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Epidemiology and Demographics of Adolescent Idiopathic Scoliosis in Brunei Darussalam

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Abstract

Introduction: Data on the characteristics of adolescents with idiopathic scoliosis in Brunei Darussalam and factors relating to risk of curve progression (≥ 5°) are unavailable. This study aimed to investigate the epidemiological, clinical presentation and curve characteristics of patients with adolescent idiopathic scoliosis in Brunei Darussalam, and identify potential factors that can predict the risk of curve progression. **Materials and Methods:** We analysed patients (aged 11-18 years) diagnosed with adolescent idiopathic scoliosis from January 2017 to December 2020. Univariate and multivariate analyses were performed to identify the predictors of curve progression. **Results:** A total of 127 patients (girls = 100, median age = 13.8 years, median Cobb angle = 36° [interquartile range 25°]) were newly diagnosed during the 4-year study period, with an annual crude incidence of 46.9 per 100,000 adolescent population. Curve characteristics were analysed in 81 patients. Of these patients, 26 (32.1%) were surgical candidates at presentation. Twenty of the 55 non-surgical candidates developed curve progression during the study period, of which nine progressed to requiring surgery. At presentation, Risser stage 0-1 correlated with curve progression (Odds ratio, 4.07; confidence interval, 1.05–15.8). **Conclusion:** Adolescent idiopathic scoliosis is more commonly seen in girls than in boys and most cases do not require surgical correction. Additionally, a low Risser stage at presentation predicts curve progression.

Keywords: Adolescent, Epidemiology, Scoliosis, Southeast Asia, Spine diseases

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INTRODUCTION

The term scoliosis is derived from the ancient Greek word ‘skolios’, which means curved or crooked. It is a three-dimensional deformity of the spine and trunk that may include lateral deviation, rotation, or a disturbed sagittal profile.[1] It is the most common spinal disorder in children and adolescents and affects 2–3% of adolescents.[2] According to the Scoliosis Research Society (SRS), diagnosis is made when the major Cobb angle (CA) is ≥ 10° (***Supplementary figure “How to measure Cobb angle?”***). Of note, more than 80% of scoliosis cases are idiopathic and occur only after other causes of scoliosis are excluded. Ninety percent of idiopathic scoliosis cases occurs in adolescence (11–18 years).[1] Nearly 10% of patients with adolescent idiopathic scoliosis (AIS) require some form of treatment and up to 0.1% eventually require surgery.[3] The exact pathogenesis of AIS is unknown. It may be caused by the failure of vertebral formation, hormonal and neuromuscular imbalance, or alteration of soft tissue (connective tissue disorder). [4,5]

The prevalence rate of AIS has been reported to range from 0.35% to 13% owing to the varying curve angle diagnosis criteria. A study of Singaporean schoolchildren showed an overall prevalence rate of idiopathic scoliosis of 0.93% and 0.25% in girls and boys, respectively, with increasing numbers during pubertal growth.[6] Approximately 30% of patients with AIS have a family history of scoliosis. Multiple studies have described patient age, sex, curve characteristics, menarche status, and the Risser stage as important factors for predicting curve progression during growth.[4,7,8] The relative importance of each individual factor is yet to be clearly defined, and it is possible that there is an interplay between these factors in curve progression during and after growth. Menarche status or age provides prognostic information for predicting the risk of curve progression. It is commonly used as an identifiable maturity indicator and is associated with decelerated growth velocity. Studies have shown that the risk of curve acceleration in patients with AIS is noticeably higher before than after menarche. It has also been proposed that older age at menarche is associated with a higher prevalence of AIS.[9-11] Radioimaging of the pelvis is performed to determine growth. Using the Risser stage (0-5), the state of ossification of the iliac apophysis is used to determine skeletal maturity. This score is commonly used to determine the amount of growth left and correlates with the extent to which spinal growth is left.[10] In simple terms, those with Risser stages of 0 and 1 grow more rapidly, and hence, are more likely to have curve progression. In contrast, those with Risser 4-5 have generally little or no growth and are less likely to have curve progression.

As part of the school health services of the Brunei Darussalam Ministry of Health, students from Year 7 (approximately 12–13 years of age) undergo scoliosis screening by trained community nurses. This entails the use of Adam’s forward bending, in which the tested individual is asked to bend forward as if trying to reach their toes.[12] Any asymmetry seen such as uneven shoulders, prominent shoulder blades, hip, and rib arches, is considered positive and the patient is subsequently referred to the scoliosis clinic for further evaluation. In addition, “head to toe” assessments are also conducted for students during Years 1,4 and 6 and any clinical suspicions for scoliosis is also referred for further assessment. The management of scoliosis is either conservative (observation/bracing) for smaller curve angles, or surgical for severe scoliosis. As the mainstay of scoliosis management is to prevent curve progression, it is important for physicians to accurately identify patients who are at risk. Surgical treatment is generally offered to patients with CA > 45°.

No epidemiological studies have been conducted on populations with AIS in Brunei Darussalam. Understanding the differences in the characteristics of patients with AIS will be important to help guide country-specific evidence-based healthcare planning and resource allocation to reduce the burden of the disease. Thus, the objective of this study was to investigate the epidemiology, clinical presentation, and curve characteristics of adolescents with idiopathic scoliosis in Brunei Darussalam and to identify potential factors that can predict the risk of curve progression.

MATERIALS AND METHODS

This was a retrospective cohort study of patients (aged 11–18 years) who were first diagnosed with AIS from 1 January 2017 to 31 December 2020. Ethics approval was obtained from the Institute of Health Science Research Ethics Committee of the University of Brunei Darussalam (Reference: UBD/PAPRSBIHSREC/2021/61). Patients who visited the scoliosis clinic at the Department of Neurosurgery, Brunei Neuroscience, Stroke, and Rehabilitation Centre (BNSRC) during this 4-years period were identified from the scoliosis register. We excluded patients with a non-adolescent presentation age, patients seen for other neurosurgical issues, patients with secondary causes of scoliosis, nonattendees, and those without any note entry.

Using the Brunei Darussalam Healthcare Information and Management System (BruHIMS), we collected data on patient demographics including date of birth, date of first visit to the scoliosis clinic, sex, reason for presentation, symptoms on initial presentation, family history, and age at menarche. From their standing whole spine radiographs, curve type, CA of major curve and Risser stage were recorded. Curve severity (**Figure 1**) was categorised as mild (CA <30°), moderate (CA ≥30° to <50°) or severe (CA ≥50°). Curve progression was defined as an increase in CA of ≥5° between the first and final radiograph.

All statistical analyses were performed using SPSS version 23 (IBM Corp., Armonk, New York, USA). Crude incidence rates were standardised according to adolescent population and based on published population statistics for 2017-2020.[13] The sociodemographic characteristics of the patients were reported. Categorical and numerical datasets are presented as frequency and percentage and as mean (standard deviation) and median (interquartile range) for normal and non-normally distributed variables, respectively. Univariate and multivariate analyses were performed to identify predictors of curve progression. The level of significance was set at $p<0.05$.

RESULTS

A total of 127 patients with newly diagnosed AIS were identified during the 4-year period. The overall annual crude incidence per 100,000 adolescents' population was 46.9 with a girl to boy ratio of 3.7:1. The mean (standard deviation [SD]) age at presentation was 13.8 (1.70) years. A positive family history of scoliosis was observed in 24.4% (31/127) of patients. The main reasons for their presentation to the clinic were body shape concerns from family members (37.8%, 48/127) and school screening (34.6%, 44/127). In the study cohort, 85.8% (109/127) of patients did not report any symptoms at presentation (**Table I**).

Eighty-one patients were assessed for their clinical and radiological progression (**Table II**). The median CA of major curve at presentation was 36° (interquartile range [IQR] 25°). Mild, moderate, and severe scoliosis were seen in 38%, 41%, and 21% of these patients, respectively.

At presentation, 32.1% (26/81) of the patients were identified as surgical candidates (**Figure 2**). Of the 55 non-surgical candidates at presentation, 20 demonstrated curve progression at follow-up and nine developed curve progression leading to surgical candidacy. Examples of patients who had undergone surgery are shown in **Figure 3**.

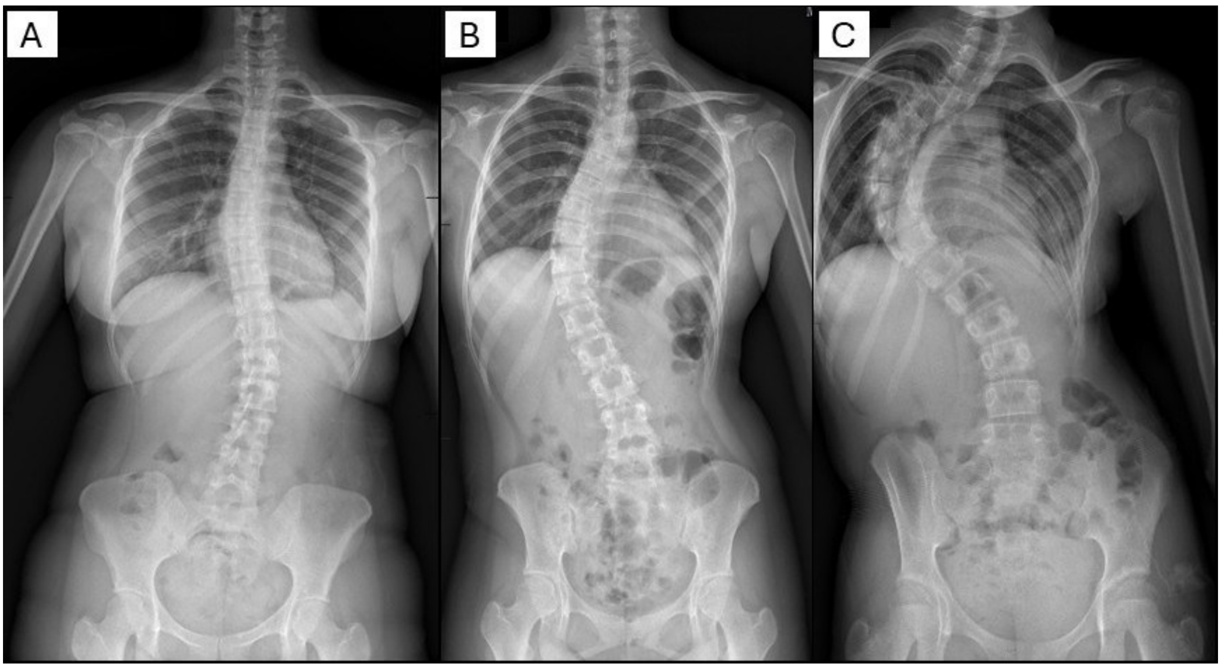

Figure 1: Categorisation of scoliosis curve severe (A) mild, Cobb angle 18°; (B) moderate, Cobb angle 49°; (C) severe, Cobb angle 95°.

Table I: Characteristics of overall adolescent idiopathic scoliosis population (n=127).

Characteristics	n (%)	Media (IQR)
Sex		
Girl	100 (78.7%)	
Boy	27 (21.3%)	
Median (IQR) age at presentation, years	-	13.8 (1.70)*
Reason for presentation		
Family reported	48 (37.8%)	
School screening	44 (34.6%)	
Self-reported	18 (14.2%)	
Incidental finding on chest x-ray	14 (11.0%)	
Others	3 (2.4%)	
Symptoms		
None	109 (85.8%)	
Pain	16 (12.6%)	
Shortness of breath	2 (1.6%)	
Positive family history		
No	96 (75.6%)	
Yes	31 (24.4%)	

IQR= interquartile range. * the distribution is skewed to the right

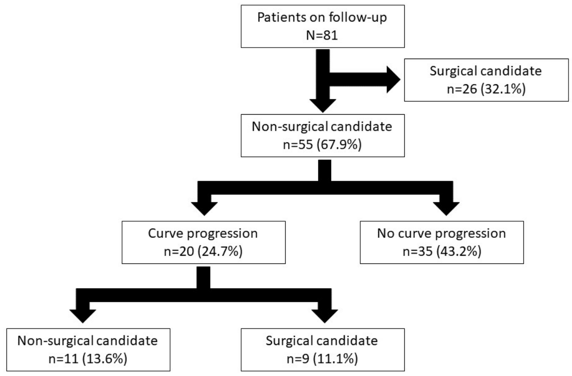

Figure 2: Clinical progress of adolescents with idiopathic scoliosis.

DISCUSSION

The data and results of this study are the first such report in Brunei Darussalam. Our findings revealed the overall annual crude incidence of AIS was 46/100,000 adolescent population.[9] New girl patients with AIS were significantly more, and a lower Risser stage (0-1) at presentation was associated with curve progression.

Epidemiology on the prevalence of idiopathic scoliosis has previously been conducted. A Montreal study reported a prevalence rate of 4.5%, and a prevalence rate of 1.2% was reported in Minnesota, USA.[14,15] A study of Singaporean schoolchildren showed an overall prevalence rate of idiopathic scoliosis of 0.93% and 0.25% in girls and boys, respectively.[6] Most studies have been conducted by estimating the overall prevalence using a screening programme within a specific population such as in schools.[16-18] In our study, an accurate evaluation of the incidence of AIS locally was conducted as we, as the only referral specialist centre providing care for scoliosis, were able to evaluate the entire adolescent population. Underestimation of the true incidence may be an issue as some patients, especially those with mild scoliosis, would not have been detected and referred or some patients may not have attended clinic assessment. However, underdiagnosis is unlikely to be an issue as mandatory school screening programme, consisting of a physical examination for scoliosis, is active in Brunei Darussalam. When there are suspicious findings in the physical examination, students are referred to our centre for further evaluation. Misdiagnosis is also minimal as the diagnosis is made following clinical and radiological evaluation and diagnostic codes are registered only by doctors who specifically conduct scoliosis clinics.

Table II: Scoliosis curve characteristics and progress (n=81).

Characteristics	
Median (IQR) age	13.0 (3)*
Mean (SD) menarche age for girls	11.7 (1.1) Range 9-14
Degree of Scoliosis	
Minor (CA<30°)	31 (38%)
Moderate (CA≥30° to <50°)	33 (41%)
Severe (CA≥50°)	17 (21%)
Median (IQR) CA of major curve at presentation	36.0 (25)*
Risser stage at presentation	
0	9 (11.1%)
1	8 (9.9%)
2	18 (22.2%)
3	16 (19.8%)
4	20 (24.7%)
5	10 (12.3%)
Curve progression ≥5°	
No	50 (61.7%)
Yes	31 (38.3%)
Median (IQR) degree of curve progression	10.0 (8)* Range 5–63

In our study, scoliosis cases among girls were nearly 4 times more than that in boys. Similar findings have been reported in multiple studies using CA> 10° as the threshold. A prevalence study in Singapore showed that the proportion of girls with scoliosis was higher than that of boys at different age groups.[6] Another prospective Korean study of over one million schoolchildren reported an overall prevalence rate of 3.26%, with a higher prevalence in girls than in boys.[19] Despite several studies, the risk of curve progression remains unclear. Reports vary owing to the different criteria or definitions of curve progression. A Greek study on

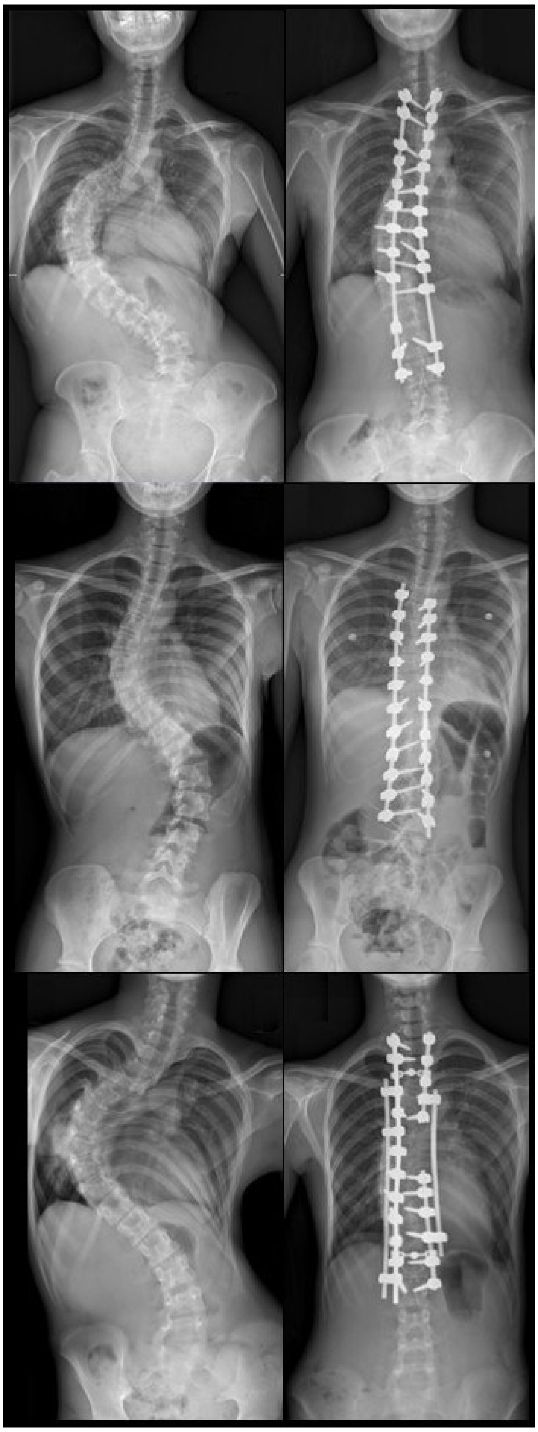

Figure 3: Pre- and post-surgery radiographs of three patients who had undergone scoliosis correction.

children demonstrated progression by ≥5° of the curve in 14.7% (123/839) children with significantly higher incidence in girls. The initial curve magnitude was also associated with a higher incidence of curve progression.[20] In a retrospective study by Lonstein and Carlson, curve progression was detected in 23.2% of 727 children with untreated scoliosis and they also reported that the curve magnitude, skeletal maturity, and curve pattern were associated with curve progression.[21]

Our study found 38.3% (31/81) patients with curve progression of ≥5° as measured by the CA difference between the first and final radiographs on latest follow up or pre surgery. This is consistent with the upper range in previous reports. This could be partly explained by the relatively high median major CA of 36° at presentation. A systematic review revealed the correlation between several individual predictive factors and a higher risk of curve progression. In particular, a higher initial curve magnitude, low skeletal maturity, younger age, and pre menarche state were the most relevant factors identified.[22] Another study looked into patients identified from school screening and it showed that CA of ≥25° at presentation when combined with girls of <12 years and pre-pubertal status has highest risk of curve progression to ≥30° at skeletal maturity.[23] Goldberg et al. also found that the incidence of curve progression was related to a younger age at diagnosis, premenarche status, older age at menarche, and a Risser stage of 0.[24]

Younger patients were at a higher risk of developing curve progression during follow-up (p=0.001). This was supported by previous studies in which the incidence decreased as the child aged.[21,24] It has been pointed out that progression of idiopathic scoliosis tends to occur at the time of most rapid adolescent skeletal growth and younger age is naturally linked to growth and skeletal maturity. Unlike other reports, our study did not show a relationship between age at menarche in girls and the incidence of scoliosis or curve progression. In one Chinese study, it was found that the age of menarche in patients with AIS was significantly older than that in normal control girls (p <0.001). The mean menarche age for healthy normal girls and girls with AIS was 12.63 +/- 0.98 years and 12.83 +/- 1.22 years respectively.[9] One interpretation of how and why late menarche is susceptible to curve progression is that late menarche is correlated with delayed skeletal maturity. This results in a prolonged period of rapid growth and an increased susceptibility to development and progression.[11]

Numerous studies have identified the magnitude of the initial curve as one of the most important risk progression factors.[20,21,23,25] Our study did not show a relationship between high initial major curve magnitude and curve progression. The occurrence of progression was 48% for those with a curve greater than 30° compared to 11.9%–20% for those with a curve between 10 -20°.[20]

Skeletal maturity is another major risk factor for curve progression. The Risser stage is one of the most widely used indicators of skeletal maturity to guide clinicians in predicting curve progression, and the decision and timing of whether to operate. Numerous studies have linked lower Risser stage to an increased likelihood of curve progression.[7,21,24-26] Lonstein and Carlson concluded that the incidence of curve progression decreased as the initial Risser stage increased (30% in patients with Risser stage 0–1 and 11% in those with Risser stage 2-4) [21]. The findings of this study are compatible with those of previous studies. Curve progression was observed in 66.7% (12/18) of patients with Risser stages of 0 and 1 and in 36.8% (21/56) of patients with Risser stages of 2, 3, and 4 (p = 0.026). The Risser staging is not without disadvantages. It correlates with skeletal age differently in boys and girls and typically appears after peak height velocity and pubertal growth have occurred.[27,28] In addition, the previous assumption that a Risser stage of 4 is a point of maturity and cessation of curve progression has been challenged. Histological studies showed that vertebral endplate growth persisted at a Risser stage of 4 and ceased only at a Risser stage of 5.[29]

Overall, the follow-up findings of this study (**Figure 2**) provide clinically relevant and, more importantly, assurance to clinicians, and patients and parents that majority of adolescents in our population who present with idiopathic scoliosis do not need surgery and in the event of curve progression, they are more likely to remain non-surgical candidates. As the sole provider of scoliosis in Brunei Darussalam, these findings reflect the "real world" management of AIS in the country and the findings are as close as possible as to a population-based experience.

This study had several limitations. As this was a retrospective study, it is subject to shortcomings commonly related to a retrospective study design, including reliance on medical records, loss of patient data, and inadequate follow-up assessment, particularly the unavailability of radiographs. Second, we excluded patients who first presented to the scoliosis clinic at age 18 years and above. This may have led to an underestimation of actual incidence of adolescent idiopathic scoliosis.

CONCLUSION

Given the scarcity of AIS data in Brunei Darussalam, this study provides insights into several aspects of AIS in Brunei Darussalam. The annual crude incidence of AIS was 46.9 per 100,000 adolescents. AIS is more common in girls than in boys and most patients do not require surgical correction. A low Risser stage at presentation was a significant predictor of curve progression during follow-up. With these findings, local clinicians can be equipped with knowledge of the natural history of AIS in Brunei Darussalam and better prognosticate and manage patients at risk of curve progression during growth. Research into the burden of AIS on health-related quality of life is currently underway to shed light on this unique adolescent population.

Abbreviations

SRS	Scoliosis Research Society
CA	Cobb angle
AIS	Adolescent idiopathic scoliosis
BNSRC	Brunei Neuroscience, Stroke and Rehabilitation Centre

Declarations

Conflict of interests

The authors declare no conflict of interests.

Ethical Statement

The study was approved by the Institute of Health Science Research Ethics Committee of the University of Brunei Darussalam (Reference: UBD/PAPRSBIHSREC/2021/61).

Acknowledgement

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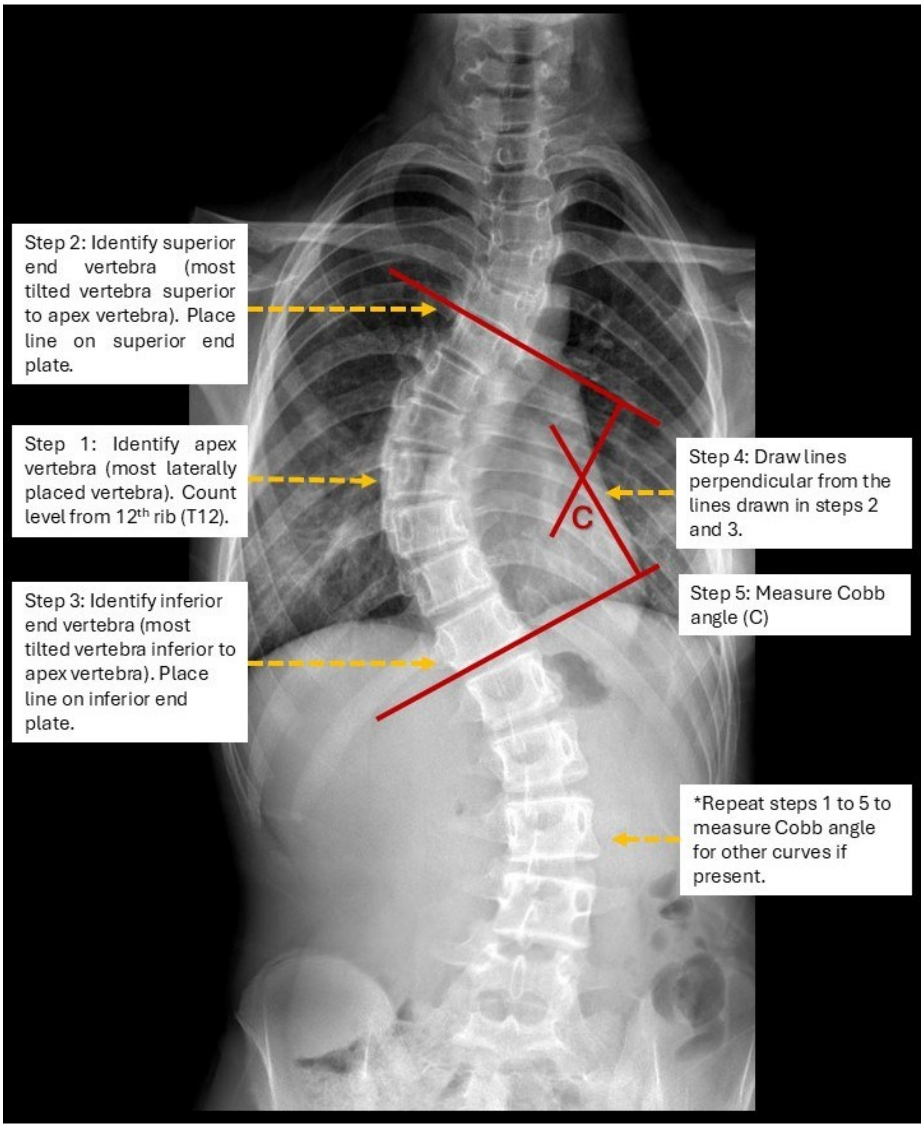

Figure: Measuring Cobb angle following the 5 steps as shown.