OPEN Research Article

Fetal Scoliosis: Natural History and Outcomes

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ABSTRACT

Introduction: Scoliosis can be detected on prenatal ultrasonography and may be associated with structural and syndromic abnormalities. Associations and pregnancy outcomes related to the prenatal diagnosis of scoliosis are poorly understood.

Methods: A retrospective cohort study was undertaken at a tertiary referral center in London. Referred cases with spinal deformities between 1997 and 2021 were identified from the prenatal ultrasonography database. Outcomes were ascertained from the database and electronic notes.

Results: One hundred twenty-three cases of fetal spinal deformities (scoliosis, kyphosis, or kyphoscoliosis) were identified from a referral population of 660,000 pregnancies, giving an incidence of approximately 0.2 per 1000 fetuses. Fifty-eight live births (47.2%) and 65 cases (52.8%) of fetal or neonatal demise or termination were observed. Most live births were isolated spinal deformities with a good postnatal outcome (n = 35, 60.3%). The commonest syndromic diagnosis in this group was VACTERL association (n = 7, 12.1%). Most cases of fetal loss were associated with severe malformations, most commonly spina bifida, body stalk anomaly and amniotic band sequence, or chromosomal abnormalities, except in 2 cases (3.1%).

Conclusions: This is the largest reported cases series to date of prenatally diagnosed fetal spinal deformity. This confirms that fetal scoliosis and associated vertebral abnormalities are underdiagnosed prenatally, with the reported incidence (0.2 per 1000) lower than the recognized incidence of congenital scoliosis (1 in 1,000). The concurrent finding of severe malformations was strongly associated with fetal loss. When an isolated finding, most fetal spinal deformities had a good postnatal outcome, while 1:8 live births were diagnosed with VACTERL association.

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The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

JAAOS Glob Res Rev 2024;8: e24.00093

DOI: 10.5435/JAAOSGlobal-D-24-00093

Congenital scoliosis is a spinal deformity present at birth, characterized by lateral curvature of the spine, present in up to one in 1,000 newborns.[1-3] Fetal scoliosis develops in the early stages of pregnancy, is characterized by an imbalance in longitudinal growth,[4,5] and can be associated with incomplete formation of vertebrae (hemivertebra), failure of

segmentation (bony bar), or a combination of both.[2,6,7] These abnormalities can be in isolation or associated with other abnormalities or syndromes and can be hereditary or sporadic.[6,8]

When detected after birth, congenital scoliosis is most commonly found in children within the first year of life or in the early teenage years.[5,9] Treatment of these children depends on various factors, including age, the severity and progression of the curve, and the underlying cause, and is nonsurgical in most cases.[3] Treatment typically involves investigation for any associated abnormalities, observation over time, and modifications and growth modulation techniques such as external bracing treatment.[2,10,11] A small proportion of patients (5% according to one study) have surgical intervention if the curve is severe or continues to progress, most commonly individuals in whom the diagnosis is made in early adolescence.[9]

Fetal scoliosis as diagnosed on an ultrasonography scan is typically detected at around 20 weeks' gestation at the planned anomaly scan, although it may be detected earlier.[12,13] There is a paucity of literature on fetal scoliosis, which consists mainly of single reports or limited series,[13-16] and little is known about the effect and natural history of this prenatal diagnosis, which may lead to termination of pregnancy.[13] If parents receive a diagnosis of fetal scoliosis, at the present time, a clinician will have difficulty counselling them about its implications, and the family may not be sufficiently supported in decision making.

This study aimed to evaluate the incidence and outcomes of fetal scoliosis and spinal abnormalities, in particular the rate of live births, terminations, intrauterine fetal demise (IUFD) and early neonatal death, as well as other associated abnormalities.

Methods

Our center is a large volume tertiary referral unit for fetal medicine, which serves a population with approximately 30,000 births per year, and a consistent referral process has been in place across the past two decades. If a fetal anomaly is detected in a peripheral center, they are referred to the fetal medicine unit, where they have additional assessment, which includes ultrasonography scanning and additional procedures when indicated.[17,18] All ultrasonography reports are stored on a database which started in 1997, which initially lacked completeness, but has complete data since 2000, providing for a 22-year period at the time of writing.

The computerized fetal medicine ultrasonography database (Viewpoint) at St George's University Hospital, London, was retrospectively analyzed and interrogated for potentially relevant cases. The text of the ultrasonography scan reports was searched for the terms "spine" and "scoliosis", and for selected tick boxes for spinal abnormalities used at the anomaly scan. The reports were manually reviewed to verify the diagnosis and to exclude duplicates and scans that showed no spinal abnormalities. Any cases with a spinal deformity (scoliosis, kyphosis, kyphoscoliosis, or hemivertebra) were included, irrespective of other anomalies present to the spine or elsewhere. Additional information was collected from ultrasonography reports and notes on events and outcomes of the pregnancy, as well as maternal demographics. The study was registered and approved locally. The NHS Research Ethics Committee decision tool excluded need for an ethical review.

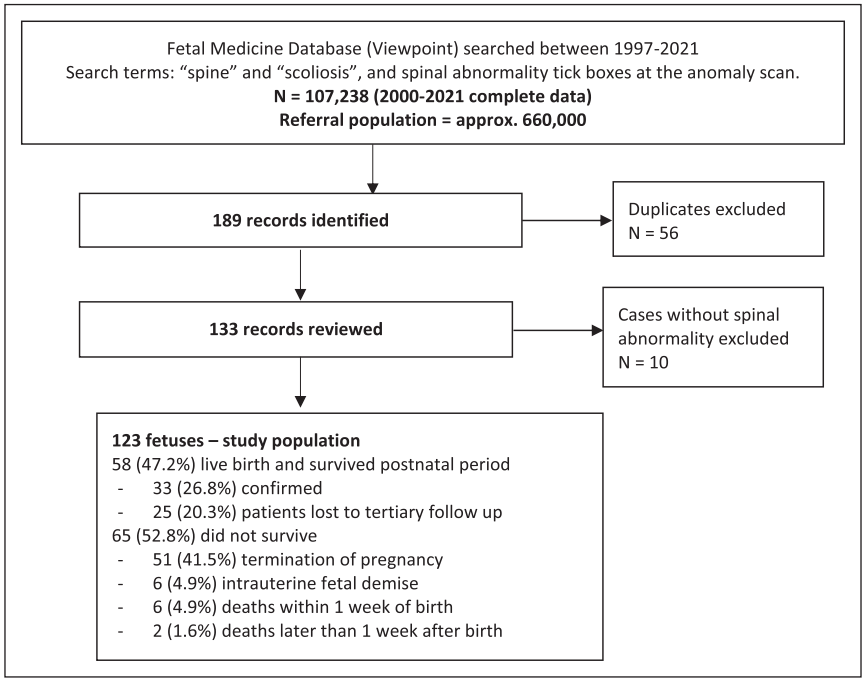
The cases were separated according to outcome (live birth versus fetal loss). For the live births, the local electronic notes were searched for any records of outcomes in childhood. For the nonsurviving fetuses, the notes and scan reports were reviewed to classify the abnormalities as either chromosomal abnormalities, multiple organ abnormalities with normal or untested karyotype, developmental structural abnormalities, or isolated spinal abnormality. Quantitative data (maternal age and gestational age at scan) were compared between groups using an unpaired two-tailed *t*-test.

Results

The database was evaluated between 1997 and 2021, and identified 189 scans. After removal of duplicate patients, 133 remained. After manual checking, 10 additional patients were removed because they did not have a fetal spinal abnormality, which left a study population of 123 patients (Figure 1). Accurate data were available for a 22-year period (2000 to 2021), with a total of 107,238 pregnancies during this period. However, this only included patients local to our center and those referred for additional evaluation. The total referral population of our region is approximately 30,000 pregnancies per year, giving a total referral population of 660,000 pregnancies during our study period. Therefore, the overall incidence of fetal scoliosis was approximately 0.2 per 1000 (1 in 5,000).

Of the 123 fetuses, 58 (47.2%) were successfully delivered and survived the postnatal period, and 65

Figure 1

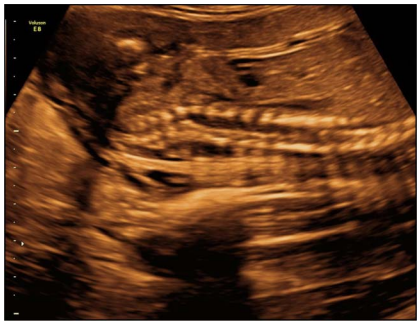


Flowchart of methodology.

(52.8%) did not. The mean maternal age at pregnancy was 30.7 years (range 17.2 to 49.2), with no statistically significant difference between the live birth and fetal loss groups. Of the 58 live births, 33 (56.9%) were confirmed to have had a live birth with no notable concerns, and the remainder (25 patients, 43.1%) were lost to tertiary follow-up—these scans showed no concerns of the pregnancy being at risk, no additional follow-up was arranged at the tertiary center, and they were assumed to have delivered with their local provider. This assumption is made because there is generally a very low threshold for referral to the Fetal Medicine Unit if any concerns arise.

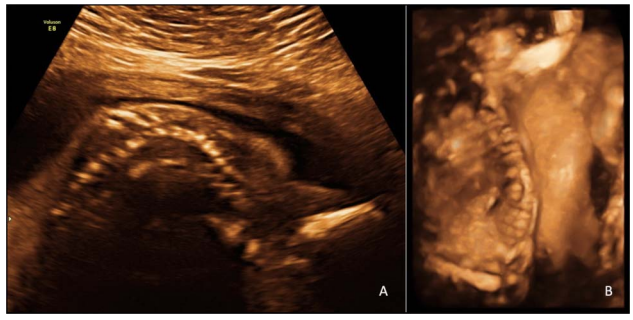
For the live births, the mean gestational age at the scan was 22 + 1 weeks. Of the 58 cases, isolated hemivertebra was reported in 30 scans (51.7%), isolated kyphosis in 4 cases (6.9%), isolated scoliosis in 7 cases (12.1%), kyphoscoliosis in four cases (6.9%), and a combination of hemivertebra with deformity in 13 cases (22.4%) (Figures 2 and 3). Thirty-five cases (60.3%) had no other abnormalities or concerns, and had normal fetal development. The remaining 23 cases (39.6%) had other nonfatal abnormalities, including skeletal, renal, and cardiac abnormalities, and VACTERL association in 7 cases (12.1%)[19,20] (Table 1).

Figure 2



Prenatal ultrasonography scan showing hemivertebra (20 weeks gestatioin).

Figure 3



Prenatal ultrasonography scan showing kyphoscoliosis (26 weeks gestation). **A,** 2D image. **B,** 3D reconstruction.

Table 1. Ultrasonography Findings of 58 Live Birth Cases of Fetal Spinal Abnormalities

Spinal Abnormality	Number (%)	Ultrasonography Scan Findings
All patients	58	33 confirmed (57%), 25 lost to tertiary follow-up (43%)
		35/58 (60%)—normal development, no other abnormalities
		23/58 (40%)—other nonfatal abnormalities
		7 (12%) VACTERL association
Hemivertebra	30 (52%)	21: Normal growth, no other concerns (36%)
		4: VACTERL
		5: Other abnormalities (e.g. talipes, renal agenesis)
Kyphosis	4 (6.9%)	2: Normal development
		1: Talipes
		1: Spina bifida
Scoliosis	7 (12%)	4: Normal development
		1: Increased nuchal translucency
		1: Exomphalos
		1: VACTERL
Kyphoscoliosis	4 (6.9%)	1: No other concerns
		1: VACTERL
		1: Exomphalos
		1: Single umbilical artery
Hemivertebra + deformity	13 (22%)	7: No other concerns (12%)
		2: Renal abnormalities
		1: Single umbilical artery
		1: Ventriculomegaly
		1: VACTERL
		1: Cardiac/vascular abnormalities

On searching local records for the live births, notes were available for 11 patients. Five patients had been referred to the local spinal surgery team for management of scoliosis (tertiary referral center for scoliosis/spinal deformity), and none had undergone a spinal operation (ages 8-16 at the time of writing).

Regarding the cases of fetal and neonatal demise, the mean gestational age at the scan was 18 + 4 weeks, which was significantly earlier than in the live birth group ($P < 0.001$). Eight pregnancies were twins (2 sets of conjoined twins), and three fetuses (4.6%) were confirmed to have a chromosomal abnormality (trisomy nine or 18). Thirty fetuses (46.2%) were characterized as having abnormalities affecting multiple organs, with either normal or absent karyotype or array testing (i.e. no proven chromosomal or genetic abnormalities). Developmental structural abnormalities (e.g. body stalk anomaly, spina bifida, or amniotic band sequence) occurred in 30 fetuses (46.2%). The remaining two fetuses had an isolated spinal abnormality detected on the scan, with normal or absent karyotype testing (Table 2).

Of the 65 cases, 51 (78.5%) had a termination of pregnancy, at a mean gestation of 18 + 3 weeks. The remaining 14 cases that were not electively terminated consisted of 6 cases of IUFD (9.2%), 6 deaths within 1 week of birth (9.2%), and 2 deaths later than 1 week after birth (3.1%). The cases of IUFD included a fetus with multiple notable abnormalities (holoprosencephaly, exomphalos, and limb abnormalities) and a fetus with generalized swelling and oligohydramnios, both with normal karyotype testing. There was also a fetus with associated ventriculomegaly and anhydramnios, and one with exomphalos and limb

Table 2. Ultrasonography Findings and Outcomes of 65 Cases of Fetal Loss With Spinal Abnormalities

Abnormalities	Number (%) + Outcomes	Findings + Outcomes
All patients	65	51 (78%) termination of pregnancy
		6 (9.2%) IUFD
		6 (9.2%) NND within 1 week of birth
		2 (3.1%) NND later than 1 week after birth
Chromosomal abnormalities	3 (4.6%) All termination	2: Trisomy 18—both with multiple abnormalities including kyphoscoliosis
		1: Trisomy 9—multiple abnormalities including scoliosis
Multiple organ abnormalities—Normal testing	20 (31%) 16 termination, 1 IUFD, 1 NND <1 week, 2 NND >1 week	17: Normal karyotype testing
		13: Multiple abnormalities including spinal deformity—termination
		2: Suspected VACTERL—termination
		1: Suspected VACTERL—NND >1 week (3 months)—cardiac abnormality
		1: Scan at 12 weeks, multiple significant abnormalities (holoprosencephaly, exomphalos, limb abnormalities) including kyphoscoliosis—IUFD and ERPC.
		3: Normal karyotype and array testing
		1: Bilateral multicystic renal dysplasia + kyphosis—NND <1 week (delivery at 36 weeks)
		1: Multiple abnormalities including scoliosis—NND >1 week (day 65) (delivery at 32 weeks)
		1: Multiple neuromuscular abnormalities including scoliosis—termination
Multiple organ abnormalities—No testing	10 (15%) 7 terminations, 2 IUFD, 1 NND <1 week	7: No testing performed (declined/not possible)
		1: Features of Holt-Oram syndrome—NND <1 week (delivery at 41 weeks)
		1: Exomphalos, limb abnormalities, scoliosis—IUFD at 29 weeks
		1: Features of VACTERL (and confirmed on postmortem)—termination
		4: Various abnormalities (cardiac, brain, exomphalos, limb)—termination
		3: Testing attempted but failed
		All: Multiple nonspecific abnormalities—2 terminations, 1 IUFD at 28 weeks

(*continued*)

Table 2. (*continued*)

Abnormalities	Number (%) + Outcomes	Findings + Outcomes
Developmental structural abnormalities	30 (46%) 25 terminations, 1 IUFD, 4 NND <1 week	9: Body stalk anomaly, no testing—termination
		1: Body stalk anomaly, no testing—NND <1 week (delivery at 30 weeks)
		1: Body stalk anomaly (monoamniotic twin), no testing—NND <1 week (delivery at 34 weeks)
		6: Spina bifida, no testing—termination
		3: Spina bifida, normal karyotype—termination
		4: Amniotic band sequence + multiple abnormalities, normal karyotype (one also had normal array testing)—termination
		1: Amniotic band sequence + multiple abnormalities (dichorionic diamniotic twin), no testing—NND <1 week (delivery at 32 weeks)
		1: Ventriculomegaly, anhydramnios (PROM), kyphoscoliosis, no testing—IUFD at 27 weeks
		1: Limb abnormalities, oligohydramnios (PROM), scoliosis, normal karyotype—NND <1 week (delivery at 27 weeks)
		1: Renal agenesis, anhydramnios, scoliosis, normal karyotype—termination
		1: Conjoined twins (thoracopagus, single heart and liver)—termination
		1: Conjoined twins (thoraco-omphalopagus, 2 hearts, single pericardium)—termination
Isolated spinal abnormality	2 (3.1%) Both IUFD	1: Scoliosis + hemivertebra, nil else detected, no testing—IUFD at 28 weeks (no additional info)
		1: Scoliosis, generalized fetal swelling and oligohydramnios but no other abnormalities, normal karyotype, negative infection screen—IUFD at 25 weeks

IUFD = intrauterine fetal demise, NND = neonatal death, ERPC = evacuation of retained products of conception, PROM = premature rupture of membranes, VACTERL = VACTERL association.

abnormalities, both with no genetic testing undertaken. In addition, there were two fetuses with a hemivertebra but no other abnormalities detected on the ultrasonography scan—other abnormalities were detected on postmortem for one of these, but no additional information was available for the other. The neonatal deaths were all associated with other abnormalities (Table 3).

Discussion

Comparison With Existing Data

This study represents the largest series to date of prenatally diagnosed fetal scoliosis. Our results show that there is a wide range of associated findings and outcomes with fetal scoliosis, with an overall incidence of

Table 3. **Ultrasonography Findings and Outcomes of 14 Cases of Fetal or Neonatal Loss With Spinal Abnormalities (Terminations Excluded)**

Outcome	Ultrasonography Findings + Outcomes
Intrauterine fetal demise (IUFD) n = 6	Scan at 12 weeks showed multiple significant abnormalities (holoprosencephaly, exomphalos, limb abnormalities) including kyphoscoliosis, normal karyotype testing—IUFD at unknown gestational age
	Scoliosis, generalized fetal swelling and oligohydramnios but no other abnormalities, normal karyotype, negative infection screen—IUFD at 25 weeks
	Ventriculomegaly, anhydramnios (PROM), kyphoscoliosis, no genetic testing—IUFD at 27 weeks
	Exomphalos, limb abnormalities, scoliosis, no genetic testing—IUFD at 29 weeks
	Multiple nonspecific abnormalities (on postmortem), genetic testing attempted but failed—IUFD at 28 weeks
	Scoliosis + hemivertebra, nil else detected, no genetic testing—IUFD at 28 weeks (no additional info)
Neonatal death (NND) within 1 week n = 6	Features of Holt-Oram syndrome, no genetic testing (late booker)—NND <1 week (delivery at 41 weeks)
	Bilateral multicystic renal dysplasia + kyphosis, normal karyotype and array testing—NND <1 week (delivery at 36 weeks)
	Body stalk anomaly, no genetic testing—NND <1 week (delivery at 30 weeks)
	Amniotic band sequence + multiple abnormalities (dichorionic diamniotic twin), no genetic testing—NND <1 week (delivery at 32 weeks)
	Limb abnormalities, oligohydramnios (PROM), scoliosis, normal karyotype—NND <1 week (delivery at 27 weeks)
	Body stalk anomaly (monoamniotic twin), no genetic testing—NND <1 week (delivery at 34 weeks)
Neonatal death (NND) later than 1 week n = 2	Large exomphalos (containing liver, bowels and stomach), cardiac anomaly, scoliosis, normal karyotype and array testing. Delivery at 32 weeks. Pulmonary hypoplasia and pulmonary hypertension, intubated at birth and gradually deteriorated, died on day 65
	Suspected VACTERL, normal karyotype testing. Significant cardiac abnormality, died after 3 months

IUFD = intrauterine fetal demise, NND = neonatal death, PROM = premature rupture of membranes, VACTERL = VACTERL association.

0.2 per 1,000, which is similar to the previous literature.[13,14]

All of the fetuses included in our study had spinal abnormalities present on a prenatal ultrasonography scan. Often no evident spinal deformities are noted on prenatal screening, but after birth, the baby is subsequently found to have a congenital spinal abnormality, with the most common being a hemivertebra. Congenital scoliosis has been reported to have an incidence ranging from to 0.5 to 1 in 1,000,[1-3] and congenital hemivertebra ranging from 0.13 to 0.33 in 1,000[13,14,21] to as high as five in 1,000 by the Fetal Medicine Foundation.[8] This wide range illustrates that spinal abnormalities are variably detected in childhood because of multiple factors. The true incidence of fetal vertebral abnormalities must be higher due to attrition (fetal loss), but this study and the limited existing literature report a lower figure, suggesting underdiagnosis. This may be because less severe spinal abnormalities are not apparent in the early stages of pregnancy and also because of

the inherent diagnostic limitations of ultrasonography scanning as a screening modality.

Prenatal Screening

Ultrasonography is the main imaging modality in pregnancy because it is a safe and effective way to diagnose prenatal abnormalities.[22,23] The main limitations of ultrasonography are that it is dependent on the operator and patient factors such as body habitus, previous surgery, and fetal position.[24-26] A study from 1989 evaluated the sensitivity and specificity of ultrasonography scans for neural tube defects in 237 patients and found 94.7% sensitivity and 98.3% specificity.[12] It is widely accepted that as technology has advanced, accuracy of these scans has improved, and they can reliably pick up abnormalities and show the exact level of a spinal abnormality,[27] and can detect severe spinal abnormalities, even in the first trimester.[28] However, neural tube defects may reveal additional clues to the sonographer (eg, cerebellar abnormalities), whereas the detection of scoliosis requires a high level of expertise with assessment of the spine alone. There is a lack of literature relating to the ability of ultrasonography to detect fetal scoliosis, but as access to imaging has improved, it is now more common to have multiple ultrasonography scans during a pregnancy when there are concerns, further increasing the rate of detection of any abnormalities.

Additional information can be obtained using 3D (three-dimensional) ultrasonography or MRI (magnetic resonance imaging). This can provide a clearer picture of the spine, particularly if the fetal or placental position makes 2D imaging insufficient.[29,30] Fetal MRI has become an increasingly used adjunct[31,32] and has higher diagnostic accuracy than ultrasonography for congenital spine and spinal cord malformations.[33]

When abnormalities are identified on prenatal scans, this is used to counsel parents on the decision to continue or terminate the pregnancy.[13,19,34] Our study provides information which may be useful when counselling families on the associations of spinal abnormalities including hemivertebra, and the risks this can pose to the fetus. Approximately half of the fetuses in our study were delivered successfully and survived, supporting the idea that having a spinal abnormality in itself is by no means guaranteed to confer a poor outcome.

Complex Fetal Spinal Abnormalities

Most unsuccessful pregnancies had ultrasonography scans showing multiple fetal malformations (Table 2). The commonest developmental structural abnormalities were spina bifida (9 cases, 13.8%), body stalk anomaly (11 cases, 15.4%), and amniotic band sequence (5 cases, 7.7%), all of which are relatively easy to diagnose and are associated with poor outcomes.[35-37] Two fetuses were reported to have isolated findings of spinal abnormalities but resulted in IUFD, suggesting that there were likely to be other abnormalities present in these cases that were not detected.

Many syndromes can be associated with spinal abnormalities, including Jarcho-Levin syndrome, Klippel-Feil syndrome, VACTERL association, and OEIS complex.[8,15] VACTERL association consists of cardiac, renal, and limb abnormalities; anal atresia and tracheo-oesophageal fistula; and the vertebral abnormality (usually hemivertebra).[19,20] This often results in notable abnormalities that can make survival impossible; however, in less severe cases, the pregnancy continues and the baby is delivered successfully, as was the case in seven of the VACTERL cases in our study. Typically, VACTERL association is diagnosed later in the pregnancy, or following birth or fetal demise, but our study reinforces findings from multiple case reports that it can be diagnosed on early prenatal ultrasonography.[38]

Isolated Fetal Spinal Deformity

In our cohort, 35 cases (28%) had an isolated spinal deformity detected on the ultrasonography scan without any other abnormalities and went on to live birth with no complications. Twenty-three cases (19%) had a spinal abnormality as well as other nonfatal abnormalities (including 7 with VACTERL association), and went on to live birth. These associated abnormalities were in keeping with other similar studies.[13,16]

Fetal scoliosis (without hemivertebra) is a relatively rare condition with limited existing literature, with only a few case reports that have identified this.[39,40] They again reveal a range of possible outcomes from termination of pregnancy with multiple other abnormalities,[39] through to successful delivery with no other abnormalities.[40] In our study, five patients had ultrasonography scans which detected fetal scoliosis but no other structural abnormalities. Four survived to term, and one resulted in IUFD during the second trimester.

There is also limited literature regarding fetal kyphosis, with a report of a case which had other associated abnormalities including horseshoe kidney, dislocated hips, and dysplastic ears.[41] In our series, only four fetuses had a finding of isolated kyphosis (no evident associated scoliosis or hemivertebra) and all went on to live birth, with two of them having no other abnormalities or concerns; of the other 2, one had talipes and the other had spina bifida. As with the findings of scoliosis, our study shows that this finding does not

necessarily mean a poor outcome, and decisions should be made in the context of any other abnormalities.

Strengths and Limitations

The referral population for this study of 660,000 was an estimate based on a well-established regional set-up,[18] and the true value may be to the order of 50,000 greater or smaller than this. However, this inaccuracy would not drastically change the incidence given the rarity of the condition. It is theoretically possible for cases to not be referred to our center for a second opinion; however, this is extremely unlikely given the fear of medical litigation, and there is no associated cost of referral to the center or the patient in our healthcare system.

The limitations of ultrasonography scanning may have meant that some abnormalities were missed, or complex cases underdiagnosed, as was the case for two fetuses with apparent isolated spinal deformity, but this would not have changed their outcomes as both resulted in spontaneous IUFD. All patients with VACTERL association (11 in total) were prenatally diagnosed, reflecting the experience of the Fetal Medicine service.

Many of the cases in our study underwent genetic testing, but these techniques evolved during the 24-year study period. Karyotype testing was used routinely at the start of the study period, but chromosomal microarray analysis and sequencing for single-gene disorders were introduced and improved during this time, allowing testing for more genetic abnormalities later on, which likely meant that some of the older cases had genetic abnormalities which were not detected.[42]

Other limitations of this study include that it is a retrospective database study, with some records having relatively minimal data, particularly the older entries. Insufficient information was available for many of the live births, or they had not had any additional contact with our center, making it impossible to search for information regarding additional follow-up and outcomes during childhood. For the more recent cases, there has not yet been long enough follow-up to assess childhood outcomes. However, there was still sufficient information to draw conclusions relating to birth outcomes and associations. This study period of 24 years with 123 cases makes this a study of notable magnitude, and the largest series to date evaluating fetal scoliosis.

Conclusions

This study evaluates the incidence and associated outcomes of fetal scoliosis, with a focus on the prenatal findings and how this may affect decision making during the pregnancy. Much of the existing literature describes congenital scoliosis diagnosed after birth, but this may not be helpful for patients and healthcare professionals faced with a diagnosis prenatally.

The incidence of fetal scoliosis was one in 5,000, and approximately half of the fetuses went on to live birth. Of these, 60% had no other notable abnormalities, and the remainder had other nonfatal abnormalities. For the other half of fetuses which did not survive the pregnancy or the early postnatal period, the spinal abnormality generally formed part of a picture of multiple abnormalities which conferred a guarded or very poor prognosis. These findings help to further clarify the outcomes associated with fetal scoliosis and can help guide informed decision making for parents and healthcare professionals.

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