

REVIEW

The influence of pregnancy on women with adolescent idiopathic scoliosis

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Abstract

Purpose The study's aim was to address three fundamental questions related to pregnancy and adolescent idiopathic scoliosis (AIS), and provide clinically applicable answers to spine specialists and general practitioners alike.

Methods The authors performed a systematic literature review using MEDLINE, EMBASE, Google Scholar, and Cochrane Database of Systematic Reviews to identify articles published between 1980 and 2015 that described pregnancy-related characteristics and outcomes in AIS patients. The search was conducted using PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guidelines and evidence was classified according to the Oxford CEBM (Centre for Evidence-Based Medicine) appraisal tool.

Results Twenty-two articles incorporating more than 3125 AIS patients were included. All studies concluded level 2b evidence or lower. Nulliparity rates were slightly higher among AIS patients, and more frequent infertility treatment was required. Pregnancy-related back pain was common, and while non-disabling, may have been more severe than in healthy women. Minor curve progression often occurred during pregnancy, though its permanence was questioned and significance unknown. Back pain and curve progression occurred independent of AIS treatment modality. With modern technology, anesthetic and obstetric complications in the perinatal period were not elevated in AIS mothers.

Conclusions Women with AIS experience slightly elevated rates of nulliparity, infertility treatment, prepartum back pain, and peripartum curve progression. However, most women are able to have children and are not at increased risk of pregnancy-related complications. Higher quality evidence is needed to better define these relationships and allow more guided counseling and treatment.

Keywords Adolescent idiopathic scoliosis · Anesthesia · Back pain · Curve progression · Pregnancy

Introduction

Idiopathic scoliosis is the most common type of scoliosis with adolescent idiopathic scoliosis (AIS) representing the vast majority of cases [1–3]. AIS affects 2–4% of patients between 10 and 18 years of age [4–6]. Because the vast majority (>85%) of AIS patients are female, issues related to women's health are of specific concern in this condition [7–11]. Naturally, as the age presentation of young females with AIS immediately precedes the childbearing age, the impact of scoliosis on pregnancy is particularly relevant.

In this study, we sought to outline a practical guide for pediatric spine specialists counseling young patients with AIS. Motivation for this endeavor arose from the authors' own experience in clinic and from the absence of a singular resource addressing this important problem. Commiserate with the frequency of patient inquiries, a focus was placed on studies describing the spinal changes surrounding pregnancy and delivery-related challenges in these patients. By means of a systematic review, the best available evidence was identified and related to each of the questions below:

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1. How does scoliosis affect the timing and success of pregnancy?
2. What spine-related changes occur during and after pregnancy?
3. What anesthetic and obstetric considerations are relevant to AIS mothers delivering a child?

Methods

A comprehensive literature search was performed using MEDLINE, EMBASE, Google Scholar, and the Cochrane Database of Systematic Reviews. Relevant combinations of the following MeSH terms were searched in articles published between the years 1980–2015: “adolescent idiopathic scoliosis”, “idiopathic scoliosis”, “adolescent scoliosis”, “pregnancy”, “pregnant”, “childbearing”, “children”, “mother”, “parent”, “marriage”, “married”, “marital”, “delivery”, “labor”, “cesarean”, “parity”, “anesthesia”, “anesthetic”, and “analgesia”. Inclusion criteria were original science manuscripts published in English between 1980 and 2015, which related AIS with one or more pregnancy-related conditions or outcomes measures, including parity, pain, spine curvature, and delivery characteristics. Review articles, case reports, and case series with less than five subjects were excluded. Records were then screened for topic relevance, specifically whether subjects with AIS were assessed in relation to the pregnancy-related conditions, attributes, or outcome measures. Full-text articles are assessed for eligibility and excluded for one or more reasons outlined in the lower right box within Fig. 1. The remaining articles were included in the qualitative analysis. A recursive search of the bibliography of target manuscripts was also performed to identify potential articles of interest.

The literature search and screening were conducted in accordance with the guidelines outlined by the Preferred Reporting Items for Systematic Reviews and Meta-Analyses statement [12]. Two reviewers independently extracted data and discussed disagreements before including/excluding manuscripts. An arbiter resolved any relevant discrepancies before final tables were constructed and data reported. The study methodology within each article was examined and assigned a level of evidence based upon the Oxford Centre for Evidence-Based Medicine 2011 guidelines [13]. The sum of the quality of evidence addressing each of the three posed questions was determined using the GRADE system of evidence classification [14, 15]. Records across study populations were not pooled, as the heterogeneous and often insubstantial nature of the available data would have rendered such an endeavor misguided, if not misleading. Our aim was to objectively

synthesize available data and present principal findings in a clinically useful manner. Key study findings of each manuscript are summarized within Tables 1, 2 and 3. Descriptive statistics are reported as proportions of a population. Where appropriate (and when made available by the article of origin) comparisons between populations are described with a significance statistic as explicitly reported within the manuscript.

Results

Study selection

A total of 22 studies were identified that met inclusion and exclusion criteria, representing more than 3125 AIS patients. Eighty-two studies were identified via database query after filtering out reviews, case reports, and duplicates. After screening for topical relevance, 41 full-text articles were reviewed for direct applicability to the 3 clinical questions posed. Finally, 19 studies are excluded for the specific reasons outlined within the lower right box of Fig. 1.

Retrospective and observational studies predominated, with only three studies utilizing a prospective design. Many authors compared AIS patients (pregnant and non-pregnant) against healthy controls, utilizing either patient-matched subjects or population-based controls for comparisons. Reported treatment approaches were widely heterogeneous, from simple observation to bracing to surgical correction. Limited numbers and vague operative descriptions precluded any reasonable comparisons between different surgery types (Harrington rods vs. segmental fusion). Rather, when appropriate, all surgical treatment was combined and qualitatively compared against more conservative treatment approaches (bracing, observation).

Clinical questions

1. How does scoliosis affect the success and timing of pregnancy?

One or more aspect of this question was addressed by 16 studies (Table 1). Three studies incorporating controls identified a higher rate of nulliparity in AIS (41–68%) relative to healthy women (20–44%) [16–18]. Combining these three populations, AIS patients were 10–24% more likely than controls to be nulliparous. Falick-Michaeli et al. on the other hand, found a 21% higher rate of nulliparity among controls [19]. In the only study specifically comparing controls and AIS patients treated conservatively and surgically, the rate of nulliparity was similar across all

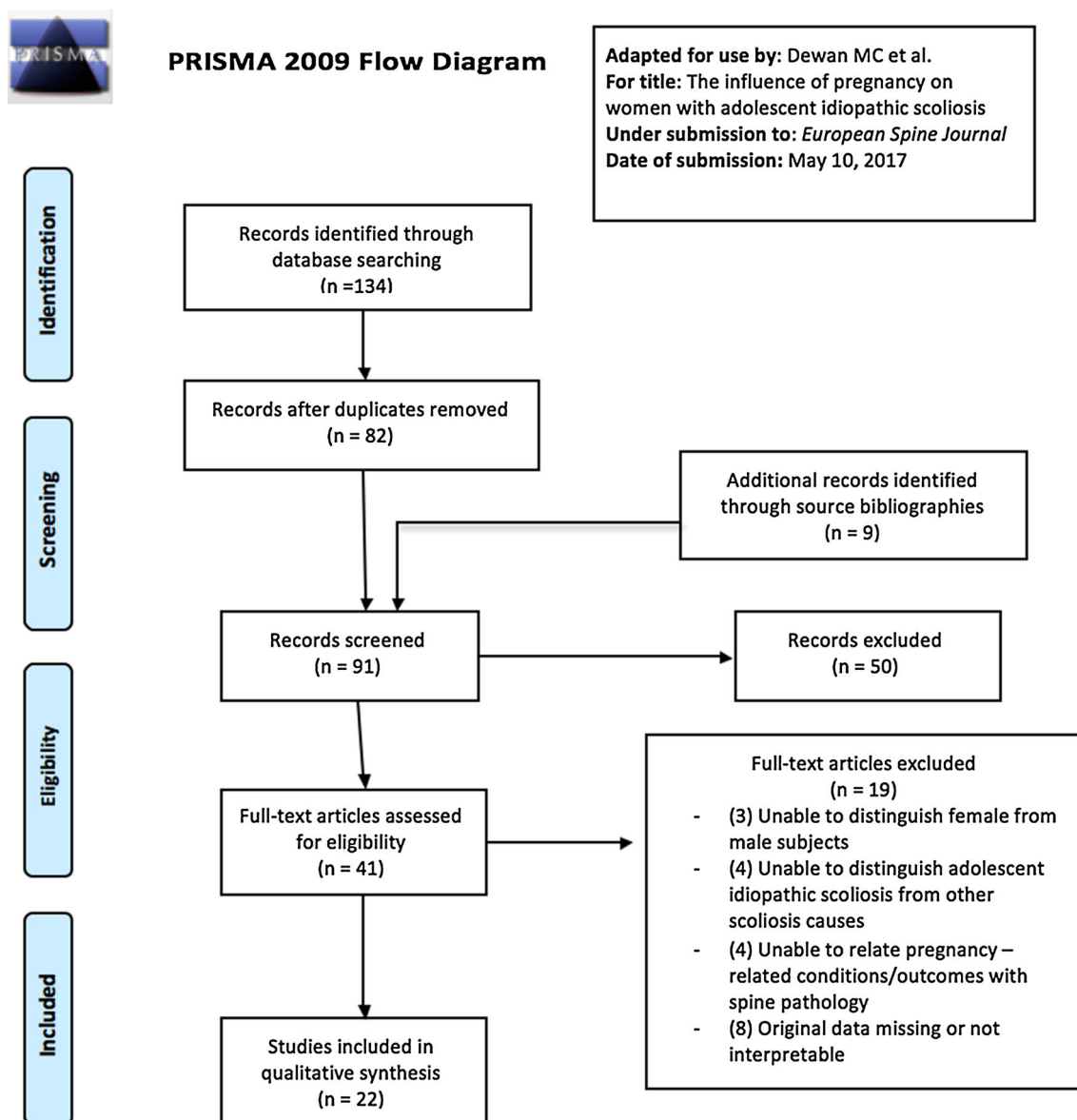


Fig. 1 PRISMA flow diagram delineating the literature search process

groups [15.6% (controls) vs. 18.4% (AIS-surgery) vs. 14.4% (AIS-bracing)]. Cochran and authors found that 56% of scoliotic women in Sweden were nulliparous—a rate that did not differ significantly from age-matched population controls at that time [20]. Among studies that lacked a non-AIS control comparator arm, the rates of nulliparity in AIS women ranged widely from 13 to 70% [20–25]. The largest study was a retrospective cohort analysis of 1292 AIS and 1178 control subjects by Goldberg et al. which demonstrated a 10% higher rate of nulliparity within the AIS cohort (49 vs. 39%, $p = \text{NR}$) [18]. This figure roughly matches the joint results of the smaller studies: a modestly higher rate of nulliparity among patients with AIS (14–68%) compared to healthy controls

(16–44%) [16, 18, 19, 24, 26]. Still, given the overlapping rates and non-significance between comparison groups in the majority of studies, a higher nulliparity rate among AIS patients—while suggested—is not concluded with certainty. Interestingly, Lebel and coauthors described a higher rate of fertility treatment among women with scoliosis (7.1 vs. 1.6%, $p < 0.001$) [17].

Four studies described the number of offspring among AIS mothers. Danielsson et al. published a series of 136 surgically treated women and 111 brace-treated women and found no difference in the number of children born compared to non-AIS controls (1.8 vs. 2.0, $p = 0.25$) [26]. Akazawa and coauthors, on the other hand, did show a reduced number of offspring in 80 patients with idiopathic

Table 1 AIS effects on success and timing of pregnancy

| Author | Year | Study design | Patient number | Treatment | Findings | Level of evidence ^a |
|------------------------|------|-----------------------------------|--------------------------------|----------------------------------|---|--------------------------------|
| Falick-Michaeli et al. | 2015 | Cohort, retrospective | 40 AIS 40 CTR | Surgery | First pregnancy an average of 7.3 years after AIS-surgery. Average maternal age was 31. 64% of AIS group nulliparous vs. 85% in controls (SNR) | 4 |
| Bauchat et al. | 2015 | Case-control, prospective | 41 AIS 41 CTR | Surgery | Average maternal age was 33 years, similar to controls. Average gestation was 39 weeks, similar to controls. 68% of AIS group nulliparous vs. 44% of controls (NS) | 3b |
| Akazawa et al. | 2012 | Case-control, retrospective | 56 non-AIS 80 AIS 80 CTR | Surgery | AIS mothers had significantly lower average number of children (1.7) compared to control mothers (2.2) | 3b |
| Lebel et al. | 2012 | Population-based, retrospective | 98 AIS | | Average maternal age (29 years), and average gestation age (39 weeks) similar to population controls. Higher nulliparity in AIS group vs. population (41 vs. 20%, $p < 0.001$). 7.1% of the AIS group received fertility treatment | 4 |
| Lange et al. | 2011 | Case series, retrospective | | Boston brace | 13% of the patients nulliparous | 4 |
| Lange et al. | 2009 | Case series, retrospective | 102 AIS | Boston brace | 13% of the patients nulliparous | 4 |
| Takayama et al. | 2009 | Case series, retrospective | 15 AIS | Surgery | 40% of the patients nulliparous | 4 |
| Smith et al. | 2003 | Cohort, prospective | 40 AIS | 24 (uncorrected) 16 (surgery) | Gestation ranged from 35 to 42 weeks. 70% of the patients were nulliparous | 2b |
| Danielsson et al. | 2001 | Case-control, retrospective | 247 AIS 90 CTR | 136 (surgery) 111 (brace) | Relative to CTR, age at first delivery higher for BT group (28 vs. 26 years, $p = 0.01$) but not ST group (27 years). Nulliparity similar across groups (ST: 18%, BT: 14%, CTR: 16%). The mothers of all three groups (ST, BT, CTR) had similar average number of children (1.8, 1.9, 2.0) | 3b |
| Goldberg et al. | 1998 | Case-control, retrospective | 1292 AIS 1134 CTR | | Higher rate of spontaneous abortion in AIS vs. controls (12.8 vs. 9.7%, $p < 0.05$). Lower rate of stillbirths in AIS vs. controls (0.5 vs. 1.5%, $p < 0.05$) | 4 |
| Orvomaa et al. | 1997 | Case series, retrospective | 146 AIS | Surgery | Gestation ≥ 38 weeks in 90% of subjects. 9% suffered miscarriages | 4 |
| To et al. | 1996 | Cohort, retrospective | 8 AIS | 7 (untreated) 1 (fusion) | Average gestation 37 weeks | 4 |
| Goldberg et al. | 1994 | Comparative cohort, retrospective | 1292 AIS 1178 CTR | Mixed | 49% of AIS cohort nulliparous vs. 39% of control (unknown significance) | 4 |
| Betz et al. | 1987 | Cohort, retrospective | 355 AIS | Mixed | 51% nulliparity among AIS cohort. 69 pregnancies ended in spontaneous or elective abortion. Average of 1.4 children per mother | 4 |
| Ascani et al. | 1986 | Case series, retrospective | 151 AIS | Observation | Nulliparity ranged from 24 to 41%; dependent upon curve location | 4 |
| Cochran et al. | 1985 | Case series, retrospective | 81 AIS | Milwaukee brace | 56.3% were nulliparous. Average of 1.4 children per mother | 4 |

AIS adolescent idiopathic scoliosis, BT brace treatment, CTR control, NS not significant ($p \geq 0.05$); SNR statistical significance not reported, ST surgical treatment

^a Levels of evidence follow the system of grading outline by the Oxford CEBM levels of evidence

Table 2 Spine-related changes during and after pregnancy

| Author | Year | Study design | Patient Number | Treatment | Findings | Level of evidence ^a |
|------------------------|------|-----------------------------|----------------------|--------------------------------------|--|--------------------------------|
| Falick-Michaeli et al. | 2015 | Cohort, retrospective | 40 AIS 40 Control | Surgery | 35% of AIS patients had severe back pain during pregnancy, and 76% had sustained back pain even after child delivery. Minimal back pain in controls | 4 |
| Lange et al. | 2011 | Case series, retrospective | | Boston brace | Back pain in 50%, similar to age-matched controls. Of these, 60–90% reported moderate to severe—but non-debilitating—back pain | 4 |
| Lange et al. | 2009 | Case series, retrospective | 102 AIS | Boston brace | 55% of the patients had back pain | 4 |
| Bjerkreim et al. | 2007 | Cohort, prospective | 76 AIS | Surgery | 62% of patients had increased back pain during pregnancy | 2b |
| Danielsson et al. | 2001 | Case–control, retrospective | 247 AIS 90 CTR | 136 (surgery) 111 (brace) | Lower back pain similar across ST, BT, and CTR groups (35, 43, and 28%). No correlation between number of pregnancies and curve progression | 3b |
| Orvomaa et al. | 1997 | Case series, retrospective | 146 AIS | Surgery | 14% of patients took ~2 weeks sick leave during first pregnancy due to back pain. 12% took leave during second pregnancy due to back pain | 4 |
| Daley et al. | 1990 | Cohort, retrospective | 18 AIS | Surgery | 17% of people had intermittent back pain since surgery. 17% had back pain only during pregnancy | 4 |
| Betz et al. | 1987 | Cohort, retrospective | 355 AIS | Mixed | 77% reported back pain during pregnancy; severe in 12%. Curve severity predicted curve progression | 4 |
| Ascani et al. | 1986 | Case series, retrospective | 151 AIS | Observation | Pregnancy-related back pain varied from 16 to 37% depending upon curve location. Curve progression increased with increased number of pregnancies, independent of curve location | 4 |
| Cochran et al. | 1985 | Case series, retrospective | 81 AIS | Milwaukee brace | All patients who became pregnant at age younger than 23 had progression of curve | 4 |
| Berman et al. | 1982 | Case series, retrospective | 8 AIS | 4 (Milwaukee brace) 4 (untreated) | Improvement in curve after pregnancy in 2 patients, no change in 3 patients, and progression (>5°) in 3 patients | 4 |
| Blount et al. | 1980 | Case series, retrospective | 10 AIS | Milwaukee brace | Mild curve progression in 30% after initial pregnancy (but no progression when stable curve antepartum). Unchanged curve in 70%. Stability of curve unrelated to age. Curve progression not correlated with curve severity | 4 |

AIS adolescent idiopathic scoliosis, BT brace treatment, CTR control, ST surgical treatment

^a Levels of evidence follow the system of grading outline by the Oxford CEBM levels of evidence

scoliosis relative to healthy controls (1.7 vs. 2.2 children, $p = 0.006$) [27]. Two other groups both reported child delivery rates of 1.4 children per mother, though a comparison against controls was not conducted [20, 24].

In the question stem, ‘timing’ refers both to maternal age and gestational age. The mean maternal age at time of first delivery ranged from 26 to 33 years, and did not differ between AIS mothers and healthy mothers [16, 17, 19, 26]. In one study, women treated with a brace were on average 1.4 years older at the time of first pregnancy than surgically treated patients [26].

Gestational age at the time of birth was similar in patients with and without AIS. Both Lebel and Bauchat reported a mean gestation age at birth of 39 weeks, suggesting that differences do not exist between patients

treated conservatively and those treated with surgery [16, 17]. In the Orvomaa population of surgically corrected patients, 90% of mothers delivered at 38 weeks and beyond, matching the population average [28, 29]. Others also reported gestation ages that did not differ from the general population [23, 30].

Finally, in a single questionnaire-based study including more than 1200 patients with AIS, spontaneous abortions were more common among women with scoliosis (12.8 vs. 9.7%), while stillbirths rates were slightly higher in non-AIS controls (1.5 vs. 0.5%) [31].

The quality of available evidence for this clinical question was low (Table 1).

1. Summary of findings:

Table 3 AIS effects on anesthesia and obstetrics

| Author | Year | Study design | Patient number | Treatment | Findings | Level of evidence ^a |
|------------------------|------|---------------------------------|-----------------------|------------------------------|---|--------------------------------|
| Falick-Michaeli et al. | 2015 | Cohort, retrospective | 40 AIS 40 Control | Surgery | Epidural anesthesia given in 70% of AIS vs. 100% of CTR. C-section rate: 24% in AIS | 4 |
| Bauchat et al. | 2015 | Case-control, prospective | 41 AIS 41 Control | Surgery | Epidural anesthesia given in 39% of AIS with spinal instrumentation vs. 5% of CTR. Longer epidural procedure duration in AIS (6.5 min) vs. CTR (4.6 min). Neuraxial failure in 12% of AIS vs. 0% of CTR. Similar anesthesia consumption, labor duration, and C-section rate (12%) | 3b |
| Lebel et al. | 2012 | Population-based, retrospective | 98 AIS | | Use of epidural anesthesia was similar to the control population (17.3%). C-section rate greater AIS vs. CTR (21 vs. 13%). AIS patients had similar rates of preeclampsia, polyhydramnion, and premature rupture of membranes as compared to the population | 4 |
| Takayama et al. | 2009 | Case series, retrospective | 15 AIS | Surgery | 19% of AIS patients had C-sections | 4 |
| Smith et al. | 2003 | Cohort, prospective | 40 AIS | 24 (obs.) 16 (surgery) | Vaginal delivery: 59%. No anesthetic requirement in 9 uncorrected and 2 corrected AIS patients. Epidural catheters were placed in 7 uncorrected AIS patients successfully. Unsuccessful continuous spinal infusion in 1/6 patient C-section delivery: 41%. Successful combined spinal and epidural anesthesia in 2/2 uncorrected AIS patients. Single shot spinal anesthesia in 2. Continuous spinal infusion in 13 (8 successful, 3 moderate, 2 failed) | 2b |
| Danielsson et al. | 2001 | Case-control, retrospective | 247 AIS 90 Control | 136 (surgery) 111 (brace) | C-section rate by treatment: Harrington rod—15.2%, brace—12.1% and control—15.2% | 3b |
| Orvomaa et al. | 1997 | Case series, retrospective | 146 AIS | Surgery | C-section rate was 23% | 4 |
| To et al. | 1996 | Chart review, retrospective | 8 AIS | 7 (obs.) 1 (surgery) | C-section rate was 13% | 4 |
| Daley et al. | 1990 | Chart review, retrospective | 18 AIS | 18 (surgery) | Successful epidural anesthesia in 18/19 pregnancies. 20/21 epidural attempts successful (single attempt in 10, patchy/excessive sedation in 11). Low back pain after multiple attempts in 2 | 4 |
| Crosby et al. | 1989 | Chart review retrospective | 14 AIS | Surgery | 8 (57%) of patients received epidural anesthesia: adequate sedation in 7, single attempt in 5. General anesthesia for C-section in 3, while remaining 3 declined regional anesthesia | 4 |
| Betz et al. | 1987 | Chart review, retrospective | 355 AIS | Mixed | 1.2% of deliveries had failed spinal anesthesia. C-section delivery rates were 7.4% (all AIS) and 2.5% (spinal fusion patients). No scoliosis-related problems identified during delivery | 4 |
| Cochran et al. | 1985 | Case series, retrospective | 81 AIS | Milwaukee brace | C-section rates were similar to population | 4 |
| Hubbert et al. | 1985 | Cohort, retrospective | 17 AIS | Surgery | 12% of AIS patients received spinal anesthesia. 53% of patients received successful epidurals. Higher epidural success rate when lower vertebrae unfused | 4 |
| Weinstein et al. | 1981 | Case series, retrospective | 194 AIS | Obs. | 1.4% of patients had C-sections, which were associated with major complications | 4 |

AIS adolescent idiopathic scoliosis, BT brace treatment, CTR control, Obs observation, ST surgical treatment

^a Levels of evidence follow the system of grading outline by the Oxford CEBM levels of evidence

- Women with AIS may be slightly less likely to become pregnant than their age-matched controls, and may be more likely to require fertility treatment.
- Independent of surgery or bracing, mothers affected by AIS can expect to have a similar number of children at the same maternal and gestational age as the general population.

2. What spine-related changes occur during and after pregnancy?

Spine-related changes are divided herein into those relating to back pain and those relating to curve progression. Twelve studies specifically examined the relationship between pregnancy and one or both of these entities (Table 2).

A significant proportion of scoliosis patients reported new back pain related to pregnancy; however, whether the incidence and severity differed from healthy women varied from study to study. In two reports of patients with AIS treated with bracing, Lange and coauthors found that 50–55% experienced new back pain during pregnancy [21, 32]. While the vast majority described moderate to severe pain, the pain was not debilitating and did not otherwise influence the performance of daily functions [21]. Similarly, in a study of 76 women treated with surgical instrumentation, 62% of those who became pregnant reported increased back pain, though the severity and duration was not described [33]. Betz et al. found that more than three-quarters of pregnant women reported pain during pregnancy relative to half of non-pregnant women who reported back pain during this age period [24]. Severe pain was reported in 12% of the former group and in 3% of the latter. In this study, AIS patients treated surgically were not more likely to experience back pain during or after pregnancy than those treated conservatively. Comparably, Danielsson et al. found no difference in pregnancy-related back pain between patients treated surgically and those treated with a brace [26]. Moreover, when both groups were compared to healthy controls, a significant difference in incidence of back pain was not detected. Likewise, in the Betz cohort at the time of last follow-up, the prevalence and severity of back pain was not different between AIS mothers and AIS women who had never been pregnant [24]. Others reported a pregnancy-related increase in back pain in 16–37% of patients [25, 34]. Ascani et al. correlated curve pattern with pain and found that women with thoracic curves were least likely to experience back pain, while those harboring a thoracolumbar curve reported the highest rates of pregnancy-related pain [25].

While most studies describe non-debilitating back pain that resolves following delivery, two exceptions are noted. In a report of 146 patients treated with Harrington instrumentation, more than 10% mandated sick leave during their first and/or second pregnancy specifically due to back pain [28]. Falick-Michaeli et al. describe a small cohort ($n = 17$) of pregnant AIS patients who required hospitalization for severe back pain and back pain that persisted beyond pregnancy in 76% of women [19].

The occurrence and degree of curve progression during pregnancy varied across studies. Betz et al. found that roughly

25% of patients experience curve progression greater than 5° and about 10% greater than 10° [24]. Pre-pregnancy curve severity also predicted curve progression: 6% of patients with a curve $<30^\circ$ experience 10° or more of curve progression, relative to 29% of patients with a pre-pregnancy curve $>50^\circ$. The use of orthosis did not mitigate these observed changes. In a small series ($n = 7$), Berman et al. found that 3 (43%) patients with a primary curve $>25^\circ$ experienced a small, but detectable progression of at least 5° [35]. Betz and colleagues also noted that the age of the patient at the time of first pregnancy had no influence on the risk of curve progression [24]. This finding is distinct from the report by Cochran et al. of 81 brace-treated women, which found that all patients who became pregnant before the age of 23 experienced late curve progression [20]. Sparse and heterogeneous reporting within source manuscripts precluded correlating curve classification (e.g., Lenke, King) with pregnancy-related curve progression or back pain.

Ascani and authors have also correlated the number of pregnancies with degree of curve progression. In this large observational study, patients were subdivided into curve type and a stepwise progression in deformity was observed. Upon reaching adulthood, the curve had progressed 10° – 13° before pregnancy, 13° – 16° (cumulative) after the first pregnancy, and 16° – 23° after multiple pregnancies [25]. Reports by Blount and Danielsson, on the other hand, demonstrated no relationship between number of pregnancies and curve progression [26, 36].

The quality of evidence for AIS-related spinal changes during pregnancy was low (Table 2).

2. Summary of findings:

- Many patients can expect to experience non-disabling back pain during their pregnancy, which typically resolves following delivery.
- Some women will experience a minor, but statistically significant curve progression during and after pregnancy; however, the clinical significance of these changes is unclear.

3. What anesthetic and obstetric considerations are relevant in AIS mothers delivering a child?

This question investigates: (1) anesthetic considerations, including type and effectiveness, and (2) delivery details, including route and complications. Eight studies provide insight into the former, while 11 address the latter (Table 3).

Bauchat et al. compared 41 surgically corrected females with AIS to 41 controls and found differences in the type and effectiveness of peripartum anesthesia [16]. Traditional epidural anesthesia was delivered in 39% of patients with spinal instrumentation relative to 5% of controls—the

remainder receiving the preferred combined spinal anesthesia. Additionally, the time required to achieve neuraxial anesthesia was 40% longer among instrumented patients. Despite these drawbacks, the failure rate of neuraxial anesthesia in this population remained modest (12%), and no serious complications were reported.

In a cross-sectional comparison by Falick-Michaeli and coauthors, 70% of surgically corrected AIS women were refused spinal anesthesia (citing an absence of catheter access site), relative to 0% of non-AIS controls [19]. Two older articles demonstrated relatively discouraging success rates for regional anesthesia in surgically corrected AIS patients. In 1985, Hubbert et al. reported a 53% success rate for epidural anesthesia [37]. In 1989, Crosby et al. reported a 75% success rate, though three minor complications were noted (2 unsuccessful attempts and 1 dural puncture) [38]. A separate report by Daley and coauthors in 1990 showed successful infusion of epidural anesthesia in 18 of 19 pregnancies; however, results were tempered somewhat by the fact that 8 (42%) required multiple attempts and only 9 (47%) patients reported adequate levels of anesthesia [34].

More recently, Smith and colleagues described an 81% success rate of spinal anesthesia among patients with an instrumented fusion. Of note, in scoliotic patients without prior instrumentation, successful spinal analgesia was achieved in all patients for whom it was attempted [23]. In 2012, a study by Lebel et al. found an equivalent rate of epidural anesthesia administration between 98 conservatively treated scoliotic women and matched controls [17].

Regarding delivery modality, four studies compared the rate of cesarean section between patients with AIS and controls. Three studies found no difference in cesarean delivery rate (12–15%) [16, 20, 26], while the other by Lebel et al. did report an elevated rate in AIS relative to controls (21 vs. 13%) [17]. The remaining studies report frequency of C-section delivery, however, a comparison with controls was not conducted. Generally, lower rates of C-section are reported across the older studies, while more recent publications describe higher rates. Overall, the range of C-section deliveries was 2–41% [19, 22–24, 28, 30, 37, 39, 40] (Table 3).

No differences were reported in rates of preeclampsia, polyhydramnios, or premature rupture of membranes [17, 24]. Additionally, the duration of labor among AIS patients was similar to that of healthy controls [16].

The quality of available evidence for this clinical question was low to moderate (Table 3).

3. Summary of findings:

- Successful spinal analgesia can be achieved in patients with instrumented scoliosis correction, though failed attempts and minor, reversible

complications occur with greater frequency than in non-instrumented patients and healthy controls.

- AIS patients can expect similar rates of cesarean delivery as their non-AIS counterparts.
- Perinatal complication rates are not elevated in patients with AIS.

Discussion

In this report, we provide a broad and comprehensive examination of the relationship between adolescent idiopathic scoliosis and pregnancy. After screening 134 articles, 22 studies were reviewed, incorporating over 3125 AIS patients.

The psychosocial impact of the diagnosis of a major medical condition on young women has been well described for many conditions, including scoliosis generally and AIS specifically [29, 41–45]. Accordingly, a patient with AIS weighing treatment options can be expected to demonstrate concern about her ability to conceive, carry, and deliver a child. The physical strain placed on a woman's body during gestation and delivery, makes this skeletal abnormality especially troubling for hopeful mothers.

The intersection between pregnancy and AIS is as complex as it is important, and pediatric spine specialists should be equipped to fluently discuss the topic with patients and their families. In this review, we offered three fundamental questions encountered by spine surgeons consulting on patients with AIS. In a stepwise fashion, the literature was interrogated, and evidence was coalesced to produce concise, direct answers, while remaining mindful of the quality of evidence available to draw such conclusions.

Pregnancy outcome

Young women with AIS hoping to have children can be reassured that their diagnosis does not preclude them from becoming a mother, but should be cautioned that their chances of pregnancy may be slightly less than that of their peers. As with nearly every conclusion drawn by this review, such counseling should be delivered while acknowledging that the best available medical evidence is non-definitive. Several important qualifiers are to be considered when interpreting this data: (1) most studies did not indicate whether AIS women were attempting or desiring to become pregnant, (2) whether all patients were followed until menopause is unclear, and (3) marital rates were not uniform across studies (and often unreported). Ignoring these three considerations risks misinterpretation of higher

nulliparity figures among AIS women and incorrectly counseling hopeful mothers.

Since the mid-twentieth century, baseline nulliparity rates among women surviving childbearing age have remained stable over time and across populations, hovering near 20% [46, 47]. It is worth emphasizing that all but two authors [21, 26] reported higher rates of nulliparity in AIS populations. And given that AIS mothers utilized significantly higher rates of infertility treatment, it is possible that nulliparity rates may have been higher among the AIS cohorts in the absence of such technology. Nonetheless, the number of children delivered between AIS and non-AIS mothers was equivalent in all but one report.

Importantly, the effect of scoliosis on twin and triplet pregnancy was not encountered in this review and, therefore, remains unclear. Conservatively, these pregnancies may be considered high-risk and followed closely by obstetricians and spine specialists alike. Considering the skeletal changes sustained during normal gestation and childbirth, it may be reasonably hypothesized that women with scoliosis are at higher risk of premature delivery. However, our findings do not support this theory and women with single-fetus pregnancies can be cautiously reassured that their diagnosis alone is not accompanied by a higher likelihood of prematurity.

Spinal changes

The role of physical strain and other biomechanical forces on the pathogenesis of non-adolescent and degenerative scoliosis has been well described [48, 49]. While some features overlap, the pathophysiology of AIS is distinct, incorporating genetic and hormonal factors [50, 51]. The physical burden that a fetus places on the spine of a mother with underlying AIS might be expected to promote curve progression in a similar fashion to adult spinal deformity. The results from this review offer little insight into how such structural changes occur; only that small changes do occur with varying degrees of permanence.

Back pain is common in pregnancy; between 25 and 65% of healthy, pregnant women report moderate or severe back pain [52, 53]. Our results indicate that AIS mothers, on average, experience somewhat higher rates of back pain during their pregnancy. However, in the majority of patients, the pain does not impair the patient's ability to conduct daily activities. Interestingly, patients requiring surgical correction did not experience more back pain during pregnancy than those treated conservatively or with bracing (though, the comparisons did not correct for original curve severity). This is particularly important to note when counseling an anxious patient with a severe or progressive curve facing the prospect of long-segment instrumentation. As with the general population, our

review indicates that the majority of pregnancy-related back pain in AIS patients resolves following delivery.

Major changes in curve progression were not observed across studies. However, minor to moderate curve progression was described in four studies [20, 24, 25, 35, 36], and related to distinct entities: baseline curve severity $>50^\circ$ [24], age <23 years at first pregnancy [20], and instability of curve antepartum [36]. Importantly, disagreement existed across studies regarding whether, and the degree to which these entities contribute to curve progression. While intriguing, the quality of the data is too fragile to endorse firm recommendations for women falling into one or more of these categories [54]. Nonetheless, the observations should serve as motivation for the design and implementation of larger, prospective studies examining such variables. The designation of a 'high-risk' subgroup more susceptible to significant curve progression—or even permanent back pain—would be tremendously useful for surgical counseling.

Anesthetic and obstetric considerations

Pain and discomfort are among the principle concerns of women approaching childbirth, and neuraxial anesthesia is accepted as the most effective form of labor analgesia [55]. Historically, rates of successful neuraxial analgesia in patients with spinal instrumentation lingered near 50% [37, 38]. Within the last two decades, however, the combination of improved spinal surgical methods and more advanced anesthetic techniques has been met with superior analgesic outcomes [16, 17]. While procedure duration is somewhat longer and multiple attempts are more frequent, the vast majority of AIS women with spinal instrumentation do achieve similar pain control as their peers.

This review also discounts the notion that women with AIS—surgically corrected or otherwise—are more likely to undergo cesarean delivery. Except for a single retrospective, population-based cohort, all studies comparing AIS with normal women found equivalent c-section rates. Between, 1965 and today, the c-section rate nationally has increased more than sixfold to 32%, likely secondary to the casual perception of surgery, efforts to maximize efficiency and optimize timing, and the litigious healthcare environment [56, 57]. Our results, however, demonstrated rates much lower (12–24%) among AIS patients in the modern era [16, 17, 19, 22, 28]. Additionally, perinatal complication rates matched those of the general population.

Limitations and future directions

As with any systematic review, the strength of our conclusions relied heavily upon the quality of evidence available for analysis. We encountered a preponderance of

retrospective case series and cohort studies as well as poorly matched case–control studies. No study met CEBM criteria for Level 1 status. Patient heterogeneity across studies and inconsistent data reporting precluded data merging for quantitative comparisons. This was especially true for variables related to curvature and surgical approach—both of which certainly influence obstetric and anesthetic considerations. As a result, while subjective consistencies germane to the three study questions permitted coherent conclusions, the quality of evidence underlying each conclusion itself was low.

Nonetheless, our findings offer clarity on this subject and provide a framework on which future studies might expand. As we consider the treatment options available to these young women, it will be important to look beyond the patient's post-op and recovery period and toward their role as potential mothers in the not-so-distant future. Prospective cohort studies as well as larger registry-based collaborations must incorporate these patient-reported quality metrics as key endpoints. Moreover, if we are to learn about the influence of curve classification and surgical approach (e.g., posterior long-segment fusion, selective thoracic fusion) on obstetric and quality of life outcomes, surgeon-scientists must include these factors among the analyzed covariates. Only then will pediatric spine specialists be armed with higher quality evidence to more assuredly guide these patients and confidently answer these important questions with greater certainty.

Conclusions

The literature relating pregnancy with adolescent idiopathic scoliosis is thin and varied, however, important clinical patterns are observed. While most women with AIS are able to have children, they may encounter more difficulty conceiving than do their peers. Non-disabling back pain is common in AIS women during pregnancy, but spinal changes as a result of pregnancy are typically minor and transient. Perinatal complications—including failed neuraxial anesthesia—are not more common in AIS patients. Future high-quality studies are needed to better elucidate these relationships to help guide the delivery of care in this important population.

Compliance with ethical standards

Funding No funding was received for this work.

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval This article does not contain any studies with human participants or animals performed by any of the authors.

Informed consent The study design consisted of a literature review of previously published data; no human participants were directly involved in any aspect of this study. Informed consent, therefore, was not applicable.

References

1. Negrini S, Aulisa AG, Aulisa L et al (2012) 2011 SOSORT guidelines: orthopaedic and rehabilitation treatment of idiopathic scoliosis during growth. *Scoliosis* 7:3. doi:[10.1186/1748-7161-7-3](https://doi.org/10.1186/1748-7161-7-3)
2. Hresko MT (2013) Clinical practice. Idiopathic scoliosis in adolescents. *N Engl J Med* 368:834–841. doi:[10.1056/NEJMcp1209063](https://doi.org/10.1056/NEJMcp1209063)
3. Asher MA, Burton DC (2006) Asher_review. *Scoliosis* 1:2–10. doi:[10.1186/1748-7161-1-2](https://doi.org/10.1186/1748-7161-1-2)
4. Stirling AJ, Howel D, Millner PA et al (1996) Late-onset idiopathic scoliosis in children 6–14 years old. A cross-sectional prevalence study. *J Bone Jt Surg Am* 78:1330–1336
5. Dickson RA (1983) Scoliosis in the community. *Br Med J (Clin Res Ed)* 286:615–618
6. Gore DR, Passahl R, Sepic S, Dalton A (1981) Scoliosis screening: results of a community project. *Pediatrics* 67:196–200
7. Roach JW (1999) Adolescent idiopathic scoliosis. *Orthop Clin N Am* 30:353–365 (vii–viii)
8. Konieczny MR, Senyurt H, Krauspe R (2013) Epidemiology of adolescent idiopathic scoliosis. *J Child Orthop* 7:3–9. doi:[10.1007/s11832-012-0457-4](https://doi.org/10.1007/s11832-012-0457-4)
9. Weinstein SL, Dolan LA, Cheng JCY et al (2008) Adolescent idiopathic scoliosis. *Lancet* 371:1527–1537. doi:[10.1016/S0140-6736\(08\)60658-3](https://doi.org/10.1016/S0140-6736(08)60658-3)
10. Raggio CL (2006) Sexual dimorphism in adolescent idiopathic scoliosis. *Orthop Clin N Am* 37:555–558. doi:[10.1016/j.ocl.2006.09.010](https://doi.org/10.1016/j.ocl.2006.09.010)
11. Watanabe K, Michikawa T, Yonezawa I et al (2017) Physical activities and lifestyle factors related to adolescent idiopathic scoliosis. *J Bone Jt Surg Am* 99:284–294. doi:[10.2106/JBJS.16.00459](https://doi.org/10.2106/JBJS.16.00459)
12. Moher D, Shamseer L, Clarke M et al (2015) Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015 statement. *Syst Rev* 4:1–9. doi:[10.1186/2046-4053-4-1](https://doi.org/10.1186/2046-4053-4-1)
13. Howick J, Chalmers I, Glasziou P et al (2011) The Oxford 2011 levels of evidence. Oxford Centre for Evidence-Based Medicine. <http://www.cebm.net/index.aspx?o=5653>. Accessed 27 June 2016
14. Atkins D, Best D, Briss PA et al (2004) Grading quality of evidence and strength of recommendations. *BMJ* 328:1490. doi:[10.1136/bmj.328.7454.1490](https://doi.org/10.1136/bmj.328.7454.1490)
15. Schünemann HJ, Jaeschke R, Cook DJ et al (2006) An official ATS statement: grading the quality of evidence and strength of recommendations in ATS guidelines and recommendations. *Am J Respir Crit Care Med* 174:605–614. doi:[10.1164/rccm.200602-197ST](https://doi.org/10.1164/rccm.200602-197ST)
16. Bauchat JR, McCarthy RJ, Koski TR, Wong CA (2015) Labor analgesia consumption and time to neuraxial catheter placement in women with a history of surgical correction for scoliosis: a case-matched study. *Anesth Analg* 121:981–987. doi:[10.1213/ANE.0000000000000690](https://doi.org/10.1213/ANE.0000000000000690)
17. Lebel DE, Sergienko R, Wiznitzer A et al (2012) Mode of delivery and other pregnancy outcomes of patients with documented scoliosis. *J Matern Fetal Neonatal Med* 25:639–641. doi:[10.3109/14767058.2011.598587](https://doi.org/10.3109/14767058.2011.598587)
18. Goldberg MS, Mayo NE, Poitras B et al (1994) The Ste-Justine adolescent idiopathic scoliosis cohort study. Part I: description of the study. *Spine* 19:1551–1561

19. Falick-Michaeli T, Schroeder JE, Barzilay Y et al (2015) Adolescent idiopathic scoliosis and pregnancy: an unsolved paradigm. *Glob Spine J* 5:179–184. doi:[10.1055/s-0035-1552987](https://doi.org/10.1055/s-0035-1552987)
20. Cochran T, Nachemson A (1985) Long-term anatomic and functional changes in patients with adolescent idiopathic scoliosis treated with the Milwaukee brace. *Spine* 10:127
21. Lange JE, Steen H, Gunderson R, Brox JI (2011) Long-term results after Boston brace treatment in late-onset juvenile and adolescent idiopathic scoliosis. *Scoliosis* 6:18. doi:[10.1186/1748-7161-6-18](https://doi.org/10.1186/1748-7161-6-18)
22. Takayama K, Nakamura H, Matsuda H (2009) Quality of life in patients treated surgically for scoliosis: longer than 16-year follow-up. *Spine* 34:2179–2184. doi:[10.1097/BRS.0b013e3181abf684](https://doi.org/10.1097/BRS.0b013e3181abf684)
23. Smith PS, Wilson RC, Robinson APC, Lyons GR (2003) Regional blockade for delivery in women with scoliosis or previous spinal surgery. *Int J Obstet Anesth* 12:17–22
24. Betz RR, Bunnell WP, Lambrecht-Mulier E, MacEwen GD (1987) Scoliosis and pregnancy. *J Bone Jt Surg Am* 69:90–96
25. Ascani E, Bartolozzi P, Logroscino CA et al (1986) Natural history of untreated idiopathic scoliosis after skeletal maturity. *Spine* 11:784–789
26. Danielsson AJ, Nachemson AL (2001) Childbearing, curve progression, and sexual function in women 22 years after treatment for adolescent idiopathic scoliosis: a case-control study. *Spine* 26:1449–1456
27. Akazawa T, Minami S, Kotani T et al (2012) Health-related quality of life and low back pain of patients surgically treated for scoliosis after 21 years or more of follow-up. *Spine* 37:1899–1903. doi:[10.1097/BRS.0b013e31825a22c2](https://doi.org/10.1097/BRS.0b013e31825a22c2)
28. Orvoma E, Hiilesmaa V, Poussa M et al (1997) Pregnancy and delivery in patients operated by the Harrington method for idiopathic scoliosis. *Eur Spine J* 6:304–307. doi:[10.1007/BF01142675](https://doi.org/10.1007/BF01142675)
29. Kersten I, Lange AE, Haas JP et al (2014) Chronic diseases in pregnant women: prevalence and birth outcomes based on the SNIIP-study. *BMC Pregnancy Childbirth* 2014:14:1–14:75. doi:[10.1186/1471-2393-14-75](https://doi.org/10.1186/1471-2393-14-75)
30. To WW, Wong MW (1996) Kyphoscoliosis complicating pregnancy. *Int J Gynaecol Obstet* 55:123–128
31. Goldberg MS, Mayo NE, Levy AR et al (1998) Adverse reproductive outcomes among women exposed to low levels of ionizing radiation from diagnostic radiography for adolescent idiopathic scoliosis. *Epidemiology* 9:271–278
32. Lange J, Steen H, Brox J (2009) Long-term results after Boston brace treatment in adolescent idiopathic scoliosis. *Scoliosis* 4:17. doi:[10.1186/1748-7161-4-17](https://doi.org/10.1186/1748-7161-4-17)
33. Bjerkreim I, Steen H, Brox JI (2007) Idiopathic scoliosis treated with Cotrel–Dubousset instrumentation: evaluation 10 years after surgery. *Spine* 32:2103–2110. doi:[10.1097/BRS.0b013e318145a54a](https://doi.org/10.1097/BRS.0b013e318145a54a)
34. Daley MD, Rolbin SH, Hew EM et al (1990) Epidural anesthesia for obstetrics after spinal surgery. *Reg Anesth* 15:280–284
35. Berman AT, Cohen DL, Schwentker EP (1982) The effects of pregnancy on idiopathic scoliosis. A preliminary report on eight cases and a review of the literature. *Spine* 7:76–77
36. Blount WP, Mellencamp D (1980) The effect of pregnancy on idiopathic scoliosis. *J Bone Jt Surg Am* 62:1083–1087
37. Hubbert CH (1985) Epidural anesthesia in patients with spinal fusion. *Anesth Analg* 64:843
38. Crosby ET, Halpern SH (1989) Obstetric epidural anaesthesia in patients with Harrington instrumentation. *Can J Anaesth* 36:693–696. doi:[10.1007/BF03005423](https://doi.org/10.1007/BF03005423)
39. Maruyama T (2008) Bracing adolescent idiopathic scoliosis: a systematic review of the literature of effective conservative treatment looking for end results 5 years after weaning. *Disabil Rehabil* 30:786–791. doi:[10.1080/09638280801889782](https://doi.org/10.1080/09638280801889782)
40. Weinstein SL, Zavala DC, Ponseti IV (1981) Idiopathic scoliosis: long-term follow-up and prognosis in untreated patients. *J Bone Jt Surg Am* 63:702–712
41. Freidel K, Petermann F, Reichel D et al (2002) Quality of life in women with idiopathic scoliosis. *Spine* 27:E87–E91
42. Freidel K, Reichel D, Steiner A et al (2002) Idiopathic scoliosis and quality of life. *Stud Health Technol Inform* 88:24–29
43. Payne WK, Ogilvie JW, Resnick MD et al (1997) Does scoliosis have a psychological impact and does gender make a difference? *Spine* 22:1380–1384
44. Lim AS, Stewart K, Abramson MJ et al (2012) Asthma during pregnancy: the experiences, concerns and views of pregnant women with asthma. *J Asthma* 49:474–479. doi:[10.3109/02770903.2012.678024](https://doi.org/10.3109/02770903.2012.678024)
45. Weckesser A, Denny E (2013) Women living with epilepsy, experiences of pregnancy and reproductive health: a review of the literature. *Seizure* 22:91–98. doi:[10.1016/j.seizure.2012.11.001](https://doi.org/10.1016/j.seizure.2012.11.001)
46. Hillier TA, Rizzo JH, Pedula KL et al (2003) Nulliparity and fracture risk in older women: the study of osteoporotic fractures. *J Bone Miner Res* 18:893–899. doi:[10.1359/jbmr.2003.18.5.893](https://doi.org/10.1359/jbmr.2003.18.5.893)
47. Opdahl S, Alsaker MDK, Janszky I et al (2011) Joint effects of nulliparity and other breast cancer risk factors. *Br J Cancer* 105:731–736. doi:[10.1038/bjc.2011.286](https://doi.org/10.1038/bjc.2011.286)
48. Veldhuizen AG, Wever DJ, Webb PJ (2000) The aetiology of idiopathic scoliosis: biomechanical and neuromuscular factors. *Eur Spine J* 9:178–184. doi:[10.1007/s005860000142](https://doi.org/10.1007/s005860000142)
49. Schultz AB (1984) Biomechanical factors in the progression of idiopathic scoliosis. *Ann Biomed Eng* 12:621–630
50. Lombardi G, Akoume M-Y, Colombini A et al (2011) Biochemistry of adolescent idiopathic scoliosis. *Adv Clin Chem* 54:165–182
51. Wang WJ, Yeung HY, Chu WC-W et al (2011) Top theories for the etiopathogenesis of adolescent idiopathic scoliosis. *J Pediatr Orthop* 31:S14–S27. doi:[10.1097/BPO.0b013e3181f73c12](https://doi.org/10.1097/BPO.0b013e3181f73c12)
52. Skaggs CD, Prather H, Gross G et al (2007) Back and pelvic pain in an underserved United States pregnant population: a preliminary descriptive survey. *J Manip Physiol Ther* 30:130–134. doi:[10.1016/j.jmpt.2006.12.008](https://doi.org/10.1016/j.jmpt.2006.12.008)
53. Mousavi SJ, Parnianpour M, Vleeming A (2007) Pregnancy related pelvic girdle pain and low back pain in an Iranian population. *Spine* 32:E100–E104. doi:[10.1097/01.brs.0000254123.26649.6e](https://doi.org/10.1097/01.brs.0000254123.26649.6e)
54. Bettany-Saltikov J, Weiss H-R, Chockalingam N et al (2015) Surgical versus non-surgical interventions in people with adolescent idiopathic scoliosis. *Cochrane Database Syst Rev*. doi:[10.1002/14651858.CD010663.pub2](https://doi.org/10.1002/14651858.CD010663.pub2)
55. Anim-Somuah M, Smyth RM, Jones L (2011) Epidural versus non-epidural or no analgesia in labour. *Cochrane Database Syst Rev*. doi:[10.1002/14651858.CD000331.pub3](https://doi.org/10.1002/14651858.CD000331.pub3)
56. Taffel SM, Placek PJ, Liss T (1987) Trends in the United States cesarean section rate and reasons for the 1980–85 rise. *Am J Public Health* 77:955–959
57. Hamilton BE, Martin JA, Osterman MJK, Curtin SC (2015) Births: preliminary Data for 2014. *Natl Vital Stat Rep* 64:1–19