Psychological Wellbeing" (*n* = 7).

6.3.3.1. Information Needs

A number of studies investigated parents' scoliosis knowledge and associated behaviours as they sought to gather information about their child's condition, which is evidently a significant aspect of their experience. Khetani et al. (2008) developed a reliable and valid 'Scoliosis Knowledge Questionnaire' to investigate parents' treatment knowledge and showed that more than 70% of parents (*n* = 30) lacked information about topics such as curve progression and postsurgical issues. To access more information about scoliosis, many parents turn to the internet, as identified in an outpatient department survey by Baker et al. (2012) and a presurgical case series by Lysenko et al. (2016) where 58% and 91.2% of parents respectively, reported searching online for this purpose. While the majority of these parents found the internet at least somewhat helpful, almost a third of parents in one study indicated that sites were confusing and caused them greater feelings of anxiety (Baker et al., 2012).

Qualitative accounts support the findings of these quantitative studies, as parents reported that they lacked knowledge about scoliosis and searched online as their main source of information (Bull & Grogan, 2010). However, learning about the implications of scoliosis online seemed to cause further anxiety and distress, suggesting that caution is required if recommending e-health resources for parents. Lysenko et al. (2016) investigated the effectiveness of their purpose-built scoliosis website, which was informed by evidence and needs assessments (MacCulloch et al., 2009; MacCulloch et al., 2010). Parents' knowledge scores on the 'Scoliosis Knowledge Questionnaire' significantly improved after using the website, while they also reported a minor increase in negative attitudes towards their child's condition. The evidence-based, user driven website was an advantage in this study, however, risk of non-response bias and the lack of a control group for comparison limited the results.

As well as searching for informative websites, some parents seek information through online support networks. Information sought and shared in an online support forum was examined by Schwieger et al. (2016b) who reported that the most frequent topic discussed by parents was the causes and progression of scoliosis. Bracing and its' effectiveness was also frequently discussed, and parents exchanged significantly more information regarding doctors and hospitals compared to adolescents who participated in the discussion forum (Schwieger et al., 2016b).

6.3.3.2. Treatment Concerns

Seven studies explored parents' concerns about medical treatment for scoliosis, highlighting the considerable fears, stressors, and emotional consequences they experienced. Of these, one study by Donnelly et al. (2004) interviewed parents whose daughters were undergoing brace treatment. Parents felt a responsibility to choose bracing as a less invasive treatment than surgery, despite difficulties with brace wear compliance and conflicting evidence regarding its' effectiveness. They reported issues including the traumatic nature of brace fitting, conflict with their child over compliance, and difficulty finding clothing.

The remaining six studies focused on parents whose children were undergoing spinal surgery. Bridwell et al. (2000) and Narayanan (2008) had parents rank or score predetermined concerns and expectations, while Chan et al. (2017) and Lonner et al. (2020) asked parents to

list concerns or operative aspirations with an open-ended question and assign scores to each. Both Salisbury et al. (2007) and Bull & Grogan, (2010) interviewed parents about key stressors. Despite differing methods of data collection there were similarities across study findings. Overall, parents were found to be more concerned about the surgery than their children (Narayanan 2008; Chan et al., 2017). Specific surgical concerns that were common across studies were the possibility of complications, particularly neurologic injury, and the amount of pain their child would experience (Bull & Grogan, 2010; Bridwell et al., 2000; Chan et al., 2017; Salisbury et al., 2007; Narayanan, 2008). Another primary stressor described by parents was the pressure of their parenting role, including the responsibility of supporting their child before surgery (Salisbury et al., 2007), and their inability to protect their child in such an uncertain situation (Bull & Grogan, 2010). Parents were also concerned about the amount of correction that surgery could achieve and possible failure of the spinal fusion (Bridwell et al., 2000; Chan et al., 2017; Narayanan, 2008). Focusing on what parents hoped would be achieved by surgery, parents' top aspirations and expectations centred around preventing progression of the spinal curve and associated future health problems, improving pain and sleep, as well as improving appearance and self-esteem (Bridwell 2000; Lonner et al., 2020; Narayanan, 2008).

Postoperatively, concerns about fulfilling their parenting role continued, as parents felt helpless (Bull & Grogan 2010) and feared the responsibility of caring for their child (Salisbury et al., 2007). Some parents felt unprepared for the amount of pain their child experienced following surgery and identified this as a main stressor (Bull & Grogan, 2010; Salisbury et al., 2007), while parents' whose children had complications self-identified this as their main cause of stress (Salisbury et al., 2007). Some parents were concerned about the level of care their child received postoperatively (Salisbury et al., 2007) and it was suggested that parents' trust in the medical and nursing staff was key to them feeling supported (Bull & Grogan, 2010). They recalled living in a state of uncertainty and intense emotional upheaval when waiting for a surgery date (Bull & Grogan, 2010), and during their child's recovery (Salisbury et al., 2007).

6.3.3.3. Psychological Wellbeing

As well as concerns directly related to medical treatment, other stressors reported by parents include the shock of diagnosis, explaining scoliosis and/ or bracing to their child, treatment delays, lack of clarity about treatment options, worry about curve progression, and concerns about their child's quality of life at present and in future (Flynn, 2007). A recent study by Wang et al. (2019) demonstrated that parents of children who were undergoing treatment for idiopathic scoliosis had higher levels of mental health difficulties in comparison to a control sample. Specifically, parents in the scoliosis group were three times more likely to report moderate to severe depressive symptoms, and about four times more likely to experience moderate to severe generalized anxiety. Maternal depression scores were also found to be significantly higher than fathers (Wang et al., 2019).

Three studies measured parental anxiety at specific times throughout their child's scoliosis treatment (Hines et al., 2015; LaMontagne et al., 2001; 2003). Firstly, a controlled study by Hines et al. (2015) reported that parents whose children were referred to the orthopaedic clinic from a school scoliosis screening experienced increased state anxiety prior to their appointment and after confirmation of the diagnosis. For parents whose children were undergoing surgery, two case series by LaMontagne et al. (2001; 2003) showed that parents reported high levels of state anxiety at the preoperative clinic visit before their child's surgery, and while anxiety decreased two days following surgery, it was still classified as moderately high (LaMontagne et al., 2003). Two studies also reported that parental anxiety and depression were weakly and moderately correlated with their children's anxiety and depressive symptoms (LaMontagne 2001; Wang et al., 2019). One further case series (Kasai et al., 2006) measured personality characteristics, showing that parents' neurotic tendencies significantly reduced from the pre- to postoperative period, suggesting that mothers felt a strong sense of anxiety pre-surgery and a sense of relief afterwards.

An intervention study by Kwan et al. (2016) sought to alleviate parental anxiety during surgery, as the intraoperative waiting period is known to be a particularly anxious time for family members due to uncertainty surrounding surgical outcomes. Parents who received intraoperative text messages with surgical progress updates experienced significantly lower levels of anxiety during, and on the day following surgery, compared to a control group of

parents who received no messages. While the authors recognise some constraints (e.g., limited information is conveyed via text compared to face-to-face interaction), this represents a promising strategy for keeping parents informed and improving their surgical experience.

Three of the included studies explored parents' coping strategies for dealing with the stress of their child's scoliosis (Flynn, 2007) and at the time of their child's surgery (Salisbury et al., 2007; LaMontagne et al., 2003). In all studies, seeking social emotional support (e.g., from friends, family) was the coping strategy used most often by parents. After this, positively reappraising the situation or optimism, planful problem solving, and seeking expert advice (e.g., from doctors) were among the three most common strategies reported by parents across studies (Salisbury et al., 2007; LaMontagne et al., 2003; Flynn, 2007). At the time of surgery, it was also shown that seeking social support decreased postoperatively, while positively reappraising the situation increased (Salisbury et al., 2007; LaMontagne et al., 2003).

6.4. Discussion

This systematic review included eighteen studies that investigated parents' experiences when their child is diagnosed with AIS. The identified literature was synthesised into three meaningful themes which reflected the experiences of this parent group, including parents' information needs, their concerns about their child's treatment, and the impact on parents' psychological wellbeing. Despite the relatively limited number of studies contributing to each theme, the current review provides a knowledge base for these facets of the parental experience and provides opportunity to consider the implications of this knowledge base. Discussion of each theme follows.

Information Needs

Parents report having little or no knowledge about scoliosis before their child was diagnosed (Bull & Grogan, 2010). After diagnosis, research also suggests that some parents may lack certain knowledge about scoliosis, in particular about surgery (Khetani et al., 2008), which is important to address given the significant role they play in their child's surgical decision making. To assist with the identification of topics where further information is required, tools such as the Scoliosis Knowledge Questionnaire (Khetani et al., 2008; Lysenko et al., 2016)

could be used collaboratively with parents in practice if their child reaches the stage of surgical consideration. This questionnaire could also benefit from being cross-culturally validated and updated as advancements in scoliosis treatments occur (Beauchamp et al., 2019).

Accessing scoliosis information online appears to be a convenient strategy for meeting parents' information needs outside of the clinical setting. However, variation in the quality or presentation of information online may contribute to feelings of confusion and distress in parents (Bull & Grogan, 2010; Baker et al., 2012). A previous review of scoliosis-specific information websites indicated that overall, information quality was poor, with academic and physician provided sites shown to contain better quality information in comparison to social media sites (Nason et al., 2012). This highlights that where possible; healthcare professionals should direct parents and their children to appropriate, evidence-based resources. There are a number of reputable resources available, such as those associated with hospitals and spinal organisations (e.g., Scoliosis Research Society webpage; The Hospital for Sick Children scoliosis learning hub, American Academy of Orthopaedic Surgeons webpage). Given parents increasing use of e-health resources (Plantin & Daneback, 2009), further research evaluating effective online scoliosis information provision for parents is timely. Some level of distress may be unavoidable when viewing information resources, as topics such as potential complications are likely to cause concern to parents. However, equipped with the appropriate knowledge, parents would be in a better position to offer support to their child before surgery (Salisbury et al., 2007), which could contribute to less anxiety and stress, and a better operative experience for families.

Treatment Concerns

Studies exploring parents' treatment concerns related to their child's AIS have primarily focused on surgery. Understandably, parents indicate many concerns about surgical intervention, such as the possibility of surgical complications, their child's level of pain, and fulfilling their parenting role both pre- and postoperatively (Bull & Grogan, 2010; Bridwell et al., 2000; Chan et al., 2017; Salisbury et al., 2007; Narayanan, 2007). Preoperative counselling and preparation addressing these main areas of concern may help to improve the surgical experience for parents and their children (Chan et al., 2017). In addition to addressing

concerns, it may be beneficial to consider parent and patient priorities for surgical outcomes (e.g., cosmetic correction), as these may have implications for postoperative satisfaction.

We identified only one study that considered parents' experiences of their child's brace treatment (Donnelly et al., 2004). Parents reported considerably different issues compared to those expressed in relation to surgery, such as conflict with their child over brace wear compliance. Previous research has investigated factors related to non-compliance with brace wear, such as increasing age of the patient, setting (e.g., in school, during summer), and concerns about the appearance of the brace (Rahimi et al., 2019; Brigham & Armstrong, 2017; Hasler et al., 2010). These studies highlight the multifaceted issues that can surround brace compliance and future research could explore the role of healthcare providers in supporting parents throughout the management of their child's bracing treatment.

Psychological Wellbeing

Studies highlight heightened anxiety levels in parents at the time of their child's scoliosis diagnosis (Hines et al., 2015) and throughout surgical treatment (LaMontagne 2001; 2003; Kwan et al., 2016). Furthermore, parents of children receiving treatment for AIS were shown to have significantly higher levels of anxiety and depressive symptoms in comparison to parents of non-affected children, with mothers and parents of children with curves exceeding 50° more vulnerable to depressive symptoms (Wang et al., 2019). Importantly, it is often recognised in practice that parents' reactions to their child's scoliosis and medical treatment may influence how their child copes with the condition and in line with this, two studies in the current review reported correlations between parent and child anxiety and depressive symptoms (LaMontagne et al., 2001; Wang et al., 2019). In the clinical setting, it is important to gain insight into parental mental health to identify families that may benefit from increased support or intervention. Future research could seek to evaluate aspects of resilience which may serve as protective factors for parental wellbeing (Barlow & Ellard, 2006), as well as psychosocial support needs relevant to this parent group.

Positive reappraisal of the situation was a commonly used coping strategy among parents, particularly during their child's surgical recovery (Salisbury et al., 2007; LaMontagne et al., 2003; Flynn, 2007), highlighting their attempts to remain optimistic at this difficult time.

However, the most frequently reported coping strategy, seeking emotional support, highlights the importance of parents' social networks as they manage the impact of their child's AIS on their lives. Decreases in this strategy postoperatively may reflect parent's presence in hospital with their child, and consequently, removal from normal support systems (Salisbury et al., 2007; LaMontagne et al., 2003). Healthcare staff therefore play an important role in supporting parents during this postoperative period (Bull & Grogan, 2010).

Limitations and Future Directions

In terms of the review process, appropriate steps were taken to minimise the risk of reporting bias (Liberati et al., 2009). These included the rigorous search strategy, strong inter-rater reliability at screening, and the inclusion of all study designs to comprehensively cover the research topic (Pluye & Hong, 2014). However, the search may have been limited by restriction to English-language studies. An important issue identified by the quality appraisal of included studies was inconsistent reporting of demographic information for parent samples. Future studies should endeavour to adequately describe their parent samples to contextualise the research and allow informed interpretation of generalisability. Of the studies that included information on parent gender (66.7% of studies), the majority were mothers, which is in line with previous reports that fathers are typically underrepresented in research concerning paediatric health conditions (Phares et al., 2005; Swallow et al., 2011). As a result, the review likely reflects mothers' experiences primarily and highlights the need to encourage future participation of fathers in AIS research. As well as this, the parents were predominantly parents of females with AIS, however given that AIS is more prevalent among females this is not unusual. Furthermore, the studies included in this review varied considerably in terms of design and results because of the broad nature of our inclusion criteria. Therefore, the focus of this review was on a narrative synthesis of the body of research, rather than any form of meta-analysis. However, this approach was appropriate given that there has been a limited number of studies thus far pertaining to parents' experiences of AIS.

It is important to acknowledge that the reviewed literature predominantly focused on the operative treatment of AIS and parents' needs, concerns, and anxieties throughout the immediate perioperative period. Further research is required to extend current knowledge

by exploring the experience of both mothers and fathers throughout all stages of scoliosis progression and treatment, in particular the presurgical stage of treatment. Future research should include a focus on the broader psychological and social implications of their child's condition, as well as their perspectives on improving the delivery of family-centered care. As also suggested by Wang et al., (2019), further assessment of parents' psychosocial support needs relevant to AIS is required. Building on the knowledge presented in this systematic review, the qualitative interview study presented in the following Chapter 7 further explored aspects of parents' psychosocial wellbeing and support needs throughout the treatment of their child's AIS prior to surgery.

Conclusion

This systematic literature review synthesised available evidence on the information needs, treatment concerns, and psychological wellbeing of parents who were accessing medical care for their child's AIS. Parents face challenges such as accepting the diagnosis of scoliosis, acquiring appropriate knowledge to participate in healthcare decisions, and coping with potentially invasive medical treatments for their child. Navigating their children's spinal surgery is a stressful and anxiety provoking experience pre- and postoperatively for parents, who may need additional support at this time to assist them in their parenting role. In collaboration with healthcare providers, parents play an important role in managing the treatment of their child's AIS and supporting their child throughout this potentially difficult time. The findings of this review demonstrate how strategies such as directing parents to appropriate resources about scoliosis and preoperative counselling and education may improve parents' experiences. This review also emphasizes the importance of prioritising a patient and family-centered approach in healthcare provision. While caring for a child with AIS, healthcare professionals can be encouraged to communicate and collaborate with parents and to consider how they can be best supported in their parenting role throughout their child's treatment.

Chapter 7

Parental Experiences of Adolescent Idiopathic Scoliosis From Diagnosis to Pre-Surgical Preparation: A Qualitative Study

Chapter Overview

This chapter details the qualitative interview study conducted with parents of children with adolescent idiopathic scoliosis (AIS). This study addressed the need for a better understanding of the wider psychosocial implications of AIS for parents throughout their child's treatment, which was highlighted in the previous systematic review in Chapter 6. Semistructured interviews were conducted with 20 parents whose children were in the presurgical stage of treatment for AIS and data were analysed using reflexive thematic analysis. To begin, the rationale and methodology for this study is detailed. The results of the thematic analysis are then narratively presented, followed by a discussion of the research findings and consideration of implications for practice.

7.1. Introduction

As detailed in Chapter 6, from the time of the AIS diagnosis, parents are required to adapt to the consequences of their child's often progressive spinal condition and research indicates that their own mental health can suffer throughout this time (Wang et al., 2019). Parents adopt new roles related to their child's care such as making important and life-long treatment-related decisions (Essex et al., 2021). For mild to moderate cases of AIS, treatment can include conservative options such as orthotic bracing and physiotherapy exercise programmes which aim to halt or reduce progression of the condition. However, for adolescents whose curvatures progress severely, they and their parents may have to contend with intervention by spinal fusion surgery (Beauchamp et al., 2019).

Although operative treatment is available to improve severe AIS curvatures and associated symptoms, patients and their parents accessing this option will often be required to manage living with the progressive spinal condition for a number of years until the curvature reaches the severity threshold for surgical intervention (i.e. a Cobb angle exceeding 45 - 50°) (Altaf et

al., 2013). Furthermore, wait times for elective orthopaedic procedures evident in many countries can result in a prolonged presurgical period for many adolescents and their parents. Adolescents may be waiting over a year from the point that surgical intervention is necessary and they can experience deterioration of their condition as they wait for a surgery date (Ahn et al., 2011; Tarrant et al., 2016). Adolescents with progressive AIS and their parents attend routine hospital appointments approximately every 6 months so that the progression of the spinal curvature can be monitored (Altaf et al., 2013). Therefore, within the clinical setting there is opportunity to provide appropriate support to both adolescents with AIS and their parents during this time from diagnosis to presurgical preparation.

Thus far, limited literature has explored the experiences of parents of children with AIS and this available literature has been detailed comprehensively in the systematic review presented in Chapter 6. This chapter synthesized key findings including the importance of appropriate information provision, as many parents seek to improve their knowledge by searching online where information quality is variable (Baker et al., 2012). Aside from research examining information needs, other research on parents' experiences of AIS has focused predominantly on the immediate surgical period. Prior to surgery, common concerns that have been identified among parents include the possibility of surgical complications and the amount of pain their child will experience (Chan et al., 2017). Bull & Grogan (2010) qualitatively explored the experiences of parents' whose children had previously undergone surgery for scoliosis. Findings indicated that parents struggled with a lack of expertise in spinal care and felt as though their parental role of 'protector' was threatened throughout the perioperative period. The importance of parents' trust in the health professionals caring for their child throughout the surgery and recovery was also highlighted (Bull & Grogan, 2010). Research examining mental health indicates high levels of self-reported anxiety among parents in the immediate pre- and postsurgical periods of their child's spinal fusion surgery (LaMontagne et al., 2003).

Previous research has mostly focused on parents' experiences in the perioperative period, however, this is just one aspect of AIS care and less is known about the experience of patients and their parents prior to undergoing surgery. No previous study has specifically examined the experiences of parents throughout the period from diagnosis to pre-surgical preparation,

and less is known about supporting parents during this stage of care. One study by Lauder et al. (2018) explored the experience of mothers who cared for children with early onset scoliosis (EOS) across the entire trajectory of the condition. Findings demonstrated the difficulties parents contended with including physical exhaustion from their child's care needs, financial burden, and constant emotional turmoil associated with the deterioration of their child's health over time. However, these experiences are not necessarily transferable to parents of children with AIS, as the conditions differ in timing of onset and prognosis. EOS is a very rare form of scoliosis present in infants, associated with high rates of cardiovascular and neurological complications and premature death (Lauder et al., 2018). On the other hand, AIS is detected around the onset of puberty and can progress over adolescence, leading to some functional and psychosocial issues that can be improved with treatment (Weinstein et al., 2008; Beauchamp et al., 2019). Notably, AIS is one of the most common spinal conditions seen in paediatric orthopaedics, and accounts for over 90% of idiopathic scoliosis cases (Konieczny et al., 2013). Similar to this previous work on EOS, more comprehensive research exploring parents' experiences of AIS throughout the presurgical period is needed. Insight into the experiences and perspectives of these parents represents a valuable contribution to the paediatric literature by furthering current knowledge on the impact that AIS can have on parents and informing delivery of clinical care.

7.1.1. The Present Study

Parents play an important role in their child's healthcare following a diagnosis of AIS. While surgical intervention is available to treat severe curvatures, the time period from diagnosis to presurgical preparation is challenging. During this time adolescents and their parents are dealing with prognosis uncertainty and possible psychosocial difficulties, but this is also a time when families are linked in with the healthcare system and can access support. Limited previous research with parents has tended to focus on the experience of their child undergoing surgery and therefore findings are specific to the immediate perioperative period. In particular, parental experiences across the trajectory of AIS, including their personal experiences and needs, the impact on family life, and their interactions with the healthcare system have received limited attention. To inform the support of parents throughout their child's treatment for AIS and improvement of patient and family-centered care, a more in-

depth understanding of parents' experiences from the time of their child's AIS diagnosis to the time of pre-surgical preparation is required.

Aim and Objective

The aim of this study was to understand parents' experiences across the trajectory of their child's AIS prior to surgical intervention. The objective was:

(i) To explore and describe the experiences of parents in relation to their child's AIS from diagnosis to presurgical preparation.

7.2. Method

A cross-sectional, qualitative study utilising semistructured interviews was conducted to achieve the research aim. Qualitative research methods are suited to describing individual experiences in the context of paediatric healthcare and are valuable in creating knowledge to inform the improvement of clinical care (Alderfer & Sood, 2016). The participant recruitment and data collection for this study was carried out concurrently with the adolescent qualitative study presented in Chapter Three. The parent participants described in this study are parents of the adolescent participants described in Chapter 3. Recruitment and data collection for this study was conducted alongside recruitment and data collection described in Chapter 3.

7.2.1. Participants and Recruitment Procedures

7.2.1.1. Eligibility Criteria

Parents and guardians (henceforth referred to as 'parents' as all participants identified as such) were eligible to participate if they had a child aged 12 – 18 years with a diagnosis of AIS, who was in the presurgical stage of treatment.

7.2.1.2. Recruitment

Parents were recruited to participate in the study through the orthopaedic department of an Irish children's hospital between October 2018 and May 2019. Parents meeting the inclusion criteria were informed of the study by a spinal nurse specialist while attending a routine outpatient appointment with their child. The nurse specialist explained that the purpose of the study was to understand the experiences of adolescents living with AIS and their parents, and that participation would involve scheduling interviews with the researcher. If they were

interested in the study or wished to find out more information, parents agreed for their contact details (phone number and/ or email) to be passed onto the researcher (GM). The researcher subsequently followed up with each parent and sent them detailed study information sheets if they had not previously received these from the spinal nurse specialist on the day they were informed about the study. Of 16 families (i.e. the adolescent with AIS and their parent(s)) in contact with the researcher throughout recruitment, two did not participate due to unsuitable timing of the study or undergoing surgery before an interview was arranged. In order to be as inclusive as possible, parents were reminded that they could participate without their child (or vice versa) if they wished to do so but this circumstance did not occur. To promote participation of both mothers and fathers, parents were reminded that both parents were welcome to participate in the study. All participants provided informed signed consent prior to participating in the research.

7.2.1.3. Participants

The final sample consisted of 20 parents (13 mothers and 7 fathers) which included six parental dyads whereby both mother and father from the same family participated, seven mothers who took part alone, and one father who participated alone. Most had a daughter with AIS (n = 15), while a smaller number had a son with AIS (n = 5), aged between 12 – 17 years. Parents were aged between 39 to 53 years (M = 44.9, SD = 4.4) and identified as White Irish (n = 19) or White English (n = 1). The total number of children in each family ranged from 2 to 4 (M = 2.9). Most parents currently worked outside of the home in various employment settings including healthcare, education, and business (n = 18, 90%). Their children were all diagnosed with moderate to severe AIS, with major Cobb angles between 46 to 100 degrees (M = 68°, SD = 14.8) as assessed by an orthopaedic surgeon using their most recent radiographic images to time of interview. The average length of time since their child's diagnosis was 12.7 months (range 3 – 26 months). At time of interview, two parents had a surgery date confirmed for their child, and the remainder were awaiting a surgical date or anticipated their child's surgery to be scheduled in future. Two parents had children who wore an orthotic brace as part of their treatment plan. Participant characteristics for parents are summarised in Table 7.1.

Table 7.1

Participant Characteristics for Qualitative Study with Parents (N = 20)

Variable		
	Range	*M*
Parent Age (years)	39 – 53	44.9
Adolescent Curve Size	46 - 100°	68°
	n	*%*
Parent Gender		
Male	7	35
Female	13	65
Home Setting		
Urban	8	40
Rural	12	60
Length of AIS Diagnosis		
<1 year	8	40
1-2 years	11	55
2-3 years	1	5
Adolescent Treatment Details		
Bracing	3	15
Awaiting surgery	13	65
Surgery scheduled	4	20

Note: All adolescents were considered surgical candidates based on the measurement of their spinal curve and potential for future curve progression at time of interview.

7.2.2. Data Collection

Prior to study commencement, ethical approval was obtained from the hospital's Medical Research Ethics Committee (GEN/546/17) and the University College Dublin Human Research Ethics Committee (HS-17-05-Motyer-Fitzgerald). Data were collected with semistructured, face-to-face interviews between October 2018 and June 2019. On the day of interview, participants completed a brief demographic form which collected information including age, employment, and details of their child's AIS diagnosis. Interviews were conducted by the same researcher (GM) with all participants, either in the participants' homes (*n* = 16), or in the outpatient department of the children's hospital (*n* = 4). The location of the interview was dependent on participant preference and a choice was provided in order to facilitate ease of participation. Data was collected on the same day that the adolescent interviews were conducted, and parents were interviewed subsequent to the adolescent interviews. Where

two parents from the same family were taking part, these parents chose to participate in the interview together.

Data collection was guided by an interview schedule that consisted of open-ended questions and related prompts designed to explore parents' views and perspectives since the time of their child's AIS diagnosis. Development of the interview schedule was informed by previous qualitative research on parents' experiences of a range of paediatric health conditions (Golics et al., 2013) and early onset scoliosis (Lauder et al., 2018), as well as qualitative research guidance (Braun & Clarke, 2013; Robson & McCartan, 2016). A paediatric orthopaedic consultant and clinical psychologist working at the hospital site were consulted regarding development of the interview content to further ensure clinical relevance and appropriateness for this study. The semistructured schedule ensured a consistent basis for each interview and also allowed for flexibility. The interview schedule is presented in Appendix C. Interview duration ranged from 15 to 60 minutes ($M_{minutes}$ = 35). Interviews where both parents took part tended to be longer in length as both parents were encouraged to contribute their perspectives. The researcher took field notes following each interview to aid interpretation of the recordings. Audio recordings were transcribed verbatim and data were anonymised at the point of transcription.

7.2.3. Data Analysis

Data were analysed in line with the reflexive thematic analysis (TA) guidelines of Braun & Clarke (2006; 2019). Reflexive TA involves a structured process of analysing qualitative data to understand patterns of meaning across the dataset and construct themes (Braun et al., 2018). This method of analysis was chosen as it is particularly effective in presenting a combination of analytic narrative and data extracts to convey subjective experiences across a group of participants. Similarly to the study with adolescents described in Chapter Three, the qualitative approach was underpinned by a critical realist position. This position recognises that knowledge is accessible through the lens of participants' subjective experiences (Braun & Clarke, 2013). Taking this approach, the objective was to understand parents' experiences throughout their child's treatment for scoliosis from their personal perspective, acknowledging that reality is framed by human interpretation.

The process of analysis involved familiarisation with the dataset through transcription and re-reading. Preliminary codes were then applied to identify data extracts which contained information relevant for addressing the research aims. Codes were refined through an iterative process, and grouped together to form broader units of meaning known as themes and subthemes where appropriate. This process of thematic analysis and steps to promote credibility and rigour of the analysis matched that which is detailed in Chapter Three Section 3.2.3. NVivo 12 qualitative research software was used to assist with data organisation and management. To promote a thorough analysis, the same process described in Chapter Three was employed, whereby the primary researcher (GM) and supervisor (AF) were involved with independently coding 25% of the dataset (i.e. data for five parents). This process of double coding the data followed by collaborative discussion throughout the analysis was conducted in order to promote a rigorous interpretation of the dataset (Braun & Clarke, 2019).

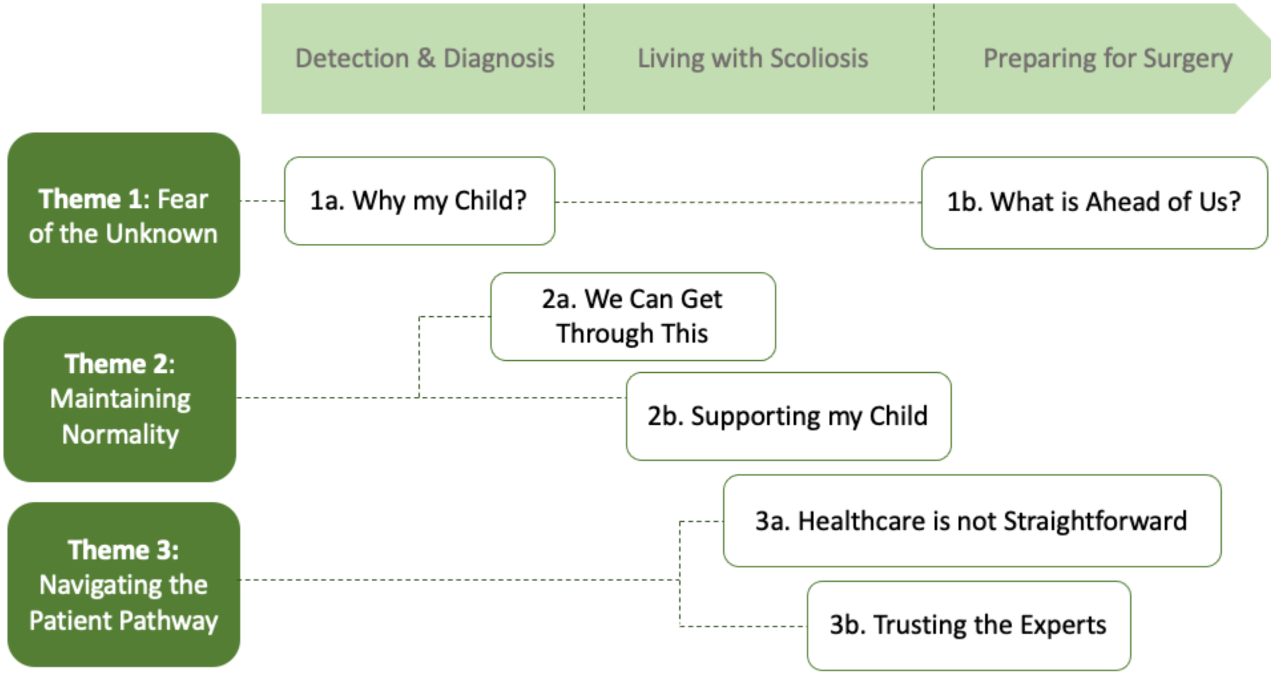
7.3. Results

Three key themes were generated from the data which captured pertinent aspects of parents' experiences from the time of their child's scoliosis diagnosis to the period of surgical preparation. The three themes titled *"Fear of the Unknown," "Maintaining Normality,"* and *"Navigating the Patient Pathway,"* each contained two subthemes. In Figure 7.1., themes and subthemes are presented across three phases of parents' trajectory with their child's presurgical AIS that were evident in parents' narratives including Detection and Diagnosis, Living with Scoliosis, and Preparing for Surgery.

Themes and subthemes are not intended to be mutually exclusive in terms of their temporal occurrence, and overlap is evident in parental experiences. For example, if the scoliosis is identified as highly progressive or severe from the outset, some parents may have been informed of the likelihood of surgery soon after the diagnosis, meaning that some parents may be coming to terms with the diagnosis, managing life with scoliosis, and preparing for surgery within a short time frame. A detailed narrative description of each theme follows.

Figure 7.1

Key Themes and Associated Subthemes Across Three Phases of Parents' Trajectory with Their Child's Presurgical AIS



7.3.1. Thematic Findings

7.3.1.1. Theme 1: Fear of the Unknown

Central to this theme was the apprehension and uncertainties that parents faced in relation to their child's scoliosis and prognosis. Being fearful of the unknown related to the diagnosis of scoliosis in their child and what this would mean for them, as described by the subtheme *"Why My Child?"* and when considering the prospect of surgical intervention and their child's future, as captured by the subtheme *"What is Ahead of Us?"*

Subtheme: Why My Child? After receiving the diagnosis of AIS, many parents went through an emotional time period when they questioned how this had happened to their child; *"the next day I kept thinking oh god why her… I don't want her to have this" (P20, Mother to 12 year old daughter).* Although AIS is idiopathic and occurs with no known cause, parents had never expected that a condition like scoliosis could develop in their child, and the discovery was associated with shock and sadness: *"It's devastating, you just think that your children are normal and then all of a sudden, you know, they're not normal and… you did*

everything with them […] you just never think that that is going to happen. I know you have to think that some things could happen but I thought that you were born with it and not that you develop it" (P8, Mother to 16 year old daughter).

For some parents, the discovery of their child's scoliosis was associated with feelings of guilt and a disbelief that they had not noticed the signs sooner: *"there's probably a bit of guilt as well of not seeing it earlier […] it was only when we actually looked, when we looked, you're kind of thinking to yourself oh my god, I don't believe that I never saw that…" (P7, Father to 16 year old daughter).* Some felt that earlier detection of their child's scoliosis could have been helpful (i.e. to try and prevent progression with conservative bracing) and questioned why school scoliosis screening was no longer a routine procedure. As articulated by one mother, there was limited awareness of the early signs of scoliosis: *"I've heard of scoliosis, you see it randomly on news things or whatever, eh but… never thought that it would affect us and I didn't know really what to look for until it got really prominent" (P10, Mother to 15 year old daughter).*

Parents were fearful of what the diagnosis of AIS would mean for them and their child. This related to the uncertainty of their child's prognosis and how severe their spinal curvature would become: *"we were shocked, terrified, upset, wondering you know how bad was it going to go and not wanting him to have to go through the surgery" (P1, Mother to 16 year old son).* It appeared that some parents had to quickly figure things out with regard to their child's healthcare, and not knowing what it entailed was stressful: *"I was upset because I didn't know what was ahead of me, I didn't know what it involved, but just as we went along, I started doing all the appointments myself…" (P16, Mother to 14 year old daughter).* A lack of information could contribute to uncertainty and fear following the diagnosis of scoliosis at the primary care level, and while waiting for a specialist orthopaedic appointment. Media coverage related to long wait times for scoliosis treatment also created heightened fear and concerns about accessing care: *"obviously you think of all the worst cases don't you, and then all the bad press you know where it's highlighted and.. everybody tags you [online] in everything to do with scoliosis" (P20, Mother to 12 year old daughter).* Media stories were fear-provoking, and this caused a few parents to be particularly cautious when it came to sourcing information.

Subtheme: What is Ahead of Us? Understandably, the prospect of their child undergoing spinal fusion surgery was a source of worry for parents, as they considered the seriousness of this treatment option and the possibility of surgical complications such as paralysis or persistent pain. Often, parents acknowledged that risks were low, but some were particularly impacted by fear of the upcoming surgery, as articulated by one mother: *"…wheeling her down to the surgery, I don't think I'll be able to, I think I'll break down. I'm very frightened of it but emm, I do know it's, like the doctor explained everything to me, and just the thought of it when you think of spine, like that's your, that's the centre link to your brain, like you know so I'm just having all these nightmares and stuff, about it" (P12, Mother to 15-year-old daughter).* For some parents, the element of uncertainty surrounding the surgery and related outcomes weighed on their minds, as they felt the decision to proceed with surgery was their parental responsibility: *"… what I'm most afraid of as the parent is… that the decision that you make for the child is the wrong decision" (P13, Father to 14 year old son).* Despite concerns about surgery, parents tended to accept surgical intervention as the only way of preventing their child's condition from becoming progressively worse and expected the surgery to be successful.

Some parents were also concerned about postoperative recovery when they would bring their child home from hospital, with some not knowing what to expect and feeling that they may be unprepared for this period: *"we're even worried for bringing her home and looking after her at home… you know after the surgery and… will we be able to manage?" (P6, Mother to 14-year-old daughter).* Specific concerns included uncertainty regarding how much time would be needed away from work to care for their child, and who to contact for support if something went wrong in the months following surgery. A few parents also questioned whether their child understood the seriousness of undergoing spinal surgery and feared how they would cope, as highlighted by a mother who was concerned about postoperative pain: *"… I don't think she realizes quite how painful it's going to be, those few days…" (P18, Mother to 16-year-old daughter).*

For many parents, their uncertainties about the future extended beyond the immediate postsurgical period. They were concerned about their child's abilities as an adult with spinal fusion, and lacked information about this. Specifically they worried how their condition may

impact upon their job opportunities, how females may manage pregnancy, and whether their physical capabilities would be restricted. This was demonstrated in a mother's concern about her son's future choices: *"he is so outdoorsy, and he's kind of a hands-on kind of kid, he likes animals, he likes all of that. Will his options down the line in his career be limited by his physicality, by this? Will he be told 'well you can't do this type of work?' […] and likewise, if he was to go down the route of more the academic side, would that be actually not good for him because he wouldn't be able to sit for long periods… there's that kind of uncertainty in my head" (P14, Mother to 14 year old son).*

7.3.1.2. Theme 2: Maintaining Normality

This theme was underpinned by parents' efforts to adapt to the reality of their child's AIS and to maintain a level of normality for themselves and for their child. There were two main elements of this theme, firstly parents desire to get on with life amidst the challenges of AIS as described by *"We Can Get Through This,"* and how they supported their child during this time described under *"Supporting my Child."*

Subtheme: We Can Get Through This. Adjusting to their child's AIS and managing their healthcare had a variable impact on parent's work life, financial implications, and a negative impact on their own health and wellbeing. For example, some parents lived a number of hours away from the hospital and taking days off work for appointments could be problematic. Parents also anticipated they would require extended leave from work when their child would be recovering from surgery at home. However, many parents felt that their workplaces were supportive and that their time off was manageable, and emphasized that their child was their priority. For some parents, there was a financial strain, particularly in relation to accessing private scans and appointments. Furthermore, some parents felt that their own health, both emotionally and physically, had suffered as a result of their child's AIS. The stress of the situation could lead to poor sleep and exacerbation of their own existing health problems in a small number of cases: *"Probably with the stress of it I've developed a terrible thyroid problem" (P16, Mother to 14 year old daughter).*

Many parents identified that following the distress of the diagnosis and despite these challenges, they had accepted the situation and decided not to dwell on it in an effort to

maintain normality for their family and stay positive. Even though some parents highlighted that their child's scoliosis was a major emotional burden and was always in the back of their mind, it was possible to continue on with their lives and function normally day to day. As described by one mother: *"you kind of get to accept it in your head, not totally but accept it in a way that you just kind of have to get on with your life and hope that when the operation is done that she gets better" (P8, Mother to 16 year old daughter).* For some parents, feeling that their child was coping well with their scoliosis was helpful to them, *"when she's relaxed about it, I'm relaxed as well" (P6, Mother to 14 year old daughter).* Some parents also found reassurance in the knowledge that AIS could be treated, and counted themselves lucky compared to those they considered to be in a worse situation; *"my only saving grace I think with this is that it's something that can be fixed, emm and that just makes me feel yeah it's going to be ok" (P3, Mother to 13 year old daughter).* Some parents made comparisons to other families affected by scoliosis whose children were more physically affected, or compared their situation to more life-threatening paediatric conditions such as cancer.

Parents with partners reported seeking support from each other and spoke about working together to manage the impact of their child's condition. However, some parents referred to the differing approaches that they and their partner took in dealing with the diagnosis, *"so it's all tension on me whereas [partner's name] doesn't carry any because of his personality he's more relaxed, chilled out…" (P5, Mother to 12 year old daughter)*, with a couple of mothers feeling that they were more likely to worry. Other parents also received support from extended family members and friends with practicalities like school collections and offers of help during the postsurgical period, as well as being put in touch with other families affected by scoliosis.

Subtheme: Supporting my Child. Parents were concerned about their child and whether they were suffering as a result of their scoliosis. A few parents acknowledged that adolescence is already a difficult developmental period, so scoliosis is an additional challenge on top of this: *"Look everything with young girls now it's all image, it's all you know, how they look, how people look or perceive them, how I suppose they feel about themselves. She's had to tackle the braces, skin issues, like she's got all these things going on. I'm always going to be worried or concerned you know" (P2, Father to 13 year old daughter).*

For many parents, open communication within their family was important in relation to their child's AIS. Parents described not wanting to keep things from their child, perhaps feeling it was their parental responsibility to keep their child informed and 'give them the facts' particularly in relation to surgery. Although parents were keen to keep their child informed, it appeared that some parents were also putting on a brave face, and hiding the emotional impact the AIS was having on them when communicating with their child;*"if we (parents) were alone we'd discuss it and talk about it and shed our tears or do whatever we did, but outwardly to her we wanted her to remain positive" (P19, Father to 12 year old daughter).* Some parents also reassured their children by being optimistic about the surgery and that things would improve in future. They were attentive to their children's needs and tried to help their child in managing their scoliosis.

By supporting their child, parents were given back some sense of control in what was an uncertain time period as they monitored progression of their child's condition. This is demonstrated by one mother who encouraged her child to continue with sports to keep him fit and healthy: *"I am reassured by the stuff we have put in place for [child] and the fact that he does sports. So I suppose, yeah, I am not too traumatized because I think we're doing all that we can do and we're supporting him so I don't feel too bad you know emotionally, and I suppose the shock is still there and the not wanting him to have it, but we just keep going" (P1, Mother to 16 year old son).* Some parents became conscious of protecting their child's back and avoiding strain or pain. For example, they could be less inclined to have their child do household jobs in case their back would be aggravated.

A lot of parents had communicated with their child's school to seek support for their AIS. They made practical arrangements such as an extra set of books to avoid unnecessary carrying to and from school, time off for appointments, and in some cases made plans for keeping up with schoolwork while their child would be recovering from surgery at home. A few parents had negative experiences where they felt there was a lack of awareness of their child's scoliosis among teachers at school which could lead to misunderstandings. The lack of awareness among teachers may have resulted from the sometimes 'invisible' nature of scoliosis, as some curvatures may not be obvious. This was articulated by one mother whose daughter had been in trouble at school for wearing her school tracksuit rather than skirt, as

this was more comfortable with her brace: *"I think people looking from the outside they almost want to see a sick child to make any sort of exceptions" (P20, Mother to 12 year old girl).*

7.3.1.3. Theme 3: Navigating the Patient Pathway

The process of accessing healthcare and having to figure out how to navigate the health system while preparing for their child's surgery was an ongoing strain for parents. Importantly, managing their child's healthcare could be a major challenge as described by the subtheme *'Healthcare is not Straightforward.'* Support was available from their child's medical team and sometimes from parents who had gone through the same process as captured by the second subtheme *'Trusting the Experts.'*

Subtheme: Healthcare is Not Straightforward. Managing their child's healthcare was not straightforward for parents and this effortful process was certainly exacerbated by the issue of extended waiting times in paediatric orthopaedic care. A sense of responsibility for their child's healthcare was clear among parents: *"I'd push every button I could to get her seen and I have. I'm sure a lot of them were sick of me at times, ringing to try get these appointments […] I've been down a lot of roads, trying to move along, but anyway I was always fortunate enough to get them. Perseverance paid off" (P16, Mother to 14-year-old daughter).* There was a clear perception among parents that they needed to be proactive to make things happen for their child, in terms of ensuring timely appointments and paying for private scans. When accessing appointments, the need to *"battle for it"* was sometimes fuelled by a lack of confidence in the public healthcare system, as described by a father who endeavoured to access timely appointments for his daughter: *"you have to be willing, able, and also financially secure enough to go and kind of make things happen because like I could imagine you'd be pulling your hair out waiting for the public system to come through" (P2, Father to 13 year old daughter).*

While waiting for appointments or surgical dates, a fear of being forgotten about, or a feeling of being lost in the system was evident among some parents. This was often caused by a perceived lack of communication with health care providers. Parental anxiety in instances where their waiting period is prolonged could be reduced by receiving reassurance: "*I think*

even if they send.. even an email or a phone call to say you know your child isn't forgotten.. you're, you're still on the list, we're just checking in to see how is she getting on and if you know, you have any questions or anything like that" (P6, Mother to 14 year old daughter).

Negative experiences with waiting times were not consistent across all parents, as a few did feel 'lucky' when it came to managing their child's healthcare. This was in instances where parents felt that they had accessed care relatively quickly: *"it's been a bit of a whirlwind I suppose, is how I'd describe our experience of it, it's moving very fast.. and I imagine.. the majority of people we're going to meet will have the opposite of that experience" (P10, Mother to 15 year old daughter).* This parent compared their situation to stories of families who were worse off, or to negative news stories, and considered their experience as lucky rather than what should perhaps be the normal standard of care.

Subtheme: Trusting the Experts. Most parents highlighted the positive interactions they had with the spinal nurses and their treating consultant. Amid the often stressful process of managing their child's appointments and surgical preparation, these trusted members of the medical team were a key source of support and reassurance. This was described by a mother reflecting on her daughter's pre-operative assessment day: *"they made it as nice a day as it could be you know […] they've obviously thought it through, the whole journey of it, what can help, all the things that you need to know, there was nothing left, unturned, I felt they'd covered most of the things that you would've wanted to ask questions about, so there was a level of comfort over that" (P18, Mother to 16 year old daughter).* Parents had a lot of confidence in their child's consultant which appeared to reduce their concerns about surgery: *"You know even though you'd be afraid going in… but the team up there are absolutely fantastic… you know even [consultant], [they are] top class… I'd be very confident of [them] doing it, put it like that" (P16, Mother to 14 year old daughter)*.

Parents appreciated the ways in which doctors and nurses communicated with their children in the hospital setting. Specifically, involving their child in important discussions at appointments was valued: *"I think the guys like [doctor's name] has really explained it to her, talks to her around it which is brilliant, and everyone she's met talks directly to her and not to us, so I think that helps her almost cope with it" (P19, Father to 12 year old daughter).*

Transparency when discussing topics such as the surgery and recovery was also important to parents; *"he didn't sugar coat things" (P9, Father to 15 year old daughter)*. Feeling fully informed in this way may have reassured parents that they and their child were more prepared for undergoing surgery.

Another trusted source of expertise or support for many parents were other parents of children with AIS, often those who were further along in their treatment (i.e. post-surgery). Meeting other parents could be helpful in terms of practical advice and information about healthcare and what to expect from surgery. Other parents were also a source of moral support: "*it's the positivity I see in the parents, and that's helpful, and you kind of get a little bit of well if they can do it I can do it you know" (P1, Mother to 16 year old son).* However, for a small number of parents, hearing stories from parents or adults who had previously had surgery for scoliosis caused concern if their child had experienced ongoing pain post-surgery.

7.4. Discussion

This qualitative study explored parental experiences throughout the presurgical period of their child's treatment for AIS and developed three key themes evident from diagnosis, living with scoliosis, to preparing for surgery. The first theme, *"Fear of the Unknown,"* captured parents' apprehension and uncertainties in relation to their child's prognosis. This included apprehension around the unexpected diagnosis and what lay ahead of them, the daunting prospect of spinal surgery and recovery, and uncertainty regarding their child's future abilities. *"Maintaining Normality"* was underpinned by parents efforts to adapt to the reality of their child's AIS, including a desire not to dwell on the situation and a need to get on with life despite the challenges, while also supporting their child through this time. Finally, *"Navigating the Patient Pathway"* encompassed the challenging process parents went through in managing their child's healthcare in the lead up to surgery, which could be a significant source of stress. Accessing care was not necessarily straightforward and this was a major strain for parents who could feel lost in the system, but there was support from trusted experts including members of the medical team and other parents, along the way.

Many parents experienced feelings of shock and guilt around the diagnosis of AIS, often questioning how their child's scoliosis had not been detected sooner. Parents also questioned the lack of school scoliosis screening (SSS) programmes in their children's schools. Implementation of such screening programmes has decreased due to concerns about their effectiveness in terms of rates of false-positive referrals and economic costs. However, previous research on this topic has suggested that parents endorse SSS programmes despite potential false positive referrals to orthopaedic clinics (Hines et al., 2015). The findings of the current study also demonstrate that SSS could function to provide families with reassurance that AIS was detected as early as possible. Future research on SSS programmes, and the development of policies around such initiatives should seek to include the perspective of families who would be accessing this service. In the absence of SSS, increased awareness of the signs of scoliosis may be an important consideration for health service campaigns to facilitate timely diagnosis as it is often parents or family members who first notice signs of AIS.

Parents showed signs of resilience as they endeavoured to maintain normality and remain positive despite challenges associated with AIS. Knowing that treatment was available for their child's AIS and expectations of improvement post-surgery may have facilitated parents' coping. This experience differs to reports of Lauder et al. (2018) on parents of children with early onset scoliosis who feel a 'pervasive burden' as caring for their child with EOS affected every aspect of their lives and they could become full-time carers (Lauder et al., 2018). Although many parents had positive experiences in making arrangements with their child's school, the issues experienced by a small number of parents related to lack of understanding of scoliosis was notable. Similarly, in a study by Loades et al. (2020), parents of adolescents with cancer reported a lack of understanding for their child's post treatment recovery needs at school. Misunderstandings around AIS may occur due to the limited visibility of scoliosis in some cases. We also understand from the qualitative study in Chapter 3 that adolescents can have difficulty in communicating about their scoliosis to others. Avoidance of misunderstandings related to AIS in the school setting could be aided by the distribution of standard scoliosis information to staff when a pupil is diagnosed with AIS. For example, this may include guidance around expectations of students with AIS participating in physical education. Other recommendations regarding the needs of adolescents with AIS in school are

likely to be individualised (e.g. time off, locker storage) and this may rely on clear communication between the parents and the school.

Uncertainty and fear of the unknown was pertinent for parents not only at the diagnosis of their child's scoliosis, but also when considering the prospect of surgical intervention. In line with previous quantitative research, concerns included possible surgical complications and postoperative pain (Chan et al., 2017). Parents were also apprehensive about their role in their child's recovery when they returned home from the hospital. Although many parents accepted that spinal surgery was a necessary intervention to prevent worsening of their child's condition and improve their future quality of life, this still appeared to be a difficult decision to make considering the seriousness of the operation. Many uncertainties remained regarding expected outcomes of surgery. Concerns extended beyond the immediate postsurgical period as parents were uncertain about their child's physical capabilities in the future due to having spinal fusion. Although parents were hopeful regarding positive surgical outcomes, more information about life postoperatively and long term outcomes could have benefitted them in feeling that they were well informed in their surgical decision-making. A review of long term health-related quality of life outcomes for AIS has recently been published, indicating generally good quality of life at least 10 years after treatment (Essex et al., 2021). However authors noted limitations of available evidence and a need for further research on this topic.

It was evident that the ongoing strain of managing their child's healthcare in the lead up to surgery was a major stressor for parents. Concern about being "lost in the system" and feeling as though they were "battling" to access care were reported amid uncertainty regarding treatment waiting times. This is comparable to qualitative reports of McDonagh et al. (2021) studying experiences of parents of children with Down's arthritis with themes describing a "Struggle for help" and "A fight for everything." Challenges among the parents interviewed by McDonagh et al. (2021) related more to difficulties in obtaining diagnoses or access to services such as pain management, whereas our findings related more to appointment or treatment waiting times; however, both capture difficulty in accessing required care. These insights demonstrate how taxing interactions with healthcare systems can be for parents amid an already difficult time when they are emotionally processing the diagnosis of a serious

health condition affecting their child. This highlights a more systemic issue whereby adequate resourcing may be lacking in some paediatric health settings. To help parents who experience difficulties with their child's healthcare, clear information regarding patient pathways and navigating the health care system is required in order to clarify this process for parents and perhaps provide them with increased sense of control. Practically, ensuring regular communication with families and providing transparent timeframe information may aid in reducing uncertainty among parents whose children face delays in their treatment for AIS.

While there may have been a lack of trust in the healthcare system, parents demonstrated considerable trust in the experts involved in their child's care. This emphasized the important role that healthcare providers play in supporting parents throughout their child's treatment for AIS. The *"Trusting the Experts"* subtheme is comparable to a theme developed by Bull & Grogan (2010) "Confidence in Professionals" which described how parents' trust in their child's consultant to ease their anxieties when their child underwent surgery. Our findings reiterate this point and also demonstrate the role of the spinal team in the lead up to the surgical period. In particular, when providing information in preparation for surgery, parents valued the transparent and honest approach of spinal nurses and doctors, and their involvement of the adolescent in these important conversations. Considering the clinician's role within modern healthcare to promote patient empowerment and shared decision making, communicating with families in this way is a key step to involving adolescents in their own care (Stein et al., 2019).

The findings of this study build on the systematic review reported in Chapter 6 by providing an in-depth account of parents' experiences from the time of their child's AIS diagnosis to the stage of surgical consideration. Furthermore, research with parents of paediatric patients has predominantly focused on mothers (Swallow et al., 2011; Alderfer, 2017), and the systematic review in Chapter 6 showed that approximately 85% of parent participants in previous AIS studies were mothers. In this qualitative study, fathers represented over one third of the sample representing a move toward a more inclusive understanding of parents' experiences. In practice, a barrier to including both parents was that often one parent would attend healthcare appointments with the child, resulting in only that parent becoming invested in the research project.

Limitations and Future Directions

As noted in the qualitative study with adolescents, this study provides insight into the experiences of a relatively small and ethnically homogenous group of parents recruited from one hospital site, and therefore the insight gained and recommendations related to clinical practice require further research to ascertain their relevance and suitability within different contexts. A further consideration regarding the sample is that recruitment was moderated by staff at the spinal department, and it is possible that parents who had supportive relationships with staff may have been more inclined to participate. In relation to findings regarding navigating the healthcare system, it is important to note that experiences may differ depending on access to private care versus public care. While some parents did discuss accessing private appointments or scans, we did not formally collect information on whether participants had private health insurance and this may be important to document in future studies. Furthermore, only two children of the parents involved in this study had worn a brace as part of their treatment plan and of these, both reported no major challenges in terms of their child's compliance with brace wear. However, it is reported in the literature that compliance with brace wear can be a challenge for adolescents (Brigham & Armstrong, 2017; Rahimi et al., 2019). Had we recruited more parents whose children were wearing a brace, accounts may have varied. As also suggested in the systematic review study, future research specifically exploring brace wear and parents' role in brace wear would be worthwhile, considering the importance of compliance for treatment efficacy.

Conclusion

This study has contributed to the AIS literature by providing insights into parents' experiences related to their child's AIS across the presurgical stage, and how parents adapt to their child's AIS and manage associated healthcare. A diagnosis of AIS can have a pervasive impact on parents, who contend with prognosis uncertainty, adjusting to life with AIS, managing healthcare demands, and the prospect of their child undergoing an invasive surgical procedure. By representing the personal views and perspectives of paediatric patients and their families, qualitative research plays an important role in informing clinical care (Alderfer & Sood, 2016). Study findings have provided the basis for a number of practical suggestions that may improve parents' experiences throughout their child's treatment for AIS and inform delivery of patient and family-centered care. These include tailored and comprehensive

information provision that allows parents to make fully informed treatment decisions, addressing knowledge gaps in the school setting, further investigation of the value of scoliosis screenings from patient and parent perspectives, and enhanced communication with parents regarding navigation of the healthcare system.

Chapter 8

Overall Discussion

Chapter Overview

This final chapter presents an overall discussion of the research presented in the thesis. Firstly, a review of the aims, the associated research studies, and key research findings are provided. This is followed by a consideration of the methodological approaches employed in the thesis. A discussion of the practical implications of the research findings is presented and finally, reflections on future directions in terms of research and practice are provided.

8.1. Review of Aims, Research Studies, & Main Findings

The overarching purpose of this thesis was to further current understanding of psychosocial aspects of adolescent idiopathic scoliosis (AIS), from the perspectives of adolescents and their parents. To this end, a multimethod programme of four research studies was completed, with findings making a number of important contributions to the literature. Under the headings of the overall research questions listed in Chapter 1, a review of the aims, research studies, and key findings is presented.

(i) How do adolescents experience living with idiopathic scoliosis?

With the aim of understanding adolescents' experiences of living with presurgical AIS, a qualitative interview study was conducted with a sample of 14 adolescents recruited from the orthopaedic department of a children's hospital. Using reflexive thematic analysis, four main themes were developed which provided insight into the psychosocial impact of this condition **(Chapter 3)**. The first theme *"Proceeding with Caution"* described adolescents' adaptation to the physical impact of their AIS, as they approached activities with increased caution and avoided straining their back. The second theme *"Am I Different?"* encompassed adolescents' perceptions of their changing appearance. Some adolescents found the appearance aspect of their condition to be particularly burdensome, while some adolescents were generally happy with their appearance despite the changes caused by their spinal curve, highlighting the need to look at factors which may contribute to this variation. The subtheme

"A Hidden Condition" provided insight into adolescents uncertainties and concerns surrounding the visibility of their condition, and potential difficulties in communicating about their scoliosis to peers. The third theme *"An Emotional Journey"* captured the rollercoaster of emotions from shock at diagnosis to the daunting realization of the severity of their condition, while knowing others with AIS could ease the emotional burden. In the final theme, *"No Pain, No Gain,"* adolescents' voiced surgical concerns, but they also expressed positive expectations about their prospective surgery and were often keen to put surgery behind them.

(ii) Are psychosocial processes associated with variation in wellbeing among adolescents with idiopathic scoliosis?

To examine the contribution of psychosocial processes to key outcomes health-related quality of life (HRQOL) and body image, an online survey, the "SCOLI Survey" was conducted with an international sample of 115 adolescents with non-surgically corrected AIS **(Chapter 4).** A preliminary framework was developed to guide this study, informed by the working framework previously proposed by Rumsey et al. (2008). This proved to be a useful approach that acknowledged the relative importance of psychosocial processes and indeed the psychosocial outcomes in the context of AIS, in line with an overall biopsychosocial perspective. Specifically, the contribution of coping strategies to HRQOL, and the contribution of appearance-related cognitions (investment, visibility, subjective perceptions) to body image satisfaction and disturbance were examined with multiple hierarchical regression analyses.

Firstly considering HRQOL, when relevant clinical and demographic variables were controlled for, problem-focused engagement coping was related to better HRQOL. Conversely, the use of problem or emotion-focused disengagement coping was related to lower HRQOL. To further investigate the relationship between coping and HRQOL, a cluster analysis of the coping data collected as part of the SCOLI survey was performed to identify combinations of coping strategies and whether they were associated with differences in HRQOL **(Chapter 5).** Findings demonstrated that the "highly engaged" cluster (i.e., those who more frequently used a range of engagement coping strategies and acceptance and distance, with lower use

of disengagement strategies) had better HRQOL in comparison to the "emotionally disengaged" and "limited strategy" clusters.

In terms of body image outcomes, after controlling for relevant clinical and demographic variables, the appearance-related cognitions (subjective perception of appearance, perceived visibility, appearance investment) accounted for an additional 26% and 24% of variation in appearance satisfaction and body image disturbance, respectively. These findings support the need to acknowledge the multidimensional and subjective nature of body image in understanding body image outcomes among those with chronic health conditions (Pruzinsky et al., 2004). Of interest, subjective perception of appearance (TAPS) had more explanatory power for levels of body image disturbance than the Cobb angle of the spinal curve. Furthermore, higher levels of perceived visibility of scoliosis and higher levels of investment in appearance were associated with lower appearance satisfaction and higher levels of body image disturbance.

(iii) What are parents' experiences related to their child's adolescent idiopathic scoliosis?

To examine what is known about the experiences of parents in the context of their child's AIS, a systematic literature review of this topic was conducted in line with PRISMA guidelines **(Chapter 6).** A total of 18 relevant studies were included, and a synthesis of knowledge related to parents information needs, treatment concerns, and psychological wellbeing was provided. Acquiring knowledge about scoliosis and its' treatment could be a challenge for parents, and online information could be a source of distress. Assessment of parents' scoliosis knowledge has focused on surgical information, however examination of parents' information seeking on a support network indicated that the causes and progression of scoliosis and bracing effectiveness are other key topics for information provision. Parents had many concerns about surgery, including the possibility of surgical complications and postoperative pain. Evidence also suggested that parents of children with AIS are at increased risk for depression and anxiety symptoms, and that the time of diagnosis and undergoing surgery cause heightened anxiety. Parents often sought social emotional support to cope with the stress of their child's AIS, among other strategies. Overall, there was a predominant focus

in the literature on the immediate perioperative period, and less was known about parents' experiences across the trajectory of the presurgical period.

With the aim of understanding the experiences of parents across the trajectory of their child's AIS from diagnosis to presurgical preparation, a qualitative interview study was conducted with a clinically recruited sample of 20 parents whose children were at the presurgical stage of treatment **(Chapter 7).** Reflexive thematic analysis was used to generate three main themes which revealed new insights that built on the findings of the systematic review. The first theme *"Fear of the Unknown"* captured parents' apprehension and uncertainties in relation to their child's diagnosis and prognosis. The findings provided insight into how the diagnosis phase can be anxiety inducing for parents, as they face concerns about whether detection was timely, and guilt around not noticing the signs sooner. From diagnosis, parents worried about what lay ahead for their child, and concerns went beyond the surgical period to their child's future abilities as an adult. Second, *"Maintaining Normality"* described parents' efforts to adapt to the reality of their child's AIS by getting on with life despite the challenges and supporting their child. The emotional burden could often be in the back of their mind but parents were reassured that their child's condition could be treated. Finally, the third theme, *"Navigating the Patient Pathway,"* highlighted parents' difficulties in navigating the healthcare system and managing their child's care prior to surgery. While some parents felt lucky in accessing care, others felt as though they were fighting for it. This theme also captured the supportive role the medical team play during this period.

8.2. Methodological Considerations

8.2.1. Design

This research project was multimethod in nature, including two qualitative interview studies, a survey-based quantitative study, and a systematic literature review. A key advantage of the multimethod approach was the flexibility it provided to use different types of data and analyses to investigate the research topic and address the set of interrelated research questions (Frost & Shaw, 2015; Hesse-Biber, 2015). It should be distinguished that the different methods used were not "mixed" in the sense that the data, methods, or analyses for each study were not strategically integrated with one another throughout the research process (Anguera et al., 2018). Rather, a programme of independent yet complementary

studies was conducted with multiple methods which were best suited to each enquiry. As described by Frost & Shaw (2015), this can be understood as a methods "toolkit" approach whereby different methods were pragmatically selected because of their appropriateness for answering the research questions. Although not integrated, the studies did function to inform each other in some respects. Specifically, the adolescent qualitative data helped to inform parts of the SCOLI survey development, and the parent systematic review provided rationale for the qualitative study with parents. When considering appropriate presentation format for the multimethod approach (Bazeley, 2015) it was deemed most suitable to present each component study separately and then review the findings together in this final chapter. In this way, the findings from each study were intended to complement each other.

It is recognised that research in paediatric psychology has traditionally been dominated by quantitative designs (Berlin et al., 2017) and as outlined in Chapter 3, there have been a very limited number of qualitative studies on the topic of AIS specifically. However, the value of qualitative research is increasingly recognised as an approach that can provide a more in-depth understanding of personal experiences, answer the questions of "how" and "why," and shed light on support needs in paediatric healthcare (Wu et al., 2016; Alderfer & Sood, 2016). In this thesis, Chapters 3 and 7 which detailed interview studies with adolescents and parents have provided rich insight into participants' lives which could not have been obtained through alternative approaches. It is anticipated that qualitative enquiry will continue to play an important role in the future of research related to AIS. For example, in Chapter 5 clustering techniques identified a "highly engaged" subgroup of participants who used a broad range of coping strategies (with lower levels of disengagement strategies) and had better HRQOL compared to those using different coping combinations. To further understand this dynamic process of coping with AIS, it was suggested that qualitative enquiry could elucidate how adolescents employ various coping strategies and the contexts they are used in, to understand why certain strategies might be helpful in different situations.

In comparison to the qualitative studies, the quantitative component of this thesis detailed in Chapters 4 and 5 allowed us to identify specific variables contributing to variation in adolescents HRQOL and body image. By recruiting a relatively large sample size and collecting numeric data, through statistical analysis this research was able to assess the utility of the

conceptual framework developed for the SCOLI survey consisting of predisposing factors, psychosocial processes, and outcomes. Furthermore, the clustering analysis applied in Chapter 5 provided a novel approach that was valuable in generating a more informed understanding of how combinations of coping related to HRQOL. Finally, the value of multiple different methods in the investigation of a research topic was also demonstrated in Chapter 6. All study designs were eligible for inclusion in the systematic literature review (i.e. a mixed studies review) which allowed comprehensive coverage of the literature pertaining to parents of children with AIS. As discussed by Pluye & Hong (2014), a mixed studies review combines “the power of stories and the power of numbers” in synthesizing knowledge on a topic.

8.2.2. Participants

As well as differing methods, this thesis also incorporated different perspectives. While a core focus was placed on the adolescents with AIS, parents perspectives were also sought. This approach was conducive to producing knowledge in line with a patient- and family-centered care (PFCC) approach, acknowledging the importance of meeting the psychosocial needs of patients and concurrent recognition of the role of families in the patient’s life (Eichner et al., 2012). It is recognised that researchers in paediatric psychology are well positioned to make contributions to inform the delivery of PFCC by considering the implications of our research for patients and parents (Alderfer et al., 2017).

It should be highlighted that although parents are typically the focus of PFCC in the context of paediatrics, we did not consider the perspectives of other family members in this study. The need to reach “beyond mothers and patients” (Alderfer et al., 2017) has been emphasised for paediatric research, and to this end, we endeavoured to include representation of fathers in Chapter 7. However, inclusion of other family members in future work (e.g., siblings) could provide a more complete understanding of the impact that AIS can have on the family overall. Research regarding the impact of paediatric chronic conditions on siblings is mixed regarding the impact on sibling wellbeing, but generally, they can experience changes in routines, changing roles in the family, and concern about their affected sibling (Barlow & Ellard, 2006; Lummer-Aikey & Goldstein, 2021). Characteristics specific to the condition can determine the impact on siblings, and most siblings adapt well, with some reporting increased maturity and

empathy (Vermaes et al., 2012; Kazak et al., 2017). To our knowledge, no studies have yet investigated the experiences of siblings of those with AIS.

Furthermore, there were differences across the study samples included in this thesis which are worth considering in the interpretation of results. In the qualitative studies, (Chapters 3 & 7) a purposive sampling strategy was employed within the clinical setting and the participating adolescents were candidates for surgery (all curves 45°+). More heterogeneity was observed in the adolescent survey sample (Chapters 4 & 5) in terms of curve size and treatment status (42% curves 45°+), and some participants in this study may never reach the surgical threshold. As discussed in Chapter 3, the findings of the qualitative study are likely to be most applicable to presurgical candidates. However, it is plausible that patients with smaller curvatures experience similar issues and challenges as reported in the qualitative analysis, possibly to a lesser extent. However, it should be emphasized that while condition severity is an important consideration in AIS, it is not a definitive indicator of how AIS impacts upon the wellbeing of adolescents, and other variables such as the psychosocial processes studied in Chapters 4 & 5 play a role. The parent sample in Chapter 7 were homogenous in relation to anticipating surgery for their child. However, parents did reflect on their experiences since time of diagnosis, providing retrospective insight into earlier stages of the trajectory.

The study samples also differed in relation to the age range of adolescents. The range for the qualitative study in Chapter 3 was 12 – 18 years and this was slightly extended for the survey (Chapters 4 & 5) where participants were aged 12 – 21 years. This difference was partially due to practical reasons as only those up to age 18 were accessible within the paediatric clinical setting where recruitment for Chapter 3 took place. As survey recruitment was community-based through patient support networks, those up to age 21 were present. This extended age range was chosen for the survey in light of the understanding that adolescence as a period of development extends into the early twenties (Alderman et al., 2019), as well as clinical guidance that there are individuals aged over 18 with AIS who have not had surgical intervention and that the survey would be highly relevant to their experiences. However, it is notable that the majority of the resulting survey participants (83%) were aged 18 years or younger.

8.3. Practical Implications

Across the studies conducted within this thesis, a secondary objective of the investigations was to inform the delivery of care and support to patients with AIS and their parents. Together, the findings have implications for practice. However, it should be noted that overall, research into psychosocial support for the AIS group is in the early stages and as articulated previously in the thesis, recommendations are considered tentative and require research to evaluate their suitability and efficacy.

Based on the theme *"Proceeding with Caution,"* it was suggested that adolescents may require guidance in the clinical setting on suitable activity participation when living with AIS, in order to promote healthy physical activity in this group, as there is no definitive guidance available (Tarrant et al., 2014). Descriptive findings from the SCOLI survey showed that the participants who reported stopping or reducing their activity participation had significantly larger curves in the surgical range (M = 49°) compared to participants who reported participation as normal (M = 37°). Therefore, this may be particularly relevant to address for those with larger curves. Also relevant to those at the stage of surgical consideration is surgical expectation management. As discussed in Chapter 3, while focusing on benefits of their upcoming surgery may help adolescents to cope during this time, management of postsurgical expectations may be required as part of presurgical counselling to ensure realistic expectations (Sieberg et al., 2018).

Addressing health-related communication needs was also identified as an area where adolescents can require more support based on the qualitative subtheme *"A Hidden Condition."* Furthermore, the SCOLI survey identified that the majority of participants perceived that their scoliosis was visibile to others "sometimes," meaning that the decision to discuss or disclose their scoliosis may often be up to them. Educational strategies aimed at preparing adolescents to communicate about their health conditions in terms of how and what adolescents may want to tell others and taking initiative in informing others (e.g., at school, in peer groups) about issues related to their condition (e.g., reasons for absences, limitations) may be worthwhile (e.g., Last et al., 2007; Douma et al., 2019). Moreover, 79% of participants rated the strategy of "learning how to explain my scoliosis to others" as definitely or probably helpful in the SCOLI survey. Most participants (90%) also rated "increased

awareness about scoliosis in their community" as probably or definitely helpful. This highlights that while preparing adolescents to communicate their condition to others is worthwhile, a wider educational approach at the school or perhaps national level to promote awareness of scoliosis may also improve the experience of those living with AIS.

As demonstrated in Chapter 4, psychosocial processes were shown to be associated with variation in key outcomes HRQOL and body image among adolescents with AIS, accounting for similar if not more variance in these outcomes compared to clinical characteristics. In relation to body image, this underscores the importance of assessing patients' subjective perceptions of their appearance (i.e., in terms of trunk appearance perception and perceived visibility). These may be important indicators of levels of body image disturbance as opposed to more objective or clinical measurements of condition severity, a finding that is previously suggested in relation to other conditions (Moss, 2005; Feragen & Stock, 2018). It was also demonstrated in Chapters 4 and 5 that coping can play an important role in adolescents' HRQOL. Screening of coping strategy usage may be helpful to identify those who exhibit lower levels of adaptive strategies and may therefore be at risk for lower HRQOL. Efforts to promote the use of adaptive strategies, and adaptive combinations of coping strategies, so that adolescents have a coping "toolkit" (e.g., engagement strategies, acceptance, distance) would be worthwhile, as previous research indicates effectiveness of coping skills training for other paediatric populations (Grey et al., 2000; Serlachius et al., 2012).

Findings across Chapter 3 and 4 also indicate potential utility of peer support strategies among adolescents with AIS, building on previous suggestions for an online information resource with opportunity for peer interaction (MacCulloch et al., 2009). Qualitative insight obtained in Chapter 3 demonstrated that adolescents who had interacted with others with AIS could gain emotional and practical support, feeling as though they were *"not the only one."* Further insight gathered in the SCOLI survey in Chapter 4 demonstrated that 97% of those who had met another young person with scoliosis (online or in person) had rated this as a positive experience, and there was relatively high endorsement of the usefulness of peer support strategies, whether online or in person. Peer support strategies provide opportunity for individualised and flexible psychosocial support, and while peer support could be facilitated through community support groups for AIS, there is also possibility of delivering

such support through online or remote programmes (e.g. Ahola Kohut et al., 2018; O'Higgins et al., 2019).

For parents, the importance of information provision was evident in the findings of Chapter 6 and 7. As well as information about treatment options and undergoing surgery, parents required more knowledge on looking after their child postoperatively, and their child's future and long-term surgical outcomes. There can also be many uncertainties for parents in the earlier stages of AIS following diagnosis, in terms of why their child was affected and what is ahead of them, as captured by the theme *"Fear of the Unknown"* in Chapter 7. Existing tools such as the Scoliosis Knowledge Questionnaire (Khetani et al., 2008) which focuses on surgical treatment could be expanded to create a more holistic compilation of knowledge that parents may need over the course of their child's care, to provide parents with a comprehensive list of queries or topics that they may wish to discuss with members of the healthcare team. This would promote tailored information provision, enhanced communication between healthcare providers and parents, and informed decision making, all key aspects of patient and family-centered care (Eichner et al., 2012; Nightingale et al., 2015). Of note, parents' experiences of sourcing information online could cause distress and this further emphasizes the important role that healthcare providers play in addressing parents information needs.

In the absence of widespread screening programmes for scoliosis, the diagnosis of scoliosis can be associated with feelings of guilt in parents and worry about timely detection. As discussed in Chapter 7, routine screening could improve this experience for parents providing reassurance that their child's AIS is treated early. The rollout of such programmes has been halted in may locations due to cost effectiveness and concern regarding false positive referrals and improvements of screening strategies is required (Hines et al., 2015). In the meantime, an alternative and complementary strategy for increasing early detection of AIS could be to increase awareness of the signs of scoliosis among the general population. Furthermore, from the point of diagnosis, parents face uncertainty as to what lays ahead for them and their child. This experience can be further complicated by issues with timely care for AIS as parents face difficulty in ensuring the best care for their child. Findings reported in Chapter 7 under *"Navigating the Patient Pathway"* demonstrate how taxing interactions with healthcare systems can be for parents amid an already difficult time when they are

emotionally processing the diagnosis of AIS in their child. Transparent communication with parents regarding the patient pathway and their child's treatment status is imperative to reduce potential feelings of being "lost in the system" or as though their child may be forgotten about.

8.4. Future Directions

Focusing on the practical implications detailed above and throughout the thesis, recommendations for improving support could be used to inform the future development or adaptation of a psychosocial intervention for adolescents with AIS, beyond the standard delivery of care. Specifically, some of the strategies recommended here have been incorporated into web-based programmes for adolescents with other chronic conditions to reduce or prevent psychosocial problems. For example, the *Op Koers* small group-based programme includes components such as teaching the use of engagement coping skills (e.g. cognitive restructuring), how to communicate about your condition to others, condition-specific information/ education, and use of relaxation techniques (Last et al., 2007; Douma et al., 2019; 2021). A recent RCT demonstrated that the online program had a positive effect on use of adaptive coping and HRQOL among adolescents with conditions including asthma, diabetes, IBD, and arthritis (Douma et al., 2021). Of interest, there is also an Op Koers parent module that can teach parents adaptive coping skills related to their child's condition, that has been shown to decrease parental anxiety and depression (Douma et al., 2020), and a recent review has also provided support for the use of psychoeducational programmes for parents of children with chronic health conditions (Costa et al., 2021). For adolescents with juvenile idiopathic arthritis in Ireland, researchers have recently adapted an integrated programme of the *iPeer2Peer* and *Teens Taking Charge* programmes with components including peer support, self-management strategies, and condition-specific information, and two modules for parents, with evaluation ongoing (O'Sullivan et al., 2018; O'Higgins et al., 2019). It is worthwhile to consider these programmes that have previously been developed, as similar programmes could be adapted in future work for AIS. In addition, given the potential for AIS to impact upon body image (Auerbach et al., 2014), inclusion of strategies to prevent body image dissatisfaction could also be considered (Pruzinsky, 2004; Yager et al., 2013).

Researchers have already begun to adapt an online support strategy for patients with AIS, specifically the *iCanCope PostOp* smartphone app designed to assist with self-management of postoperative pain (Birnie et al., 2019; O'Reilly et al., 2019). Given the specific focus on pain and the postsurgical period, future developments aimed toward the presurgical period are warranted. Web-based programmes offer the advantage of flexible participation in the patient's own time and are considered a relatively convenient and low cost option. However, it is worth cautioning that Douma et al. (2021) reported difficulties with recruitment for their RCT evaluation of the *Op Koers Online* as did Durand et al. (2021) for their pilot testing of the integrated *iPeer2Peer* and *Teens Taking Charge* programme, potentially indicating a lack of motivation among adolescents. Furthermore, the SCOLI survey showed that 82% of participants deemed "in-person peer support" definitely or probably helpful, and 62% rated "online peer support" as definitely or probably helpful, indicating a slight preference for in-person strategies or a combination of both. Feasibility testing and attention to adolescents preferences would be necessary in the development or adaptation of psychosocial interventions for AIS in order to ensure interest and uptake. To assist with this, inclusion of a strong Public and Patient Involvement (PPI) component from the outset of any such projects would be beneficial (Bate et al., 2016).

In this research project, adolescents and parents perspectives were sought in order to understand psychosocial aspects of AIS within a patient and family-centered approach. However, their perspectives were studied separately. Taking a family systems perspective, future research could incorporate dyadic assessment and analysis in order to investigate interrelations between adolescent and parent. Family systems theory postulates that the family operates as a unit and that individual members are interdependent on each other, so a change in one member is reciprocated by changes in other members (Alderfer & Stanley, 2012). As such, while a diagnosis of AIS in their child may lead to stress and concern for parents, it is also plausible that parents responses to their child's AIS can in turn, influence their child's adjustment (e.g., in terms of HRQOL) (Kazak et al., 2017). In this regard, preliminary evidence included in the systematic review in Chapter 6 indicated that parental anxiety and depression were moderately correlated with children's anxiety and depressive symptoms in AIS samples (LaMontagne 2001; Wang et al., 2019). To build on this, future

research could seek to investigate how parental responses and functioning (e.g., coping, mental health) may influence their child's adjustment to AIS.

In this thesis, reliable measures assessing psychosocial domains affected by scoliosis were included in the SCOLI survey (e.g., SRS-22r, BIDQ-S). While these are comprehensive measures, it is possible that other outcomes of importance to patients were not assessed in this study. Ongoing research in Queen's University Belfast is working toward the development of a core outcome set of measures for AIS, representing the outcomes of most importance to adolescents, their families, and health care professionals (Graham-Wisener et al., n.d.). Importantly, the voice of patients is essential in ensuring a core outcome set is of relevance to the patients themselves (Sherratt et al., 2020), and in line with this, the process is set to involve qualitative research and young person advisory groups. This work may uncover additional psychosocial domains to be included in future studies with the AIS population.

It would be remiss not to highlight the systemic issue of timely access to care in the treatment of scoliosis, a topic which has been touched on within this thesis. While healthcare waiting times are a complex and multifaceted issue, there is evidently a lack of appropriate resourcing of paediatric orthopaedic services, as reported in Ireland and in other countries (Clark, 2008; Miyanji et al., 2015; Tarrant et al., 2016; Anthony et al., 2021; Ombudsman for Children, 2017). Treatment delays and difficulties in accessing care can lead to deterioration of the condition and exacerbate the psychosocial challenges faced by adolescents and their parents. Importantly, progression of spinal curvatures far beyond the surgical threshold is likely to lead to increased distress in adolescents that could be avoided. Insight into parents difficulties were captured within the *"Navigating the Patient Pathway"* theme in Chapter 7, as parents could find themselves battling to ensure timely care for their child. Of note, the COVID-19 pandemic has compounded treatment delays for some patients, as demonstrated in the survey findings in Chapter 4. However, the studies presented in this thesis did not have many participants with curves far exceeding the surgical threshold or participants with complex care needs related to scoliosis of non-idiopathic causes. Issues with access to care may therefore be a more significant problem among other samples. It is acknowledged that healthcare professionals involved in treating those with AIS often go beyond what is required of them (Ombudsman, 2017) and the trust parents place in these professionals was

highlighted in Chapter 7. Clearly, the issues related to inadequate resourcing and operational capacity is a key priority in improving the experience and health of those affected by AIS.

8.5. Conclusion

To conclude, it is suitable to return to the question posed in the case vignette presented in the first chapter of this thesis: *"How should the patient be evaluated and treated?"* Modern medicine continues to advance the efficacy and safety of conservative and surgical treatment for adolescents with AIS, and providing timely access to required care is a priority for all patients and their families. Alongside appropriate medical care, the research presented in this thesis has highlighted the importance of considering the psychosocial implications of living with AIS, for both the adolescent and their parents. Through a multimethod programme of research, this thesis has provided a number of original contributions to the literature on AIS by gaining in-depth insight into the experiences of adolescents living with idiopathic scoliosis, and identifying psychosocial processes leading to variation in wellbeing among this patient group. Knowledge on the role of parents in their child's AIS was synthesized and their experiences explored across the trajectory of their child's condition from diagnosis to presurgical preparation. Overall, the value of a patient and family-centered approach to care in AIS and indeed to research on this topic has been emphasised throughout and findings have a number of implications for practice. While further research is required to advance the research agenda on psychosocial aspects of AIS and the development of supports for this patient group, the studies included in this thesis should inform and encourage continued progress.

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Appendix A: Publication Relating to Chapter 3

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Original Research Article

Adolescents' Experiences of Idiopathic Scoliosis in the Presurgical Period: A Qualitative Study

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Abstract

Objective Adolescent idiopathic scoliosis (AIS) is a sideways curvature of the spine that can progress severely during adolescent development and require surgical intervention. This qualitative study was conducted to explore the psychosocial experiences of adolescents with idiopathic scoliosis during the presurgical stage of treatment. **Methods** Fourteen adolescents with moderate-to-severe AIS aged 12–17 years participated in semistructured interviews and data were analyzed using inductive reflexive thematic analysis. **Results** Four key themes were generated from the analysis. "Proceeding with Caution" described adolescents' adaptation to the physical impact of their AIS, while "Am I Different?" encompassed adolescents' perceptions of their changing appearance and visibility of their condition. "An Emotional Journey" captured the rollercoaster of emotions from shock at diagnosis to the daunting realization of the severity of their condition, while knowing others with AIS could ease the emotional burden. Finally, adolescents' concerns and expectations about their prospective surgery were captured by the theme "No Pain, No Gain", whereby they were often keen to put surgery behind them. **Conclusions** Understanding and addressing adolescents' psychosocial support needs as they manage the challenges associated with idiopathic scoliosis is a key component of promoting better outcomes among this patient group. Clinical implications and opportunities for support provision are discussed.

Key words: adolescents; chronic illness; psychosocial functioning; quality of life.

Introduction

Adolescent idiopathic scoliosis (AIS) is a sideways curvature of the spine that is typically diagnosed in the early teenage years (Weinstein et al., 2008) and is one of the most common pediatric spinal conditions seen by primary care physicians, pediatricians, and orthopedic surgeons, affecting approximately 1–3% of adolescents (Altaf et al., 2013). Although the etiology of AIS is uncertain, growth spurts during adolescent development are understood to contribute to progression of the spinal curve (Beauchamp et al., 2019). Curve progression can be associated with symptoms including postural asymmetries, imbalanced shoulders and hips, rib prominence, back pain, and pulmonary issues (Weinstein et al., 2008; Altaf et al., 2013). Living with this often progressive condition can be burdensome for adolescents, who are already experiencing the normative developmental challenges of adolescence including pubertal changes, increasing independence from parents, and a desire for peer conformity. Adolescents with AIS are more likely to report appearance self-consciousness, associated problems with social interactions (Auerbach et al., 2014), and higher levels of body image dissatisfaction compared with nonaffected peers (Tones et al.,

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Appendix B: Publication Relating to Chapter 6

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Parents' information needs, treatment concerns, and psychological well-being when their child is diagnosed with adolescent idiopathic scoliosis: A systematic review

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ABSTRACT

Objective: We systematically reviewed the experiences of parents who have a child with adolescent idiopathic scoliosis in order to understand their needs and concerns related to their child's healthcare, and assist health professionals in supporting parents of this paediatric patient group.

Methods: A systematic search strategy identified eighteen relevant studies published between 2000 and 2020. Quality was assessed using the Mixed Methods Appraisal Tool and the literature was narratively synthesised.

Results: Three main themes were evident across the literature including information needs, treatment concerns, and psychological well-being. Studies predominantly focused on the surgical treatment of scoliosis.

Conclusion: Parents face challenges such as acquiring appropriate knowledge about scoliosis to participate in healthcare decisions and coping with their child undergoing invasive spinal surgery. Throughout this time, their psychological well-being can be negatively impacted. Considering parents' experiences and support needs throughout this anxiety-provoking time is an important step in delivering family-centered care and promoting better outcomes for paediatric patients.

Practice Implications: Providing parents with appropriate resources and addressing concerns around surgical complications, postoperative pain, and how they can best support their child before and after surgery, may alleviate some of the emotional burden that parents experience.

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Appendix C: Protocol for Qualitative Data Collection

The following protocol includes guidelines followed for the qualitative data collection. These guidelines were intended to ensure consistency over the course of data collection.

1. Arranging Data Collection

The researcher will make contact with the parent/guardian via their phone or email details provided. Alternatively, the parent/guardian may phone or email the researcher. The following points will be covered to arrange a meeting for data collection;

- Clarify whether the parent/guardian or their child (with AIS) wish to take part in the research. If no, seek reason as to why they do not wish to participate on this occasion and document this reason.
  If yes, explain that data collection can take place in their home or in a private location in the paediatric hospital. Arrange a date and time which suits participants.
- Advise that a quiet and private space will be required for conducting the interviews.
- Check whether the participants have any questions about the research.

2. Conducting Data Collection

Data collection which takes place in participants homes will be conducted in accordance with the UCD Home Visit Guidelines (UCD SIRC Office, 2015). For data collection the researcher will require: Consent and assent forms, Audio recording device, Printed copy of interview guides, Participant information questionnaires, Data storage box.

Obtaining Informed Consent and Assent

- Ensure participants have read the information sheets (which they will have received previously) and clarify any questions they may have.
- Briefly go through the points on the consent form with the participants and clarify any questions they may have. Remind participants that the interviews will be audio recorded and that no identifiable information will be published or presented.
- The parent/guardian will then sign the consent form and the researcher will also sign and date the form. An assent form will also be available for the young person (under age 18 years) to sign.

Conducting the Interviews

Adolescent interview to be conducted first, followed by parent interview.

- Give participant the participant information questionnaire (presented below) to complete while getting the interview materials ready and setting up the recording device. Check with the participant whether they would like any help in completing this.
- Chat to the participant casually (e.g. about their day) to build rapport and help the participant to feel at ease.

(i) Prior to Interview:

For Adolescents: "Thanks again for joining me today to contribute to this research. I am interested in finding out about your experience of scoliosis, so we will be talking about things like how you feel about your scoliosis and how this might impact other areas of your life. Sharing your personal experience in this way will help us to have a better understanding of what it's like to have scoliosis as a young person. We hope that this information will be used to inform the support of young people like you."

For Parents: "Thanks again for taking part in this research study. I am interested to hear what your experience has been like as a parent of someone with scoliosis. Sharing your personal experience in this way will help us to have a better understanding of how scoliosis can affect young people and their families."

- Remind participant that if there are any questions they do not want to answer that is fine and they can stop the interview at any time.
- Provide the participant with an opportunity to ask questions before beginning the interview.

(ii) Follow interview guides for adolescent or parent (presented below).

Guidelines for Handling Instances of Distress

It is possible that the participants may become distressed while talking about sensitive topics during the interview. If a participant shows signs of distress (i.e. upset, uncomfortable), the researcher will handle the situation sensitively, as follows;

(i) It is important to acknowledge the participants distress. Remind them that they can have a break or end the interview at any time.

(ii) Empathise with the participant and convey understanding that these topics can be difficult to discuss. Ask the participant if they are okay to continue the interview.

(iii) If the participant is happy to continue, resume the interview. If the participant appears unable to carry on, or expresses that they wish to end the interview, discontinue the interview.

(iv) Sensitively notify the young person's parent/guardian following the interview that their child was upset and suggest they may discuss this further with their child to ensure they are okay.

(v) In the event that a young person participating in the interview is in significant distress or discloses information which indicates their wellbeing is at risk, the designated psychologist at the paediatric hospital will be informed following the interview and the parent will be sensitively informed as appropriate.

___

Participant Information Questionnaires for Qualitative Study

Adolescent Information Questionnaire

Participant ID	
Age	
Date of Birth (dd/mm/yyyy)	
Gender	
Ethnicity (e.g. White-Irish, Asian-American)	

Please ask your parent/guardian or the researcher about these questions if you are unsure:

Scoliosis Type	Thoracic(upper back) ☐	Lumbar(lower back) ☐	Thoracolumbar ☐
Curve Magnitude *(degrees)*			
Length of Diagnosis *(month/year you found out you have scoliosis)*			
Treatment Details			

Parent/Guardian Information Questionnaire

Participant ID	
Relation to adolescent participant (e.g., mother)	
Age	
Date of Birth (dd/mm/yyyy)	
Gender	
Ethnicity (e.g., White-Irish, Asian-American)	
Occupation	
Number of children	

Summary of the Interview Schedule for Adolescents with Idiopathic Scoliosis

- To get started, can you tell me a bit about yourself?
  (e.g., hobbies, school)
- Can you tell me about when you first found out that you had scoliosis?
  How did you find out? How did you feel about it?
- Could you tell me about the treatment you have been receiving for your scoliosis?
  Are you considering surgery?
- Is there anything in your life that you think is affected by your scoliosis?
  (e.g., school, activities, relationships)
  Has anything been particularly difficult for you since you've had scoliosis?
  Has your scoliosis had any positive impact on you?
- Do you have any concerns about your scoliosis?
- Have your thoughts and feelings been affected by your scoliosis?
  (e.g., mood, self-esteem)
- Some people with scoliosis can experience changes to their body, do you think your scoliosis affects the way that you look?
  How happy are you with the way your body looks?
- Can you tell me about how you cope with having scoliosis?
  (Helpful or unhelpful)
  (e.g., can you think of a time when you found your scoliosis challenging and remember how you dealt with it?)
- Are there any resources or services that have been helpful in supporting you?
  (e.g., healthcare system, scoliosis groups, meeting others with scoliosis)
  Anything you would change or improve?
- That's just about everything I had to ask you, is there anything you would like to add?
- Are you happy for the interview to end?

Note. Main questions indicated by bullet points with possible probes presented underneath.

Summary of the Interview Schedule for Parents

- To get started, can you tell me a bit about yourself and your family?
- Can you tell me about your child's diagnosis of scoliosis?
  - Can you tell me about their stage in treatment?
- How would you describe your own reaction or your approach to dealing with the diagnosis?
  - Can you tell me how you felt about the diagnosis?
  - How do you feel about it now?
- Since the diagnosis, what kind of impact do you think scoliosis has had on your child?
  - Is there anything that you do to help manage this impact on your child?
- Do you have any concerns about your child's scoliosis?
  - How are you feeling about the future?
- Thinking about your personal experience, can you tell me what kind of impact your child's scoliosis has had on your life?
  (e.g., personal life, family, work, social)
- Personally, what kind of impact has your child's scoliosis had on you and how you feel?
- How do you manage or cope with the impact that your child's scoliosis has on you/ your family?
- Are there any supports which have been helpful to you over the course of your child's scoliosis?
- How has your experience been within the hospital/ healthcare system?
- If there was one thing that could have been better about your experience so far, what would it be?
- That's just about everything I had to ask you, is there anything you would like to add?
  - Is there anything you would like to further discuss?

Note. Main questions indicated by bullet points with possible probes presented underneath.

Appendix D: COVID-19 and Survey Timeline

Time Period		Feb – Apr 2020	May – Aug 2020	Sept 2020
Survey Status		Survey design ongoing	Ethical approval granted in July	**Recruitment commenced Sept 7th**
COVID Status	IRE	First confirmed case of coronavirus 29th Feb. Restrictions implemented 12th Mar (e.g. school closures), national 'stay at home' order until May.	'Roadmap' to easing of restrictions followed. Restrictions varied in severity throughout this time.	New restriction 'level' system introduced. Many protective measures still in place but schools reopened.
	UK	First cases of the virus. Restrictions introduced, lockdown regulations come into effect.	Restrictions vary over summer months. Partial opening of schools in June (optional).	Students returned to school. Other restrictions remain & vary.
	USA	Public health emergency declared Jan 31st. Public school closures began in Mar. Social distance measures introduced from Mar 19th.	Restrictions (e.g. mask mandates) vary among states.	Limited part-time school reopening, varied depending on school districts.
	CAN	First case of COVID on Jan 25th. Between Mar 12th and 22nd, all provinces had introduced restrictions/ social distancing.	Varying levels of restrictions among provinces over summer, relaxing of measures in June.	Return to school with mixture of in person and remote learning differing by province.

(Contd.)

Time Period		Oct - Nov 2020	Dec 2020	Jan - Feb 2021
Survey Status		Data collection ongoing	Data collection ongoing	**Data collection completed Feb 28th**
COVID Status	IRE	Full lockdown restrictions from Oct 21st to Dec 1st, with schools remaining open.	Easing of restrictions during Dec (e.g. opening of non-essential retail, restaurants). First vaccine administered Dec 29th.	National lockdown re-introduced from 31st Dec 2020. School reopening after holidays postponed to March.
	UK	COVID regulation tier system introduced. Month lock down from Nov 5th, schools/universities open.	Commencement of vaccine rollout Dec 8th. Relaxation of restrictions for holidays. Emergence of new COVID variant.	Surge in cases. Lockdown from 5th Jan including school closures, to reopen in March.
	USA	Celebration of thanksgiving causes concern for surge, leading to some tightening of restrictions.	First vaccine administered Dec 14th.	School reopening varied across states, Mar – Apr.
	CAN	Localized lockdowns in areas experiencing surge in cases, as second wave heightens.	On Dec 14th, vaccinations began. Recommendations in many areas not to gather for holidays.	Schools began to reopen in some regions from late Jan/ early Feb.

IRE: Ireland; UK: United Kingdom; USA: United States of America; CAN: Canada.

Appendix E: The SCOLI Survey

SURVEY

For Adolescents with Idiopathic Scoliosis

Section 1: General Information

This section asks questions about you and your scoliosis.

1. **What is your age?** __________

2. **What is your gender?**

☐ Female ☐ Male ☐ Other______________

3. **What is the size of your main scoliosis curve, according to your most recent measurement?** *Please write the number of degrees (known as the Cobb angle). If unsure, you could check with your parent/guardian.*

____

4. **Where are you living?**

☐ Ireland ☐ Northern Ireland (UK) ☐ England, Scotland, or Wales (UK)

5. **What is your ethnicity?**

☐ White ☐ Asian
☐ Mixed/Multiple ethnic groups ☐ Black
☐ Another ethnic group

6. **How would you describe where you live?**

☐ In a city/suburb ☐ In the countryside outside a town/city
☐ In a town ☐ On a farm
☐ In a village ☐ Other ______________________

7. **What stage of school/education are you in?**

☐ Primary school ☐ Secondary school ☐ College / University
☐ Other _______________________________________

8. **What is your height?** ______________ (cm) OR ________________ (feet and inches)

9. **What is your weight?** ______________ (kg) OR _________________ (stone and lbs)

10. What is your scoliosis curve type?

- ☐ Thoracic (upper back)
- ☐ Lumbar (lower back)
- ☐ Thoracolumbar (upper and lower, double curve)
- ☐ Not sure

11. How long has it been since you were first diagnosed with scoliosis?

- ☐ 0 - 6 months
- ☐ 7 – 12 months
- ☐ 1 – 2 years
- ☐ 2 – 3 years
- ☐ 3 – 4 years
- ☐ 4 – 5 years
- ☐ Over 5 years

12. My Scoliosis (TAPS Measure)

(a). Please draw a circle around the figure that you think looks most like your body:

(b). Please draw a circle around the figure that you think looks most like your body:

(c). Please draw a circle around the figure that you think looks most like your body:

(Females) OR (Males)

(d). Do you think your scoliosis is visible to other people?

☐ Never visible ☐ Visible sometimes ☐ Visible most of the time ☐ Visible all of the time

13. What treatment are you receiving for your scoliosis? (tick all that apply)

☐ I attend hospital appointments to get X-rays of my back

☐ I wear a brace (on my torso/ back)

☐ I am waiting for a surgery date to be organised

☐ My surgery is scheduled

☐ I am not receiving any treatment for my scoliosis

☐ Other treatment, please write in space: ___________________________________

14. Do you receive physiotherapy for your scoliosis & how often? (tick all that apply)
We understand in recent times this may have changed because of Coronavirus, but please select what is normal for you.

☐ Never had physiotherapy for my scoliosis

☐ Physiotherapy at a local hospital/practice

If so, around how many times per month? _____

☐ Scoliosis-specific exercise physiotherapy (e.g. Schroth)

If so, around how many times per month? _____

☐ I do a daily exercise programme for my scoliosis

15. Has your medical treatment for your scoliosis been impacted by the Coronavirus pandemic? (for example, surgery or hospital appointments delayed or cancelled, unable to go to physiotherapy)

☐ No ☐ Yes ☐ Not Sure

If yes, please tell us how it was impacted: _____________________________________

____

16. Are you experiencing any health complications related to your scoliosis? (e.g. shortness of breath)

☐No ☐Yes, please write them here ____

17. Are you currently receiving treatment for any diagnosed medical conditions other than scoliosis? (e.g. athsma, diabetes)

☐ No ☐ Yes, please write them here ____

18. Do you normally take part in any sport or extra-curricular activities? (e.g. Swimming, Soccer/Football, Dancing etc.)*

☐ No ☐ Yes, please list: ____

If so, how many hours per week (on average)? _____________

**(We understand in recent times this may have changed because of Coronavirus, but please report what is normal for you).*

19. Have you had to stop participating in any sports or activities <u>because of your scoliosis?</u> If so, please list the sports/activities you have stopped.

☐ No, I still take part in my sports/ activites as normal

☐ Yes, I have completely stopped going to all or some of my sports/ activities

Which ones? ___________________________________

☐ I have had to reduce how much I take part in sports/ activities

Which ones? ___________________________________

20. How often is your scoliosis a problem your life?

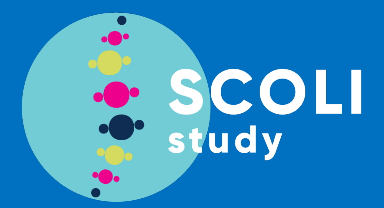
☐ Never ☐ Sometimes ☐ A lot of the time ☐ All of the time

In what ways?

____

____

CONTINUE TO NEXT SECTION →



This section asks questions about how you feel about your appearance. Remember there are no right or wrong answers, everyone's answers are personal to them.

Part 1. Please read each statement about your overall/general appearance and tick how often you agree with each statement. (BESAA-A)

	Never	Seldom	Some-times	Often	Always
I like what I look like in pictures.	☐	☐	☐	☐	☐
I like what I see when I look in the mirror.	☐	☐	☐	☐	☐
There are a lot of things I'd change about my looks if I could.	☐	☐	☐	☐	☐
I wish I looked better.	☐	☐	☐	☐	☐
I wish I looked like someone else.	☐	☐	☐	☐	☐
My looks upset me.	☐	☐	☐	☐	☐
I'm satisfied with how I look.	☐	☐	☐	☐	☐
I feel ashamed of how I look.	☐	☐	☐	☐	☐
I worry about the way I look.	☐	☐	☐	☐	☐
I look as nice as I'd like to.	☐	☐	☐	☐	☐

Part 2. Please read each statement about your overall/general appearance and tick how much you agree with each statement. (MBSRQ-AO)

	Definitely disagree	Mostly disagree	Neither agree nor disagree	Mostly agree	Definitely agree
Before going out in public, I always notice how I look.	☐	☐	☐	☐	☐
I am careful to buy clothes that will make me look my best.	☐	☐	☐	☐	☐
I check my appearance in a mirror whenever I can.	☐	☐	☐	☐	☐
Before going out, I usually spend a lot of time getting ready.	☐	☐	☐	☐	☐

(continued)	Definitely disagree	Mostly disagree	Neither agree nor disagree	Mostly agree	Definitely agree
It is important that I always look good.	☐	☐	☐	☐	☐
I use very few grooming products *(e.g. cosmetics, make up, hair styling products).*	☐	☐	☐	☐	☐
I am self-conscious if my grooming isn't right. *(worried about my appearance).*	☐	☐	☐	☐	☐
I usually wear whatever is handy without caring how it looks.	☐	☐	☐	☐	☐
I don't care what people think about my appearance.	☐	☐	☐	☐	☐
I take special care with my hair grooming *(styling).*	☐	☐	☐	☐	☐
I never think about my appearance.	☐	☐	☐	☐	☐
I am always trying to improve my physical appearance.	☐	☐	☐	☐	☐

Part 3. Sometimes, young people with scoliosis can be worried about how their back looks. **Please read each question carefully and tick the answer that best describes your thoughts and feelings about your back. Some questions ask you to write in your own answers. (BIDQ-S)**

1(a). Are you worried about the appearance of your back shape?

☐ Not at all worried (skip to Q3) ☐ Somewhat worried ☐ Moderately worried ☐ Very worried ☐ Extremely worried

1(b). If so, what are these concerns? (Tick all that apply)

☐ My shoulders are uneven (one is higher or lower than the other)
☐ My shoulder blade sticks out
☐ My chest is asymmetric from the front (one side higher or lower than the other side)
☐ My hips are asymmetric (one hip is higher or lower than the other)
☐ My rib bump

2(a). If you are at least somewhat concerned or worried, do these concerns/worries preoccupy you? That is, you think about them a lot and they're hard to stop thinking about?

☐ I don't think about them
☐ I think about them from time to time
☐ I think about them a moderate amount
☐ I think about them a lot
☐ I think about them constantly

2(b). If you identified concerns about the way your back looks, do these concerns affect your life in any way?

____

____

____

3. Has the way your back looks caused you to feel upset?

☐ Not upset at all
☐ A little bit upset
☐ Somewhat upset
☐ Very upset
☐ Extremely upset

4. Have worries about how your back looks caused you any problems at school, at your job, or with your friends and family?

☐ No problems
☐ A few problems, but overall, I do what I need to do & my performance is not affected
☐ Several problems, but the problems are manageable
☐ A lot of problems that limit what I can do
☐ Extreme problems that keep me from doing almost everything I want to do

5. Has your back shape caused problems with your friends, family members, or dating?

☐ Never ☐ Occasionally ☐ Sometimes ☐ A lot ☐ All the time

If so, how?

____

6. Has your back shape caused problems with your schoolwork, your job, or your ability to do other things that are important to you? (e.g., play sports, be social with your friends)

☐ Never ☐ Occasionally ☐ Sometimes ☐ A lot ☐ All the time

If so, how?

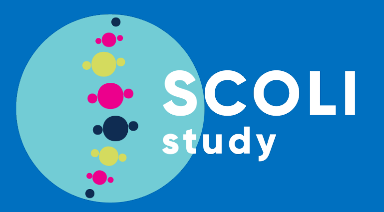
____

7. Do you ever avoid things because of your back shape?

☐ Never ☐ Occasionally ☐ Sometimes ☐ A lot ☐ All the time

If so, how?

____



This section asks questions about your life, health and wellbeing. Remember there are no right or wrong answers, everyone's answers are personal to them.

Part 1. Please tick your answer for each of the following questions about your scoliosis: (SRS-22r)

1. Which of the following best describes the amount of pain you have experienced during the past 6 months? (due to scoliosis)

☐ None ☐ Mild ☐ Moderate ☐ Moderate to Severe ☐ Severe

2. Which of the following best describes the amount of pain you have experienced over the last month? (due to scoliosis)

☐ None ☐ Mild ☐ Moderate ☐ Moderate to Severe ☐ Severe

3. During the past 6 months, have you been a nervous person?

☐ None of the time ☐ A little of the time ☐ Some of the time ☐ Most of the time ☐ All of the time

4. If you had to spend the rest of your life with your back shape as it is right now, how would you feel about it?

☐ Very happy ☐ Somewhat happy ☐ Neither happy nor unhappy ☐ Somewhat unhappy ☐ Very unhappy

5. What is your current level of activity?

☐ Don't get out of bed ☐ Mostly no activity ☐ Light work and light sports ☐ Moderate work and moderate sports ☐ Full activities, no restrictions

6. How do you look in clothes?

☐ Very good ☐ Good ☐ Fair ☐ Bad ☐ Very bad

7. In the past 6 months, have you felt so down in the dumps that nothing could cheer you up?

☐ Very often ☐ Often ☐ Sometimes ☐ Rarely ☐ Never

8. Do you experience back pain when at rest?

☐ Very often ☐ Often ☐ Sometimes ☐ Rarely ☐ Never

9. What is your current level of work/school activity (compared with other people your age)?

☐100% normal ☐75% normal ☐50% normal ☐25% normal ☐0% normal

10. Which of the following best describes the appearance of your trunk; defined as the human body except for the head and limbs?

☐Very good ☐Good ☐Fair ☐Poor ☐Very poor

11. Which one of the following best describes your pain medication use for back pain?

- ☐ None
- ☐ Non-opioid painkillers weekly or less (e.g. paracetamol, Panadol, neurofen)
- ☐ Non-opioid painkillers daily (e.g. paracetamol, Panadol, neurofen)
- ☐ Opioid painkillers weekly or less (e.g. Solpadeine)
- ☐ Opioid painkillers daily (e.g. Solpadeine)

12. Does your back limit your ability to do things around the house?

☐Never ☐Rarely ☐Sometimes ☐Often ☐Very often

13. Have you felt calm and peaceful during the past 6 months?

☐All of the time ☐Most of the time ☐Some of the time ☐A little of the time ☐None of the time

14. Do you feel your back condition affects your personal relationships?

☐None ☐Slightly ☐Mildly ☐Moderately ☐Severely

15. Are you and/or your family experiencing financial difficulties because of your back?

☐Severely ☐Moderately ☐Mildly ☐Slightly ☐None

16. In the past 6 months, have you felt down hearted and blue?

☐Never ☐Rarely ☐Sometimes ☐Often ☐Very often

17. In the last 6 months, have you taken any sick days from work (including household work, or schoolwork) **due to back pain and if so, how many?**

☐0 days ☐1 day ☐2 days ☐3 days ☐4 or more days

18. Does your back condition limit your going out with friends/family?

☐ Never ☐ Rarely ☐ Sometimes ☐ Often ☐ Very often

19. Do you feel attractive with your current back condition?

☐ Yes, very ☐ Yes, somewhat ☐ Neither attractive nor unattractive ☐ No, not very much ☐ No, not at all

20. Have you been a happy person during the past 6 months?

☐ None of the time ☐ A little of the time ☐ Some of the time ☐ Most of the time ☐ All of the time

21. Are you satisfied with how your scoliosis is being managed/treated? *(e.g. appointments, physio, bracing)*

☐ Very satisfied ☐ Satisfied ☐ Neither satisfied nor unsatisfied ☐ Unsatisfied ☐ Very unsatisfied

22. Would you have the same management/ treatment again if you had the same condition?

☐ Definitely yes ☐ Probably yes ☐ Not sure ☐ Probably not ☐ Definitely not

Part 2: Please rate how you feel at the moment by marking an X on the following scales:

1. How **anxious** does your scoliosis make you feel? *(worried, nervous, scared)*

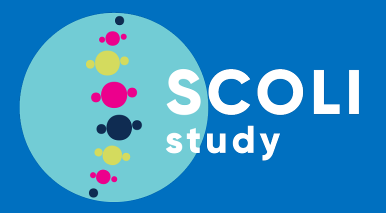
0 1 2 3 4 5 6 7 8 9 10

Not At All — Very Much

2. How **sad or down** does your scoliosis make you feel? *(low mood, unmotivated)*

0 1 2 3 4 5 6 7 8 9 10

Not At All — Very Much



This section asks you about how you cope or manage with your scoliosis. There are no right or wrong answers, we are interested in your personal experience.

Part 1. Please think about times when your scoliosis might bother you or cause you stress. Read each item below and answer them based on how you cope with your scoliosis. (CSI)

	Never	Rarely	Sometimes	Often	Very Often
I work on solving the problems in the situation.	☐	☐	☐	☐	☐
I try to look on the bright side of things.	☐	☐	☐	☐	☐
I let out my feelings to reduce the stress.	☐	☐	☐	☐	☐
I find somebody who's a good listener.	☐	☐	☐	☐	☐
I keep going as if nothing is happening.	☐	☐	☐	☐	☐
I hope a miracle will happen	☐	☐	☐	☐	☐
I feel that I am responsible for my problems and really lecture myself.	☐	☐	☐	☐	☐
I spend more time alone.	☐	☐	☐	☐	☐
I make a plan of action and follow it.	☐	☐	☐	☐	☐
I look at things in a different light and try to make the best of what is available.	☐	☐	☐	☐	☐
I let my feelings out somehow.	☐	☐	☐	☐	☐
I talk to someone about how I'm feeling.	☐	☐	☐	☐	☐
I try to forget the whole thing.	☐	☐	☐	☐	☐
I wish the situation would go away or somehow be over with.	☐	☐	☐	☐	☐
I blame myself.	☐	☐	☐	☐	☐
I avoid my family and friends.	☐	☐	☐	☐	☐

(continued)	Never	Rarely	Sometimes	Often	Very Often
I tackle the problem head on.	☐	☐	☐	☐	☐
I ask myself what's really important, and discover that things aren't as bad as they seem.	☐	☐	☐	☐	☐
I let my emotions out.	☐	☐	☐	☐	☐
I talk to someone I'm very close to.	☐	☐	☐	☐	☐
I don't let it get to me; I refuse to think about it too much.	☐	☐	☐	☐	☐
I wish the situation had never started.	☐	☐	☐	☐	☐
I criticize myself for what happened.	☐	☐	☐	☐	☐
I avoid being with people.	☐	☐	☐	☐	☐
I know what has to be done, so I double my efforts and try hard to make things work.	☐	☐	☐	☐	☐
I convince myself that things aren't quite as bad as they seem.	☐	☐	☐	☐	☐
I get in touch with my feelings and just let them go.	☐	☐	☐	☐	☐
I ask a friend or relative I respect for advice.	☐	☐	☐	☐	☐
I avoid thinking or doing anything about the situation.	☐	☐	☐	☐	☐
I hope that if I wait long enough, things will turn out OK.	☐	☐	☐	☐	☐
Since what happened is my fault, I really give out to myself.	☐	☐	☐	☐	☐
I spend some time by myself.	☐	☐	☐	☐	☐

CONTINUE TO NEXT PAGE →

Part 2. People with scoliosis might think about their condition in different ways. Please read each statement below and tell us how often you think in these ways. (CODI)

	Never	Rarely	Sometimes	Often	Very Often
I am able to manage my scoliosis.	☐	☐	☐	☐	☐
I have got used to my scoliosis.	☐	☐	☐	☐	☐
I cope well with my scoliosis.	☐	☐	☐	☐	☐
I accept my scoliosis.	☐	☐	☐	☐	☐
I take my scoliosis easy (doesn't bother me).	☐	☐	☐	☐	☐
I face my situation with humour.	☐	☐	☐	☐	☐
I don't care about my scoliosis.	☐	☐	☐	☐	☐
I think my scoliosis is no big deal.	☐	☐	☐	☐	☐
I think my scoliosis is not so serious.	☐	☐	☐	☐	☐
I forget about my scoliosis.	☐	☐	☐	☐	☐

Part 3. Please answer these questions about your access to scoliosis information.

1. **Where would you normally get information about scoliosis?** (tick as many that apply)

☐ Parents
☐ A medical professional (for example: doctor, nurses)
☐ Online/ the internet
☐ Other, please specify ___________________________

2(a). Do you ever feel like you need more information about scoliosis?
☐ No, I am happy with the amount of information I have (skip to Q3)
☐ Yes

2(b). If you answered yes, **please tick topics that you would like further information on:**
☐ What is scoliosis?
☐ How to look after myself/ manage my scoliosis
☐ Patient stories
☐ Bracing treatment
☐ Surgery for scoliosis
☐ Other, please describe
____

2(c). Where would you like to get this information?
☐ From your doctor/ nurse/ medical team
☐ Online website
☐ A leaflet
☐ Other __________________________

3. How regularly would you talk about your scoliosis with family members?

☐ Never ☐ Rarely ☐ Sometimes ☐ Often ☐ Very often

4A. Have you ever met another young person with scoliosis?

☐ Yes (in person) ☐ Yes (online) ☐ Never

4B. If so, was this a good experience?

☐ Yes ☐ No

In what way? ____

Part 4. We are learning about how best to support young people with scoliosis.

Please tick how helpful each of the following things would be for you and other young people like you with scoliosis:

	Definitely helpful	Probably helpful	Not sure	Probably not helpful	Definitely not helpful
I would like other people to know more about scoliosis (more awareness about scoliosis in my community).	☐	☐	☐	☐	☐
I would like to learn about how to explain my scoliosis to other people.	☐	☐	☐	☐	☐
I would like to meet other people my age with scoliosis in person.	☐	☐	☐	☐	☐
I would like to interact virtually with other people my age with scoliosis (online chat, video chat, for example).	☐	☐	☐	☐	☐
I would like to meet someone who has already had surgery for their scoliosis.	☐	☐	☐	☐	☐
I would like to participate in an exercise group for people my age with scoliosis (like a swimming club for example).	☐	☐	☐	☐	☐
I would like to talk with a professional about my scoliosis and how I feel (a psychologist, counsellor, therapist).	☐	☐	☐	☐	☐

Do you have any other ideas or suggestions about what would be helpful to young people with scoliosis?

____

____

Final Questions

As a result of the Coronavirus pandemic/ COVID-19 outbreak, life has been a bit different recently. Please answer these last few questions so we can get a more accurate understanding of your life and your wellbeing at the moment:

1. **Overall, how much has the COVID-19 outbreak, and the resulting changes to daily life, affected your life in a negative way? (CASPE)**

☐ Not at all (skip to Q2) ☐ A little ☐ Somewhat ☐ A lot ☐ A great deal

1(a). **What event or change to daily life has been the most negative? (tick up to 3)**

☐ Worried about someone who has or has had the virus
☐ Having to stay at home
☐ Not seeing friends in person
☐ Not going to school
☐ Thinking about how many people are dying because of the virus
☐ Spending more time with family
☐ Increased stress or disorientation from not having a schedule
☐ Not having access to things I need (like food, products)

2. **Overall, how much has the COVID-19 outbreak, and the resulting changes to daily life, affected your life in a positive way?**

☐ Not at all (skip to Q3) ☐ A little ☐ Somewhat ☐ A lot ☐ A great deal

2(a). **What event or change to daily life has been the most positive? (tick up to 3)**

☐ Reduced amount of schoolwork or no schoolwork
☐ Less stress/pressure from school and activities
☐ More time to relax
☐ Getting to do things I don't usually have time for (i.e., art, music, writing, cooking)
☐ Getting more recreational time on the phone/computer (i.e., texting, social media)
☐ Getting to watch more TV/movies
☐ More time to exercise or go outside
☐ Getting more sleep
☐ Spending more time with family
☐ Spending more time with my pet(s)
☐ Not having to have unwanted interactions with other kids at school
☐ Feeling like I have more control in creating my own schedule

3. Has the coronavirus pandemic/covid changed any of the following:

(a) The way you feel about your general appearance

☐ No, I feel the same
☐ Yes, I feel better about my appearance
☐ Yes, I feel worse about my appearance

(b) The way you feel about the appearance of your scoliosis

☐ No, I feel the same
☐ Yes, I feel better about the appearance of my scoliosis
☐ Yes, I feel worse about the appearance of my scoliosis

(c) The way you cope with your scoliosis

☐ No, I cope the same as always
☐ Yes, I have been coping better with my scoliosis
☐ Yes, I have been coping worse with my scoliosis

(d) Your level of pain (scoliosis-related)

☐ No, my pain is the same
☐ Yes, my pain is not as bad
☐ Yes, my pain is worse
☐ Not applicable to me

(e) Your ability to function/do activities

☐ No, I function about the same
☐ Yes, I have been better able to do work and sports
☐ Yes, I have been less able to do work and sports

(f) Your happiness/ mental wellbeing

☐ No, I feel the same
☐ Yes, I feel better
☐ Yes, I feel worse

Thank You for Completing this Survey!

Appendix F: Exploratory *t*-Test Results for Gender Differences on Key Survey Variables

Table F.1

Exploratory t-Test Results for Gender Differences on Key Survey Variables

	Female		Male		Mean Difference	*df*	*t*	*p*
	M	*SD*	*M*	*SD*				
Quality of Life (SRS)	3.41	0.93	3.82	0.58	-0.41	28	-1.45	.158
Function	4.07	0.97	4.33	0.64	-0.27	28	-0.89	.380
Pain	3.48	1.15	3.95	0.65	-0.47	28	-1.37	.181
Self-Image	2.95	1.02	3.48	0.74	-0.53	28	-1.64	.111
Mental Health	3.15	1.04	3.52	0.86	-0.37	28	-1.07	.294
Body Image Disturbance (BIDQS)	2.60	1.01	1.93	0.79	0.67	28	2.02	.054
Appearance Satisfaction (BESAA)	1.83	1.15	2.54	0.69	-0.71	27	-1.99	.057
Appearance Investment (MBRSQ)	3.69	0.76	2.82	0.89	0.86	27	2.84	.008
Perceived Visibility Coping (CSI)	1.27	0.88	1.07	0.46	0.20	21	0.78	.445
PF Engagement	3.38	0.88	3.36	1.01	0.03	28	0.07	.943
EF Engagement	3.34	0.65	3.43	1.04	-0.08	28	-0.83	.794
PF Disengagement	3.16	0.59	3.28	0.52	-0.13	28	-0.62	.544
EF Disengagement (CODI)	2.59	1.26	1.97	0.60	0.63	20.06	1.74	.098
Acceptance	3.59	0.94	3.71	1.08	-0.12	28	-0.33	.744
Distance	2.10	0.83	2.35	1.19	-0.25	24.93	-0.66	.511

Appendix G: Use of Cobb Angle Measurements

The Cobb angle is the degree of a scoliosis spinal curvature, used to quantify the magnitude of the curvature and to track progression. Clinicians assess the Cobb angle using radiographs, and of note, there is a 5° margin of error in calculation. Widely accepted categories are mild (10° – 24°), moderate (25° - 44°), and severe (45°/50°+). Some patients have two curvatures (e.g. S shape curves). The Cobb angle is the measurement of the major curve (i.e. the largest).

Cobb Angle Data Qualitative Study – Comparison of Self-Report & Clinician Assessed

For the qualitative study (Chapter 3) Cobb angles were measured by the orthopaedic surgeon using the closest available scan to the date of the participants' interview. Self-reported Cobb angles were also collected from participants in this qualitative study for comparison.

Self-reported Cobb angle measurements ranged from 33° – 89° (*M* = 59.9°, *SD* = 13.28) and the clinician measurements ranged from 46° – 100° (*M* = 68.07°, *SD* = 14.84). An exploratory correlation analysis demonstrated a positive relationship between the two sources of Cobb angle measurements (r = 0.83). Discrepancies between self-reported and clinician measurements ranged from 1° – 27° (*M* = 9°, *SD* = 6.7). In 13 of 14 cases, self-report Cobb angles were smaller than the clinician measurement. Potential reasons for discrepancies include:

(i) Some discrepancy may be attributable to the 5°± margin of error. The clinician assessment of Cobb angles was conducted retrospectively for the study. The Cobb angle discussed with participants at their clinic appointments prior to interview was therefore measured at a different time and is subject to the margin of error.

(ii) Ideally, clinician assessment would have used a radiograph taken *prior* to the participant's interview as the most recent x-ray assessment would be what the participant is likely to recall. This was the case for 10 participants, as a radiograph was available 1 – 2 months prior to interview. For these participants, self-report and clinician measurements were more similar, as the average discrepancy was 7.2° (range 1°-13°). For 4 participants, radiographs 3 – 6 months post interview were available and may not be as appropriate. These participants could have accessed private scans explaining why there was not a recent radiograph available on the hospital system.

Appendix H: Data Screening for Hierarchical Regression Analyses

All skewness and kurtosis values fell within an acceptable range of +1/-1 (range -0.717 – 0.830) and kurtosis values within an acceptable range of +3/-3 (range -0.626 – 0.496) with the exception of one variable. This was Perceived Visibility (skewness = 1.289, kurtosis = 3.149). A square root transformation was applied to the Perceived Visibility variable to obtain a nearer-normal distribution (resulting skewness = 0.5, kurtosis = 2.345). As a one item indicator this variable is unlikely a robust measure, however it was retained for analysis as a novel concept of interest to be examined in this sample. On inspection of the histograms, the function subscale of the SRS-22r quality of life measure appeared negatively skewed, with a clustering of scores toward the higher end of functional ability. This could be expected due to a limited number of participants with highly severe curvatures which would indicate a lower likelihood of functional disability in our sample (Asher et al., 2006). However, skewness and kurtosis values fell within the acceptable range and the overall SRS-22r scores appeared normally distributed (skewness = -.643, kurtosis = -.003). Post analyses, inspection of the normal probability plots of the regression standardised residuals also indicated no major deviations from normality and the scatterplots demonstrated appropriate homoscedasticity.

The data were screened for univariate and multivariate outliers. Box plots, histograms, and standardised *z*-scores were inspected for independent and dependent variables to be used in the regressions. Prior to analysis, a small number of suspected outliers were examined but on inspection, no cases had *z*-scores exceeding the accepted cut-off of >3.29 (Tabachnick & Fidell, 2014, p. 107), and trimmed mean values were not substantially altered with removal of outliers. Therefore, a conservative approach was taken and these cases were retained for analysis. Post analyses, standardised residuals and Mahalanobis distance values were examined indicating no concerns regarding multivariate outliers for regression 1. For regression 2 with BIDQ-S as the dependent variable, one case with a standardised residual exceeding 3.29 was removed. For regression 2 with BESAA-A as the dependent variable, there were no concerns regarding multivariate outliers.

A review of associations between the independent variables showed no potential issues with multicollinearity for Regression 1 (Pearson's $r < .538$). The tolerance and variance inflation

factor (VIF) values fell within an acceptable range with tolerance > .1 and VIF < 10 (tolerance range = .528 - .935; VIF range = 1.07 – 1.89). For Regression 2, there were relatively strong Pearson correlations between the Trunk Appearance Perception (TAPS) scores and Perceived Visibility (*r* = -.662), and between TAPS scores and Curve Size (*r* = -.617). These correlations are not surprising given that each of these variables assessed a perspective on scoliosis appearance. While these correlations were notable, the tolerance and VIF values were within acceptable ranges indicating no serious concerns for multicollinearity (tolerance range = .424 - .921; VIF range = 1.08 – 2.36).

Appendix I: Expanded Correlation Table for Demographic & Clinical Variables, Coping Strategies, and HRQOL

Table I.1 *Pearson Correlations Among Demographic & Clinical Variables, Coping Strategies, and HRQOL*

Measure	1	2	3	4	5	6	7	8	9	10	11	12	13
1. Age	-												
2. Curve Size	-.048	-											
3. Length Diagnosis	.538***	-.024	-										
4. Brace (Yes/No)[a]	.380***	.206*	.031	-									
5. CSI PF[b] Engagement	-.046	-.278**	-.008	-.262**	-								
6. CSI EF[c] Engagement	.048	-.155	.022	-.075	.363***	-							
7. CSI PF Disengagement	.038	.242*	.019	-.007	-.243**	-.195*	-						
8. CSI EF Disengagement	.199*	.080	.272**	.213*	-.343***	-.365***	.369***	-					
9. SRS Quality of Life Total	-.270**	-.368***	-.284**	-.331**	.490***	.287**	-.446***	-.616***	-				
10. SRS Function	-.197*	-.264**	-.217*	-.266**	.343***	.204*	-.415***	-.479***	.853***	-			
11. SRS Pain	-.253**	-.327***	-.318**	-.272**	.352***	.261**	-.275**	-.486***	.871***	.742***	-		
12. SRS Self-Image	-.191*	-.406***	-.175	-.290**	.485***	.312**	-.459***	-.561***	.839***	.624***	.620***	-	
13. SRS Mental Health	-.259**	-.241*	-.233*	-.285**	.466***	.188*	-.367***	-.544***	.801***	.538***	.549***	.589***	-

*** p < .001, ***p* < .01; **p* < .05 (two-tailed).[a]Point biserial correlation reported for dichotomous variable. [b]PF = Problem Focused, [c]EF = Emotion Focused.
Note. Cohen's Pearson *r* effect size guidelines: small = 0.10, medium = 0.30, large = 0.50.

Appendix J: Expanded Regression Output Tables for SRS-22r Subscales

Table J.1

Hierarchical Regression Analysis Predicting Function (SRS-22r)

Variable	*B*	*SE B*	95% CI	β	ΔR^2	R^2
Step 1					.16**	.16
Age	-.00	.04	-.08, .07	-.01		
Curve size	-.01	.00	-.02, -.00	-.23*		
Length diagnosis	-.07	.04	-.16, .01	-.19		
Brace	-.33	.16	-.65, -.02	-.21*		
Step 2					.21***	.37
Age	-.01	.03	-.07, .06	-.02		
Curve size	-.00	.00	-.01, .00	-.12		
Length diagnosis	-.04	.04	-.12, .04	-.11		
Brace	-.24	.15	-.53, .06	-.15		
PF Engagement	.12	.09	-.07, .29	.12		
EF Engagement	.01	.08	-.16, .17	.01		
PF Disengagement	-.31	.10	-.51, -.10	-.28**		
EF Disengagement	-.22	.08	-.39, -.06	-.27**		

*** $p < .001$, ** $p < .01$, * $p < .05$. *Note.* Adjusted R^2 for Step 1 =.13, Step 2 = .33.
Step 1: $F(4, 107) = 5.116$, $p = .001$. **Step 2:** $F(8, 103) = 7.673$, $p < .001$.
CI = confidence interval (lower limit, upper limit).

Table J.2

Hierarchical Regression Analysis Predicting Pain (SRS-22r)

Variable	*B*	*SE B*	95% CI	β	ΔR^2	R^2
Step 1					.25***	.25
Age	-.01	.04	-.09, .07	-.02		
Curve size	-.01	.00	-.02, -.01	-.29**		
Length diagnosis	-.15	.05	-.25, -.05	-.31**		
Brace	-.35	.18	-.71, .01	-.19		
Step 2					.15***	.40
Age	-.02	.04	-.10, .06	-.05		
Curve size	-.01	.00	-.02, -.00	-.24**		
Length diagnosis	-.10	.05	-.19, -.01	-.20*		
Brace	-.18	.18	-.53, .16	-.10		
PF Engagement	.14	.11	-.08, .36	.12		
EF Engagement	.07	.10	-.13, .27	.06		
PF Disengagement	-.10	.13	-.35, .15	-.07		
EF Disengagement	-.30	.10	-.49, -.11	-.26**		

*** $p < .001$, ** $p < .01$, * $p < .05$. *Note.* Adjusted R^2 for Step 1 =.23, Step 2 = .36.
Step 1: $F(4, 107) = 9.095$, $p < .001$. **Step 2:** $F(8, 103) = 8.712$, $p < .001$.

Table J.3

Hierarchical Regression Analysis Predicting Self-Image (SRS-22r)

Variable	*B*	*SE B*	95% CI	β	ΔR^2	R^2
Step 1					.25***	.25
Age	-.02	.04	-.09, .06	-.05		
Curve size	-.02	.00	-.02, -.01	-.38***		
Length diagnosis	-.07	.05	-.16, .02	-.16		
Brace	-.31	.16	-.64, .02	-.18		
Step 2					.30***	.55
Age	-.02	.03	-.08, .04	-.07		
Curve size	-.01	.00	-.02, -.00	-.26**		
Length diagnosis	-.02	.04	-.09, .05	-.05		
Brace	-.13	.14	-.40, .14	-.08		
PF Engagement	.23	.09	.06, .39	.21**		
EF Engagement	.04	.08	-.12, .19	.03		
PF Disengagement	-.28	.10	-.47, -.09	-.22**		
EF Disengagement	-.31	.08	-.46, -.16	-.34***		

*** $p < .001$, ** $p < .01$, * $p < .05$. *Note.* Adjusted R^2 for Step 1 =.22, Step 2 = .51.
Step 1: $F(4, 107) = 8.679$, $p < .001$. **Step 2:** $F(8, 103) = 15.616$, $p < .001$.

Table J.4

Hierarchical Regression Analysis Predicting Mental Health (SRS-22r)

Variable	*B*	*SE B*	95% CI	β	ΔR^2	R^2
Step 1					.18***	.18
Age	-.05	.04	-.13, .03	-.15		
Curve size	-.01	.00	-.02, -.00	-.21*		
Length diagnosis	-.06	.05	-.15, .04	-.12		
Brace	-.35	.18	-.71, .01	-.19		
Step 2					.30***	.48
Age	-.06	.03	-.13, .01	-.17		
Curve size	-.00	.00	-.01, .00	-.10		
Length diagnosis	-.00	.04	-.09, .08	-.01		
Brace	-.12	.16	-.43, .19	-.07		
PF Engagement	.34	.10	.14, .53	.29**		
EF Engagement	-.09	.10	-.27, .09	-.08		
PF Disengagement	-.23	.11	-.45, -.01	-.16*		
EF Disengagement	-.36	.09	-.53, -.19	-.37***		

*** $p < .001$, ** $p < .01$, * $p < .05$. *Note.* Adjusted R^2 for Step 1 =.15, Step 2 = .44.
Step 1: $F(4, 107) = 5.725$, $p < .001$. **Step 2:** $F(8, 103) = 11.720$, $p < .001$.

Appendix K: Search String for Systematic Review

Database	Search String	Limits
MEDLINE; PsycINFO; CINAHL Plus; EMBASE; SCOPUS	AB,TI(parent* OR famil* OR mother* OR father*) AND AB,TI(scoliosis) AND AB,TI(child* OR adolesc* OR son* OR daughter* OR patient*) AND AB,TI(experience* OR perspective* OR perception* OR assessment* OR concern*)	Since 2000 English language

AB: Abstract; TI: Title.

Appendix L: Summary of Studies Included in the Systematic Review

Citation	Parents Age (*M*) Gender	Children Age (*M*) Gender	Treat-ment Status	Study Design	Measures used	Findings
Information Needs						
Khetani 2008 Canada	n=30 - -	n=34 11-18 (14.7) 94% F	Braced & Post-op	Survey	Scoliosis Knowledge Questionnaire	Parents scored higher than children on the scoliosis knowledge survey. >70% of parents incorrectly answered questions on curve progression, radiation exposure, non-union, and long-term postsurgical concerns.
Baker 2012 Ireland	n=168 - 81% F	- 1-19 (11.9) -	All outpatie nts	Survey	Internet Usage Questionnaire	58% of parents reported using the internet for scoliosis information. 77% of responses indicated that the internet was helpful. 29% of responses showed that the internet created more anxiety for parents, while 30% encountered confusing websites.
Lysenko 2016 Canada	n=71 (45.9) -	n=74 10-18 (14.3) 90.5% F	Pre-op	Case series	Scoliosis Knowledge Questionnaire; MIQ; CHIP	91.2% parents previously searched online for scoliosis information, with 63.9% rating it as somewhat helpful and 18.1% as very helpful. Scoliosis knowledge improved after exposure to website intervention ($p < .05$), while parents also demonstrated a small increase in negative attitude toward illness and a small decrease in positive attitude towards illness ($p < .05$).
Schwieger 2016 USA	n=300 - 92% F	- - -	All stages	Content analysis	Purpose-made coding frame	Parents with newly diagnosed children were most likely to seek information. Parents most often exchanged information about the causes and progression of scoliosis (48% of online posts), brace types (25%), brace effectiveness (21%), and doctors/hospitals (21%).
Treatment Concerns						
Bridwell 2000 USA	n=93 - 82% F	N=91 9-18 82% F	Pre-op	Survey	Concerns and Preferences Questionnaire	Neurologic deficit, pseudarthrosis, and wound infection were ranked as greatest surgical concerns. Main reasons/expectations for surgery were to prevent scoliosis progression and future pain, and cosmetic correction. Patients ranked postsurgical lifestyle adjustment as more concerning and return to function as more important than parents ($p < .05$).

(contd.) Citation	Parents Age (*M*) Gender	Children Age (*M*) Gender	Treat-ment Status	Study Design	Measures used	Findings
Donnelly 2004 USA	n=10 - 80% F	n=12 13-18 100% F	Braced & Post-op	Qualitative	Semi-structured Interview	*Decision making*: Parents felt they had a responsibility to proceed with bracing as a less invasive treatment over surgery. *Effect on life & compliance with brace wear*: Parents reported that brace fitting was traumatic, fights with child over brace wear, difficulty finding clothing, and a difficult recovery from surgery.
Salisbury[a] 2007 USA	n=92 - 100% F	- (13.6) 80.4% F	Pre – to post-op	Mixed methods	Semi-structured interview; WCQ	Presurgical stressors (in order): *parental role loss*, *possibility of poor surgical outcomes, uncertainty about recovery, pain,* and *accepting reality.* Postsurgical stressors (in order): *pain*, *parental role loss*, *uncertainty about recovery*, *concerns about care*, and *complications* Most frequent pre- and postsurgical coping strategies were social support, positive reappraisal, and planful problem solving.
Narayanan 2008 Canada	n=55 - -	n=55 12-18 (14.3) 87% F	Post-op	Survey	Concerns/ Expectations Questionnaire	Parents had higher levels of overall concern about surgery than children (p = .001). 91% parents were very or extremely concerned about child's appearance pre-op. Highest surgical concerns were pain (M = 4.45/5), stiffness (M = 3.98), & paralysis (M = 3.91). Strongest desires were to prevent deformity progression (M = 4.6/5), prevent future health problems (M = 4.38), & reduce pain (M = 4.25).
Bull & Grogan[b] 2010 UK	n=13 (46) 91.6% F	- 3.5-18 (12.9)[c] 92% F	Post-op	Qualitative	Semi-structured Interview	Themes: 1. *Information* (parents lacked knowledge, often used the internet); 2. *Parenting role* (fear/anxiety related to diagnosis and treatment threatened protector role); 3. *Confidence in professionals* (importance of relationship with consultant & nursing team); 4. *Pain* (pain management was major source of stress); 5. *Effect on life* (living in a state of uncertainty & emotional upheaval).

(contd.) Citation	Parents Age (*M*) Gender	Children Age (*M*) Gender	Treat-ment Status	Study Design	Measures used	Findings
Chan 2017 USA	n=48 - -	n=48 10-19 (14.2) 90% F	Pre-op	Survey	Concerns questionnaire	Parents had higher presurgical concern (M = 6.9/10) than children (M = 4.6). Age of child was correlated with parents level of concern (r = -.37, p = .009). Parents listed top three surgical concerns as pain, neurologic injury, and amount of correction. Patients listed pain, ability to return to activities, and neurologic injury.
Lonner 2020 USA	n=44 - -	n=44 10-18 (14.3) 84% F	Pre-op	Survey	PGI	Parents' primary operative aspirations were to improve sleep, self-esteem, and pain, while patients most wanted to improve pain, self-esteem, and sports. Parents reported they would be most regretful about surgery if sleep (M = 8.6/10), future health (M = 7.3), and appearance (M = 6.75) were not improved.
Psychological Wellbeing						
LaMontagne 2001 USA	n=74 - -	n=74 11-18 74% F	Pre- to post-op	Case series	STAI; VAS-P	Parents' anxiety decreased from the presurgical clinic visit to the second postsurgical day ($p < .01$). Parents' and children's anxiety positively correlated at presurgical ($r = .28$, $p = .01$) and postsurgical ($r = .32$, $p > .01$) timepoints, but parents' anxiety was not related to children's pain.
LaMontagne 2003 USA	n=60 - 100% F	- (14) 85% F	Pre- to post-op	Case series	LOT; STAI; WCQ	Parental anxiety decreased from pre- to post-op ($p < .01$), still moderately high. Seeking social support was the most common coping strategy used at pre- and postsurgical timepoints, followed by positive reappraisal and planful problem solving.
Kasai 2006 Japan	n=30 (40.4) 100% F	n=30 10-26(15.1) 80% F	Pre- to post-op	Case series	MPI	Parents reported reduced neurotic tendencies from the presurgical period to one year after surgery ($p < .001$). No significant correlations were identified between parents and patients psychological states (extroversion, neuroticism).

(contd.) Citation	Parents Age (*M*) Gender	Children Age (*M*) Gender	Treat-ment Status	Study Design	Measures used	Findings
Flynn 2007 UK	n=41 (47.2) 90% F	- (17.3) 75% F	All stages	Survey	FDC	Parents most frequently mentioned predominant life stressor was the health of their child with scoliosis (constituting 34% of all 103 life stressors listed). Three most frequently reported coping styles for parents were emotional social support, instrumental social support, and optimism.
Hines 2015 USA	n=54 - 78% F	n=54 9-17(14.1) 74% F	Referred to clinic	Non-randomized cross-sequential	STAI	Parents and children referred from school scoliosis screening experienced elevated levels of anxiety pre-appointment compared to controls (p = .02; p = .04). Those diagnosed with scoliosis continued to report elevated anxiety levels post-appointment while anxiety declined for those not diagnosed. Over 70% of parents rated the SSS evaluation as helpful.
Kwan 2016 Malaysia	n=96 - 51% F	- - -	Pre- to post-op	Non-randomized ed cohort	VAS-A; HADS	Intraoperative text messages (SMS) were effective in reducing parental anxiety during children's spinal fusion surgery. Compared to controls, parents receiving SMS had significantly lower anxiety during surgery ($p < .05$) and one day post-surgery ($p < .05$).
Wang 2019 China	n=64 (42.4) 73% F	n=64 11-18(14.3) 92% F	Braced & pre-op	Survey	PHQ-9; GAD-7	Parents of children with AIS were more likely to report moderate to severe levels of depressive symptoms and generalized anxiety compared to a controls (14.1% vs. 4.7%, p = 0.045, & 14.1% vs. 3.5%, p = 0.019, respectively). Parent and child depression & anxiety scores were correlated (r = .45 - .5, p < .01). Scoliosis Cobb angle ≥50°, child depression, and low parental education level were independent risk factors for parental depressive symptoms.

Studies are placed under theme they aligned with most and ordered by date of publication. [a] Contributed to treatment concerns and psychological well-being. [b] Contributed to treatment concerns and information needs. [c] 2 of 13 cases were early onset scoliosis. Abbreviations: MIQ: Meaning of Illness Questionnaire; CHIP: Coping Health Inventory for Parents; WCQ: Ways of Coping Questionnaire; PGI: Patient Generated Index; STAI: State-Trait Anxiety Inventory; VAS-P/A: Visual Analogue Scale for Pain/Anxiety; LOT: Life Orientation Test; MPI: Maudsley Personality Inventory; FDC: Functional Dimensions of Coping; HADS: Hospital Anxiety and Depression Score. PHQ-9: Patient Health Questionnaire; GAD-7: Generalized Anxiety Disorder Scale.

Appendix M: MMAT Assessment Summary

Table L.1. *Summary of the MMAT Assessment for Systematic Review*

Study	Screen		Qualitative					Quantitative Non-Randomized					Quantitative Descriptive					Mixed Methods				
	S1	S2	1.1	1.2	1.3	1.4	1.5	3.1	3.2	3.3	3.4	3.5	4.1	4.2	4.3	4.4	4.5	5.1	5.2	5.3	5.4	5.5
Donnelly et al., 2004	✓	✓	✓	✓	✓	✓	✓															
Bull & Grogan, 2010	✓	✓	✓	✓	✓	✓	✓															
Hines et al., 2015	✓	✓						✓	✓	✓	✓	✓										
Kwan et al., 2016	✓	✓						✓	✓	✓	×	✓										
Bridwell et al., 2000	✓	✓											✓	✓	✓	✓	✓					
LaMontagne et al., 2001	✓	✓											✓	--	✓	✓	✓					
LaMontagne et al., 2003	✓	✓											✓	✓	✓	×	✓					
Kasai et al., 2006	✓	✓											✓	✓	--	×	✓					
Flynn 2007	✓	✓											✓	✓	--	--	✓					
Khetani et al., 2008	✓	✓											✓	--	✓	✓	✓					
Narayanan 2008	✓	✓											✓	✓	✓	✓	✓					
Baker et al., 2012	✓	✓											✓	✓	✓	×	✓					
Lysenko et al., 2016	✓	✓											✓	--	✓	×	✓					
Schwieger et al., 2016	✓	✓											✓	--	✓	✓	✓					
Chan et al., 2017	✓	✓											✓	--	✓	✓	✓					
Wang et al., 2019	✓	✓											✓	✓	✓	✓	✓					
Lonner et al., 2020	✓	✓											✓	--	✓	✓	✓					
Salisbury et al., 2007	✓	✓																--	✓	✓	✓	✓

MMAT: Mixed Methods Appraisal Tool. As no randomized controlled trials were identified in this review, the '2. Quantitative Randomized Controlled Trials' category was omitted from this table.'✓' indicates 'criterion was met'; '×' indicates 'criterion was not met'; and '--' indicates 'unclear information related to the criterion'. **Issues Identified:** 3.4: Potential confounders not accounted for in study design; 4.2: Details of parent participants (gender) not reported; 4.3: Justification for measures used not provided; 4.4: Risk of non-response bias; 5.1: Rationale for study design not reported.

Table L.2. *MMAT Appraisal Criteria (adapted from from Hong et al., 2018)*

Category	Quality Criteria
Screening (all studies)	S1. Are there clear research questions? (*Clear study aim presented)* S2. Do the collected data allow the research questions to be addressed? (*The data relate to the study aim)*
1.Qualitative	1.1. Is the qualitative approach appropriate to answer the research question? (*Approach named and justified)* 1.2. Are the qualitative data collection methods adequate to address the research question? (*Appropriate method described, interview schedule outlined, form of data named, adequate description of sample)* 1.3. Are the findings adequately derived from the data? (*Appropriate analysis described)* 1.4. Is the interpretation of results sufficiently substantiated by data? (*Adequate reference to quotations)* 1.5. Is there coherence between qualitative data sources, collection, analysis and interpretation?
3. Quantitative Non-Randomized	3.1. Are the participants representative of the target population? (*Adequate description of the sample and recruitment process)* 3.2. Are measurements appropriate regarding both the outcome and intervention? (*Measures are justified and appropriate)* 3.3. Are there complete outcome data? (*Were there any participant dropouts in the study)* 3.4. Are the confounders accounted for in the design and analysis? (*Relevant confounders considered and accounted for)* 3.5. During the study period is the intervention administered as intended? (*Were any changes reported in the intervention)*
4. Quantitative Descriptive	4.1. Is the sampling strategy relevant to address the research question? (*Is the source of recruitment appropriate e.g. hospital clinic)* 4.2. Is the sample representative of the target population? *(Eligibility criteria; Clear description of the sample, Recruitment process stated)* 4.3. Are the measurements appropriate? (*Measures are justified and appropriate)* 4.4. Is the risk of nonresponse bias low? *(Consider missing data, did study consider differences between those who completed and those who did not, or reasons for non-completion; For case series, is there complete data)* 4.5. Is the statistical analysis appropriate to answer the research question? (*Statistics adequately described and justified)*
5. Mixed Methods	5.1. Is there an adequate rationale for using a mixed methods design to address the research question? (*Explanation for using mixed methods)* 5.2. Are the different components of the study effectively integrated to answer the research question? (*Consideration of how and when the data was brought together)* 5.3. Are the outputs of integration of the components adequately addressed? *(Interpretation of findings shows value of mixed methods)* 5.4. Are divergences and inconsistencies between the components adequately addressed? 5.5. Do the different components of the study adhere to the quality criteria of each method? (*Are there important threats to trustworthiness of either component? (consult other relevant sections of the MMAT i.e. 1 & 4))*