A systematic scoping review and textual narrative synthesis of the qualitative evidence related to adolescent idiopathic scoliosis

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Abstract

Introduction: This systematic scoping review sought to summarise and synthesise the qualitative evidence on adolescent idiopathic scoliosis (AIS) to understand the experiences of AIS, and of its diagnosis and treatment, and effective coping strategies to inform directions for future research.

Method: A systematic scoping review and textual narrative synthesis was undertaken. Qualitative or mixed-methods studies with extractable qualitative data were included if participants had AIS, and patient or patient relative perspectives about AIS or its treatment were reported.

Results: Fifteen papers were included. These suggested that AIS can influence self-image and perceptions of appearance and can cause those with the condition to reluctantly adjust parts of their lives and avoid some activities. The diagnosis and treatment of AIS is a major event and is often accompanied by shock, uncertainty, and anxiety. Whilst some find surgery stressful, others were relieved to have the surgery, and were generally happy with the outcome. Immediate family members may be distressed by the diagnosis and treatment of AIS.

Discussion: AIS diagnosis and treatment has a substantial impact on adolescents and their families that is only addressed in part in this review. There is a pressing need for more qualitative research to understand needs, perspectives and experiences of adolescents with AIS and their families from the point of diagnosis, throughout treatment, and in the longer-term.

Introduction/Background

Scoliosis is the most common spinal disorder in children and adolescents. It is most likely to develop during adolescence, defined by the World Health Organisation (WHO) as the 'phase of life between childhood and adulthood, from ages 10 to 19' when there is 'rapid physical, cognitive and psychosocial growth' (WHO 2021). Adolescent scoliosis is described as 'idiopathic' because it arises spontaneously and the cause is, as yet, unknown. Adolescent Idiopathic Scoliosis (AIS) has a global prevalence of 0.47 – 5.2% and affects more girls than boys (Erwin et al., 2020; Konieczny et al., 2013). The condition can cause thoracic, lumbar or thoracolumbar curvatures and twists, often described as a three-dimensional curve of the spine. The size of the curve is reported as the Cobb angle, and surgery is usually offered when this reaches 50°in the thoracic spine, and 40 in the lumbar spine (Bettany-Saltikov et al., 2015).

Where intervention is required, treatment of AIS generally involves either bracing or surgery. The literature suggests that surgical outcomes are generally good, with minimal risks to patients (for example, Kepler et al., 2012; Lykissas et al. 2013). However, several gaps exist in the evidence. Less is known about longer term non-clinical outcomes of AIS, particularly regarding health-related quality of life. In our recent systematic review of quantitative evidence of longer-term outcomes in AIS (Essex et al. (2021) we found that the majority of included studies generally reported good quality of life (QoL) of participants, yet almost all studies addressed this QoL in relation to surgical outcomes, and relied on the same one or two instruments for measuring QoL. The review was critical of this, noting that relying on a small number of instruments to measure health related quality of life risks oversimplifying the concept that has otherwise been seen as dynamic, relational, and largely unsettled in the broader literature (Ferrans et al. 2005; Olsen and Misajon, 2020). Furthermore, the results of our earlier review also appeared to contrast sharply with the minimal qualitative literature on AIS, which presents a far more nuanced picture of perceptions of AIS, its treatment and quality of life throughout this process. In a trial feasibility study related to the use of exercise to treat AIS, Toye et al. (2016) concluded that including a qualitative element in their study yielded a range of benefits including contextualising research findings and providing a richer understanding of participant experiences of AIS. Further qualitative research may be needed in this area to complement the largely deductive approach inherent in quantitative research. To confirm this need, we aimed to provide a more rounded assessment of the qualitative evidence related to quality of life and patient perceptions and experiences of AIS.

The aim of this scoping review was therefore to summarise and synthesise the qualitative evidence related to AIS, to identify any knowledge gaps and to determine directions for future research. More specifically this review sought to explore and synthesise this literature to better understand 1) the experiences of AIS amongst adolescents and their parents, 2) the experiences of the diagnosis and treatment of AIS and 3) best practice and coping strategies reported in the literature.

Method

Design

A systematic scoping review was utilised to examine the extent, range, and nature of research activity with the goal of locating, appraising, and synthesizing all relevant qualitative evidence related to AIS, and to identify gaps in the existing literature. Thus, we aimed to examine all available qualitative papers, and to include mixed methods papers where there was distinct and extractable qualitative data. A scoping review was employed because of the perceived paucity of evidence in this area and as a means to map any available evidence, identify its strengths and weaknesses, and determine directions for future research (Pollock et al., 2021). Foundational concepts and evidence can be mapped, allowing for examination of practice, policy, and research and gaps in evidence and policy can be identified. The results of scoping reviews can provide indications for where further research may be required and inform the development of these research endeavours.

This review was informed by the scoping review methodology described by Levac and colleagues (2010) and involved seven steps: 1) formulating a review question; 2) developing a search strategy and conducting the search; 3) defining inclusion and exclusion criteria; 4) selecting studies; 5) extracting data; 6) summarising studies; and 7) analysing, synthesising and interpreting results. PRISMA and ENTREQ reporting guidelines have been followed (Moher et al., 2009; Tong et al., 2012).

Formulating a review question

Based on our earlier work and knowledge of the literature from a preliminary screen of the databases, our review question was: ‘What is the available qualitative evidence on the experiences of adolescents and their parents, of diagnosis, treatment and outcomes of adolescent idiopathic scoliosis?'

Search Methods

A search was conducted on 05/05/21. Five reference databases were searched: Medline, CINAHL, Web of Science, PsycInfo and Scopus. Preliminary search terms were developed to reflect the core concepts - the population of interest (those diagnosed with AIS), and the type of research we were interested in (qualitative research studies). The final search terms were ("adolescent idiopathic scoliosis" OR "idiopathic scoliosis") AND (qualitative OR interview OR "focus group" OR "case report" OR "case study" OR ethnograph* OR phenomenolog* OR "grounded theory").

Inclusion and Exclusion Criteria

The inclusion and exclusion criteria were developed to enable inclusion of all available qualitative evidence, whilst keeping the focus on AIS:

Inclusion

- Studies that employed qualitative designs, or mixed-methods designs with extractable qualitative data
- Studies which included patient or patient relative (i.e. parent) perspectives about AIS, its treatment or impact
- Studies that included participants with AIS (not more general spinal surgery or back pain, for example, nor congenital scoliosis, or that related to syndromes including Ehlers-Danlos Syndrome)
- Studies published after the year 2000 (to include modern rod and screw or fusion techniques and exclude older surgical techniques)

Exclusion

- Studies that employed quantitative designs
- Case studies with little data from a patient (or relative) perspective
- Studies published in a language other than English
- Books or conference abstracts

Selecting studies

A total of 1441 studies were identified, which was reduced to 1359 after duplicates were removed. A first screen by two authors (RE and LD) examining titles and abstracts reduced this to 31 papers. The reference lists of these papers were searched, revealing another 13 papers. A total of 44 papers were assessed against the below inclusion and exclusion criteria by two authors (RE, LD) resulting in 15 papers selected for the review (Figure 1).

Data extraction

Data were extracted by all authors; each author worked with four or five papers, so that each paper was reviewed twice, ensuring robustness in the process; data were categorised according to the source, country of where the research took place, study aims and objectives, research methods/design and sample information and main outcomes (see Table 1). These results were then collated by RE and LD. Where there were significant disagreement or discrepancies, authors were consulted to reach a common understanding regarding the study in question.

Data summary and synthesis

Due to heterogeneity across studies and even within similar study methodologies, a meta-synthesis for qualitative data was not possible. Instead, studies were combined to summarise the study characteristics, followed by a textual narrative synthesis. This approach arranges disparate study types into more homogenous sub-groups which aids in the synthesising of different types of evidence. Study characteristics, context, quality, and findings are reported according to a standard format, and similarities and differences are compared across studies (Lucas et al., 2007).

Results

This section reflects the final stage described by Levac et al. (2010) of analysing, synthesising and interpreting results. Fifteen studies were included in this review; these were conducted in Canada (MacCulloch et al., 2009), China (Du et al., 2016), Greece (Sapountzi-Krepia et al., 2006), Hong Kong (Law et al., 2017), Poland (Grantham et al., 2019), South Africa (Naidu & Shabangu., 2015), Spain (Carrasco & Ruiz, 2016), Sweden (Rullander et al., 2013; Rullander et al., 2017), Turkey (Bilik et al., 2018), United Kingdom (Honeyman & Davison., 2016), and the United States of America (Donnelly et al., 2004; Klieber & Adamek., 2012; Merenda et al., 2011; Salisbury & LaMontagne., 2007). Eleven studies were qualitative (Bilik et al., 2018;

Carrasco & Ruiz, 2016; Donnelly et al., 2004; Grantham et al., 2019; Honeyman & Davison., 2016; Kleiber & Adamek., 2012; Law et al., 2017; MacCulloch et al., 2009; Naidu & Shabangu., 2015; Rullander et al., 2013; Sapountzi-Krepia et al., 2006); four were mixed methods (Du et al., 2016; Merenda et al., 2011; Rullander et al., 2017; Salisbury & LaMontagne., 2007) from which we extracted the qualitative data. Studies were heterogenous, drawing on a number of qualitative methodological traditions; five were explicit about the qualitative design, using hermeneutic/interpretive phenomenology (Carrasco & Ruiz, 2016; Honeyman & Davison., 2016), auto-ethnography (Grantham et al., 2019), grounded theory (Law et al., 2017) and focus group methodology (MacCulloch et al., 2009); the remaining studies stated the use of a descriptive qualitative design, and used appropriate qualitative methods for data collection (focus groups, semi-structured interviews) and analysis (content and thematic analyses).

As is typical in qualitative studies where the aim is to collect rich, detailed data, sample sizes were small and ranged from n=1 each for an autoethnographic study and a case study (Grantham et al., 2019; Naidu & Shabangu., 2015) to n=18 for a hermeneutic phenomenological study (Carrasco & Ruiz, 2016); the mixed methods studies carried larger sample sizes ranging from n=31 (Merenda et al., 2011) to n=975 (Du et al., 2016) and consequently produced shallower qualitative data.

The purpose of a scoping review is to describe what evidence exists, usually in order to demonstrate a gap in knowledge and hence the need for further research. Results are typically presented descriptively (to show the knowledge that already exists) rather than interpretively (to suggest what the knowledge might mean). Detailed critical appraisal of the findings is atypical (Levac et al., 2010; Pollock et al., 2020), and any critique is reserved for assessing the availability of knowledge and evidence on a given topic. Accordingly, the below results are arranged according to our research questions, namely: 1) the experiences of AIS amongst adolescents and their parents, 2) the experiences of the diagnosis and treatment of AIS and 3) best practice and coping strategies reported in the literature, enabling us to demonstrate both what is known, and is as yet unknown about AIS.

Experiences of Adolescent Idiopathic Scoliosis

Three studies (Carrasco & Ruiz., 2016; Merenda et al., 2011; Du et al., 2016) explored experiences of living with AIS. A common theme across all three studies was related to body image and self-perception. Participants reported that they had to buy and wear different clothes to their preferred style due to their altered body shape (Carrasco & Ruiz., 2016), and there was a reported dissatisfaction with physical appearance and disruption to self-image based on perceived body shape (Merenda et al., 2011), and disrupted self-perception (Du et al. 2016). These disruptions led to feelings of "embarrassment" and "inferiority" (Carrasco & Ruiz., 2016). Scoliosis was also experienced as limiting their lives, through substantial negative impact on daily activities (Du et al., 2016) and reduced physical capacity and experiences of scoliosis-related pain which incapacitated participants and prevented them from leaving the home and socialising (Carrasco & Ruiz., 2016). Du et al's mixed methods study enabled them to demonstrate that participants with a Cobb angle ≥40° generally had a more negative perception about themselves and experienced greater compromise in their daily activities. Merenda et al. (2011:389) concluded that it is important to understand how adolescents perceive their 'diagnosis, self-image, physical appearance, and how they compare themselves to peers'.

Experiences of diagnosis and treatment

Diagnosis

Diagnosis was reported as shocking and distressing in two studies (Grantham et al., 2021; Honeyman and Davison., 2016). Grantham et al's. (2021) powerful autoethnographic study recounts one author's experience of diagnosis in Poland, when she was 12 years old. The classical physical examination, the Adams Forward Bend Test – first described by William Adams in 1865 - requires the patient to remove all upper clothing so that the torso is clearly visible, and bend forward to accentuate the curve of the spine, making any imbalance in skeletal anatomy apparent. Grantham et al. (2021:3) explain the humiliation and embarrassment of this procedure:

> *Without asking whether it was fine for me or not – I was told by a doctor, again, to strip nearly naked. I was told to bend forwards. I felt extremely embarrassed. I was a girl on the edge of puberty. I did not want anybody to look at me naked. I was not a small child anymore and it did matter to me that I was not allowed to wear a top.*

In this example, the experience of being diagnosed was as distressing as the diagnosis itself and had a profound and detrimental emotional impact on the individual. Less profound, but still meaningful, is the shock of diagnosis shock that is attributed to visualisation of the spine on x-ray: *That can't be my spine, is that my spine? It must be somebody else's spine* (Honeyman and Davison., 2016: 32).

Treatment decisions

Following diagnosis, decisions have to be made about treatment. Because the adolescent with scoliosis is often not yet old enough to give legal consent, this burden falls primarily to the parents. Donnelly et al. (2004) found that the presentation of treatment options primarily dictated the parents' decision on what treatment would be best. Patients and parents felt they had little choice but to agree to a brace as this was the least invasive method with the least risk. Those who had chosen a brace, but then still underwent surgery felt a strong sense of disappointment. For those who underwent surgery, they also felt they had few other options based on the degree of their curve and potential for improvement.

The majority of the studies included in this review explored experiences of treatment, both bracing (Donnelly et al., 2004; Law et al., 2017; Sapountzi-Krepia et al., 2006) and surgery (Bilik et al. 2018; Carrasco & Ruiz, 2016; Donnelly et al., 2004; Honeyman and Davison., 2016; MacCulloch et al., 2009; Rullander et al., 2013; Rullander et al., 2017).

Treatment with bracing

Bracing is commonly used for conservative treatment of mild scoliosis. The aim is to apply pressure and stretch on the inner aspect of the curve, thus relaxing the muscle tension and correcting the spinal curvature whilst the skeleton is still immature; successful resolution of the scoliosis is not guaranteed. Braces must be worn for up to 23 hours a day, often for several months, and are notorious for being uncomfortable and unappealing. It is often difficult for parents to encourage their child's compliance with wearing the brace.

In the reviewed papers, the brace was identified as the cause of arguments between parents and participants (Donnelly et al., 2004). The impact of visual aesthetics on the acceptance of brace use was also explored (Law et al., 2017). Participants with experience of wearing a brace commented on the colour, discomfort, and negative social and emotional experiences that came when wearing a brace. While all in the study understood that in many ways the design of the brace couldn't change, many agreed that if it looked better, they would feel better about wearing it. The importance of aesthetics to the adolescent population was further addressed by Sapountzi-Krepia et al. (2006). The study revealed the range of emotions experienced by brace-wearing adolescents, including denial, fear, stress, and shame. Almost always, these feelings were related to how participants felt the brace changed their appearance, and their concerns about reactions from peers at school. Anger was attributed to changes outside the participants' control: *I was very angry, I wasn't me, I was someone else, a snail that was carrying its shell* (Sapountzi-Krepia et al. 2006:3).

Donnelly et al. (2004) reported similar findings, with participants stating that they often felt self-conscious wearing the brace and believed others treated them differently because of it. However, they continued engaging in sports activities by simply removing the brace to be able to participate.

Treatment with surgery

Across the seven studies exploring experiences of surgery (Bilik et al. 2018; Carrasco & Ruiz., 2016; Donnelly et al., 2004; Honeyman and Davison., 2016; MacCulloch et al., 2009; Rullander et al., 2013; Rullander et al., 2017) several issues stand out. All studies were conducted before or within a year of surgery except for Rullander et al. (2017) where the follow-up period was two years from the date of surgery.

The anticipation of surgery promoted feelings of anxiety and worry, with participants feeling overwhelmed by what they were facing (Honeyman and Davison, 2016; Rullander et al., 2017). In contrast, Carrasco & Ruiz (2016) reported that their participants expressed feelings of 'relief' when surgery was scheduled, with none apparently feeling afraid about the surgery as they were 'young and it's fine'. The authors concluded that the risks associated with surgery did not seem to 'affect the young women'. Post-operatively, participants experienced multiple physical impacts of surgery (Bilik et al., 2018) including fear and helplessness (Rullander et al., 2013) and difficulties with pain, mobilisation and nausea (Bilik et al., 2018; Rullander et al., 2013; Rullander et al., 2017). Withdrawing from strong painkillers caused nightmares and hallucinations for some, and feelings of anxiety continued throughout recovery, with a number of participants having second thoughts about having had the surgery (Rullander et al., 2017).

Emotions after surgery were mixed. Whilst some participants reported anxiety related to having had the surgery, others perceived their post-surgery body more positively (Bilik et al., 2018). In contrast, MacCulloch et al. (2010) identified post-surgical appearance as a source of anxiety. There were concerns about recovery and adjusting to life after surgery including worrying about their functional capacity and pain related to daily activities (Bilik et al., 2018; MacCulloch et al., 2010); participants reported that they had to change their physical activity, some permanently, not returning to sports in which they were previously involved (Rullander et al., 2013).

Social impact and support

Diagnosis and treatment of AIS often had a negative social impact on participants. At a period in their development when relationships with peers are central to adolescents' ways of being in the world, disruptions caused by the AIS could arise, and endure. There was concern about the impact of surgery on school, peer relationships and other social interactions (MacCulloch et al., 2010); these concerns were not confined to the immediate post-operative period, as some participants lost contact with friends during their recovery, and many stated that this still had an impact two years on (Rullander et al. (2013). Whilst some hid their feelings from family and friends (Honeyman and Davison, 2016) others drew on family, friends and classmates for support (Sapountzi-Krepia et al., 2006). Parental involvement throughout treatment was found to be essential. Honeyman and Davison (2006) found that parental support, particularly from the mother, was essential during hospitalisation and when recovering at home, and was highly valued by the participants who reported feeling surprised that they needed their Mum that much; mothers were needed for practical assistance, moral support, and advocacy. The authors concluded that the diagnosis and treatment of AIS impacted all family members, including parents and siblings. Bilik et al. (2018) also noted the importance of parental involvement, with several participants reporting that not having their parents present after surgery caused distress. The only study to focus solely on parents (Salisbury et al., 2007) explored parental coping before surgery and in the first few days afterwards. All participants were mothers who were anxious for several reasons with stressors related to loss of their role as a parent and how to support their children through surgery and recovery, the potential for poor surgical outcomes, uncertainties about recovery, pain post-surgery, while at the same time hiding these concerns to avoid causing further alarm to their child.

Information and clinical support

Four studies (Bilik et al., 2018; MacCulloch et al., 2010; Salisbury et al., 2007; Sapountzi-Krepia et al., 2006) reported on the need for sufficient information to assist adolescents and their families in decision-making around treatment options, preparing for and coping with bracing or surgery, and with the recovery period following surgery. Specifically, participants wanted information to address their questions about recovery (in hospital and at home), their post-surgical appearance, the emotional impact of surgery and coping strategies, the impact of surgery on daily activities and the impact of surgery on school and social relationships. Participants also raised concerns more generally about their decision to have surgery, wanting to make the decision independently, but with the support of others. They wanted to know how others were diagnosed and came to their decisions to have surgery (MacCulloch et al. 2010). Parents reported anxiety towards the pain their child was likely to experience and received little information to know what to expect or how to support their child through post-operative pain episodes (Salisbury et al., 2007). Adolescents themselves documented surprise at the level of pain, and the changing severity, nature, and duration of that pain, following surgery and through recovery (Bilik et al., 2018; Rullander et al., 2017) suggesting that they had not been sufficiently informed of the likely severity of the pain following this major surgical intervention. Parents also needed information on what to expect on discharge: *We know nothing about what she will or won't do at home. What kind of exercise will she do?* (Bilik et al., 2018:347).

In contrast, participants who had received bracing treatment for their scoliosis reported that they were satisfied with the information they received regarding their condition and therapy (Sapountzi-Krepia et al., 2006). However, the information was not accompanied by support from the health care professionals, and this perception of poor clinical support is also reported across other studies. Some adolescents and parents felt that medical staff did not care or listen to their concerns, particularly around care practices and post-operative complications (Salisbury et al., 2007). Participants reported feeling that they were not being listened to by nursing staff, who they also perceived lacked the specialist skills to operate the technical medical equipment (Rullander et al., 2017). In the only auto-ethnographic study (Grantham et al., 2021) multiple indignities at the hand of clinicians during diagnosis and treatment had an enduring negative impact on the lead author; numerous negative interactions with clinicians changed her perceptions, trust and ultimately adherence with treatment. Small gestures like being treated as a person and not a “scoliotic back” and empathy from professionals were seen as important. Sapountzi-Krepia et al. (2006) further reported participants’ perceptions that support from a physiotherapist was not enough, with only one of their 12 participants seeing a psychologist.

Coping strategies and best practice

Coping strategies

Adolescents with AIS and their parents need support to cope with the diagnosis, and with the different forms of intervention. Bracing, whilst avoiding surgery, brings its own difficulties and challenges, as described above, whilst surgery has the added concerns relating to perceived risk, loss of control, and a potentially painful recovery. Both have psychosocial and emotional impacts, and families and adolescents cope with all these factors in different ways.

A range of coping strategies employed by adolescents with AIS were noted. To address their concerns about appearance and body image, adolescents often attempted to cover or hide their bodies, changing their dress or posture, wearing loose clothing, and avoiding certain situations, such as going to the beach (Carrasco & Ruiz., 2016). To cope during and after surgery, including on discharge home and when returning to school, adolescents engaged in active coping, social support via discussion with peers, distraction and avoidance techniques; there was a sense of being mentally/emotionally stronger than they or their parents realised (Honeyman & Davison, 2016; MacCulloch et al., 2010). Adolescents and their parents used achievable targets to motivate themselves and their child through pre-operative preparation and post-operative recovery – such as being able to get up out of bed after surgery, and seeing how much height they had gained: *I tried to prepare him by having him think positive thoughts like he will be one inch taller and the hump on his back will be gone* (Salisbury et al., 2007:220).

Two studies (Klieber & Adamek, 2013; Naidu & Shabangu, 2015) reported on interventions specifically to help alleviate adolescents’ anxiety in relation to treatment. Notably, both employed artistic distraction methods to divert participants’ focus from their situation. Drawing on the evidence that music therapy can decrease postoperative pain in children having minor surgery, Klieber and Adamek (2013) applied the approach to adolescents following surgery for AIS. They found that music therapy also helped to alleviate anxiety, promoted mental relaxation, and provided a welcome distraction from pain, although it was important for adolescents to self-select the type of music. Naidu and Shabangu's (2015) case study described the use of poetry during psychotherapy to address severe anxiety that was preventing the subject from undergoing necessary surgery to address her AIS. Drawing on centuries of evidence that poetry can ease physical and emotional ailments, the study details this use of poetry as a narrative therapy which gently enabled the participant to accept the need for surgery and agree to the procedure.

Best practice: information and preparation

Most of the studies above touched upon aspects which may need addressing to enhance best practice; for example, a recurring theme related to information provided before and after surgery (or the lack thereof), indicating that improving information provision during these time periods would benefit patients and improve clinical practice.

Adolescents rarely present with scoliosis without a family member; parents need information to enable them to provide appropriate support to their son or daughter. Salisbury et al., (2007) comment that parents fully understood that their involvement could affect their adolescent's attitudes about recovery, and they need information from the outset about all stages of treatment, to know what to expect and to be able to control their own anticipatory fears and better support their child. Rullander et al. (2017) suggested that information about coping strategies could be included in patient pre-operative education to improve patient experience.

Best practice: clinical care

The evidence reported above demonstrates that pain and anxiety are common experiences relating to AIS surgery. Rullander et al. (2013) suggested that during the peri-operative period, pain management and treatment of opiate-induced nausea should be optimised. They also suggest that there is a need for greater psychological preparation for AIS patients, including more time spent on making care plans. The need for better attentiveness from clinical staff is also identified (Salisbury et al., 2007); the experience may be unremarkable to clinical staff who encounter it repeatedly, but it is unique and immensely challenging to parents and their adolescents with AIS. Without being included and informed about progress, complications, or decisions about treatment during recovery, parents can feel ‘left out’ (Salisbury et al., 2007:227).

Better post-operative support following discharge was also recommended; there was considerable uncertainty around what to expect during the convalescence period at home (Rullander et al., 2013; Rullander et al., 2017; Salisbury et al., 2007). Rulklander et al., (2017:421) comment that ‘*It is probably during this transition from hospital to home that patients and families need the most patient education and support from staff for coping and self-care*’ and, building on their earlier (2013) work, the authors recommend that this could be achieved by follow-up calls to families after discharge.

Finally, a consideration of how brace treatment may be improved by enhanced practices. Opting for bracing may seem the less complicated choice, since it carries little physical risk to the child unlike the risks (including that of paralysis) attached to spinal fusion surgery. Yet as discussed above, adherence to and compliance with treatment is difficult and often accompanied by disagreements between parents and their adolescent with AIS. Of the three papers that addressed bracing treatment (Donnelly et al., 2004; Law et al., 2017; Sapountzi-Krepia et al., 2006) the first identified the need to address compliance with brace-wearing alongside the need for a normal adolescent lifestyle. Since adherence to brace-wearing is demonstrated to be poor, they recommend ‘prescribing’ fulltime wearing (24 hours a day, 7 days a week) so that even with inevitable periods of non-compliance, there is an effective level of adherence to treatment. Adherence is also about comfort and acceptability; Law et al. (2017) emphasised the importance of co-designing patient braces, noting that involving adolescents and parents in this process could improve adherence with this treatment and allow patients to feel more like the brace was ‘theirs’. Finally, the enduring need for ongoing support from a range of healthcare professionals including doctors, nurses, physiotherapists, and psychologist, is identified by Sapountzi-Krepia et al. (2006). Brace wearing is an endurance event lasting several months and sometimes years, and clinical support needs to go beyond prescribing the brace and monitoring adherence, to also providing emotional and psychological support.

Discussion

This systematic scoping review sought to better understand 1) the experiences of AIS amongst adolescents and their parents, 2) the experiences of the diagnosis and treatment of AIS and 3) best practice and coping strategies reported in the literature.

The fifteen studies included in this review speak loudly to issues that challenge the normal tasks of adolescent development. One well-cited psychological theory (Erikson 1968) proposes that the key task of the adolescent (aged 12-18 years) is to resolve the crisis between identity and confusion and to emerge into young adulthood with a clear sense of who and what they are. At the same time as grappling with all this psychosocial turmoil, the adolescent's body is undergoing significant physical changes. The onset of puberty and the changes in hormone levels marks the development of secondary sex characteristics and accelerated periods of skeletal growth (Hinchcliff & Montague, 1998; Smith et al., 1998). The body they were familiar with in middle to late childhood changes, as limbs lengthen and the proportions of limbs to trunk and head, alter. The harrowing account of feeling embarrassed and exposed during diagnosis (Grantham et al. (2021) shines a light on the heightened sensitivity of the adolescent to their changing pubertal body, and the need for privacy and dignity to be respected.

During this developmental period, the most important social relationships are with peers, outside of the home (Smith et al., 1998). Armed, hopefully, with a strong and secure sense of trust developed during early years, and the need to develop a growing sense of independence from their parents, adolescents turn their focus away from the family home towards their peers and the outside world and spend this time period working out their place in that world. In early adolescence, the need to be like their peers is crucial and homogenous friendships prevail; during later adolescence, as the sense of individuality and identity strengthens, heterogenous relationships become the norm (although the additional influences of gender and sexual identity are also at play here) (Herbert, 2003; Smith et al., 1998).

The diagnosis of AIS, typically at the start of puberty, throws these normal processes into disarray, and it is thus unsurprising that adolescents' concerns regarding body image, self-perception and comparisons with their peers feature so dominantly in this reviewed literature. Across all studies (except Salisbury et al., 2007 which recruited parents of adolescents with AIS) there is mention, in some way, of the need to 'fit in', and of measures taken (such as wearing different clothes to hide physical deformity, avoiding social activities and interactions) to avoid appearing as different to their peers. Whilst none of these young participants used the word ‘stigma’ to describe their feelings of being or feeling that they were or would be treated differently because of their AIS, their reported thoughts and actions speak to the seminal work of Erving Goffman (1963) and the efforts that individuals make to conceal a feature of difference which they perceive will discredit them.

The parental role also changes during adolescence. As the young person seeks to move away from dependence on their parents to independence, their parents need to relinquish the control they have necessarily had until this stage, and hand this control over to their son or daughter. A diagnosis of AIS disrupts this developmental process, throwing the adolescent back into a state of dependence and loss of control. The work of Salisbury and colleagues (2007) reveals just how acutely aware parents are of this disruption, and the conflicts they feel between needing to maintain their adolescent’s independence, yet also be involved and supportive in their diagnosis, treatment decision-making, and care. For the adolescent, their conflict arises between not wanting to need their parent - since this challenges their perception of self as independent - but at the same time, really needing and wanting their support.

The review also highlights the need for better clinical support. Whilst diagnosis and treatment decisions (bracing and/or surgery) appear straightforward, the details of clinical care (particularly post-operative pain management) and support are left wanting. Spinal fusion surgery causes severe postoperative pain (Borgeat & Blumenthal, 2008) and it is surprising that in the era of modern medicine when excellent pain control can be achieved, adolescents continue to endure significant and intolerable levels of post-operative pain.

The identification of parents’ and adolescents’ concerns about poor clinical support may suggest a need for all clinicians involved in the care and treatment of those with AIS to have a greater awareness of the impact that diagnosis and treatment has on families. On the evidence reviewed here, clinicians appear to pay little attention to the psychological needs of adolescents with AIS despite the intense impact on body image, self-perception and comparison with peers that are typical during adolescence.

An overarching aim of this literature review was to identify shortcomings and directions for future research in this area, so some comment is warranted here, particularly in relation to the broader literature that deals with AIS. The reviewed studies were predominantly focussed on the pre-operative and post-operative (hospital and home) recovery periods. Only one study (Rullander et al., 2017) followed up with adolescents whose AIS surgery had occurred ~2 years previously. Consequently, there is no insight into whether concerns raised across these studies about longer term body image, social impact, sports and activities, resolve or endure.

All studies recruited either adolescents or their parents, and none addressed the longer-term quality of life outcomes of AIS treatment, coping or best practice, from at least 10 years following conclusion of treatment. There is thus a dearth of evidence on the enduring consequences of AIS and any treatment related to it, when those affected reach school-leaving age, seek employment, and - for women - pursue motherhood. Investigating these longer-term outcomes is warranted, to enable adolescents and their parents to make a fully informed decision when treatment options are presented to them after diagnosis, and to enable clinicians to understand the long-term implications of the interventions they recommend. Second, there was also little research into the impact on AIS on parents and other immediate relatives. Parents are often central to decision-making about treatment, and to giving support throughout the process of diagnosis and treatment -if genuinely robust family-centred care is to be provided, the needs of other family members need to be understood. Third, no studies reported patient involvement in the development, design, delivery or reporting of the findings – so we do not know how well the issues presented here reflect the primary concerns and experiences of those living with AIS, either before, during or following treatment. Finally, these results stand in contrast to the quantitative literature on AIS and suggest that future studies need to give greater consideration to more fundamental considerations, related to quality of life and patient experiences of living with AIS (Essex et al., 2021). On this point, it appears that qualitative research could fill a number of gaps in this area and complement existing quantitative studies to provide a more rounded understanding of the impact of AIS and treatment on adolescents and their families, across the life course.

Conclusions

This review demonstrates that AIS has a substantial impact on adolescents and their families, but that the minimal available evidence focusses on the pre, peri and short-term post-operative periods. A recent research priority setting exercise by the British Scoliosis Research Foundation has identified (amongst other issues) the need to understand how quality of life is affected by scoliosis and its treatment, what the long-term outcomes and side-effects of surgery are, and whether scoliosis gets worse over time, with or without treatment. There is therefore a pressing need for more qualitative research to understand needs, perspectives and experiences of adolescents with AIS and their families from the point of diagnosis, throughout treatment, and in the longer-term following treatment and recovery, to augment any quantitative investigations in these areas of concern.

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