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Case report

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Scoliosis in an identical twin: a case report with literature review

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Abstract

Scoliosis is called the lateral angulation of the vertebra more than 10 degrees. Scoliosis is divided into two groups that is congenital and idiopathic scoliosis. Congenital scoliosis was presented in one of the two 14-year-old sisters who were identical twins; while the other developed adolescent idiopathic scoliosis. The sister with congenital scoliosis has hemivertebra in the thoracic third and seventh vertebras, block vertebra in the thoracic 10^{th}-11^{th} vertebras, and fusion the L3-L4 vertebras. The Cobb angle of this sister was 44.4 degrees of thoracic scoliosis. In the other sister, 13.8 degrees lumbar scoliosis was detected. Scoliosis has been shown to occur not only with the genetic background but also due to low birth weight.

Introduction

Scoliosis is a 3-dimensional deformity that occurs when the lateral angulation of the vertebra is more than 10 degrees [1]. Scoliosis is divided into two different groups. These are Congenital Scoliosis (CS) and Idiopathic scoliosis (IS) [2]. Congenital scoliosis occurs at a rate of 0.5-1 per 1000 live births [3]. The role of genetic factors and environmental factors in its etiology has not been fully explained yet. Studies suggested that the spine is challenged due to developmental disorders in the fifth and eighth weeks of pregnancy [4]. These studies emphasized the developmental defects of the spine may occur as a result of segmentation and formation anomalies or a combination of both [5]. Also, vertebral deformities seen in congenital scoliosis have been associated with genetic syndromes such as Alagille syndrome [6], spondylocostal dysostosis [7] and Jarcho-Levin syndrome [8]. IS is seen in society between 0.2% and 3% [9]. In different studies, it has been argued that IS has a genetic background [10]. It has been reported that an anomaly in the gene 17p11 may be present in genetic studies on large families with autosomal dominant transmission [11]. In this article, we aimed to contribute to the literature by publishing identical twins with congenital scoliosis in one sibling and adolescent idiopathic scoliosis in the other sibling.

Patient and observation

The two sisters were born prematurely in the cesarean method in 2006. When the medical records of the patients were examined, it was reported that a single sac was seen in the Ultrasonography (USG) performed during the mother's pregnancy. Birth weights were 1800g (twin 1) and 2300g (twin 2), respectively. Twin 1 was followed up on an incubator for one month due to low birth weight. When the mother's anamnesis was questioned, it was learned that there was no toxin exposure during pregnancy, and she did not use drugs or cigarettes. There was no history of scoliosis in other family members. Back pain of twin 1 had been going on for many years. Two years ago, the family noticed the back curvature of twin 1 and applied to the department of orthopedics and traumatology of our hospital. Both siblings were evaluated together (Figure 1). Adams forward test was performed for both siblings, and it was thought that there was scoliosis deformity in twin 1 (Figure 2). Scoliosis X-ray was taken for both siblings. When we evaluated the X-rays, left thoracic curvature in twin 1 (Figure 3) and lumbar scoliosis in twin 2 were detected. Vertebral computed tomography imaging was performed in twin 1 for further evaluation. On Computed tomography (CT) images, hemivertebra in thoracic third and seventh vertebras, block vertebra in the thoracic 10^{th}-11^{th} vertebras, and fusion in the L3-L4 vertebras were detected. The Cobb angle of twin 1 was 44.4 degrees of thoracic scoliosis (Figure 4). In twin 2, 13.8 degrees of lumbar scoliosis was detected (Figure 5). No surgical intervention has been performed on both patients so far and the patient is under close follow-up at routine intervals. These patients and their parents had gone through an informed consent procedure.

Informed consent: all procedures performed in this study were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Informed consent was received from the patients and their parents.

Discussion

The etiological factors causing the development of congenital scoliosis have not been fully explained yet. Some researchers suggested that several environmental factors may be responsible for the development of congenital scoliosis. Exposure of the mother to carbon monoxide during pregnancy [12], consuming alcohol [2], or using antiepileptic drugs such as valproic acid [2,13] and dilantin [2] are accused. Other factors are hyperthermia [2,14] and gestational diabetes mellitus [2,15]. Studies in animals have shown that

exposure to teratogenic substances such as boric acid during pregnancy can cause congenital scoliosis [12].

Researches on congenital scoliosis have been carried out in many medical departments. Some of these studies have been done in the field of genetics. In these studies, the presence of family history has been demonstrated in patients with congenital scoliosis. This genetic transition is both autosomal recessive [16] and autosomal dominant [17]. Chromosomal analysis showed that deletions in the 2p13-13, 6q13, and 15q12 regions cause congenital scoliosis [18]. Two sisters, our patients, were genetically examined for known skeletal anomalies, but no defects were found.

The first study published on identical twins with congenital scoliosis was published by Haffner [19] in 1936. In this case report, two sisters in their 20s who were identical twins were described. While one of the sisters had a left hemivertebra under the third lumbar vertebra, the other sibling had a left hemivertebra under the second lumbar vertebra. Both siblings were reported to have a slight left lumbar curvature.

In the case report published by Peterson HA and Peterson LF [20] in 1967, identical twins were mentioned. In this study, multiple hemivertebra and scoliosis were detected in one of the twin brothers at the age of 14, while the other brother was reported to have a normal spine.

In their study published in 2008 by Kaspiris *et al.* [12], they talked about two brothers with an identical twin who had congenital scoliosis. They reported that 34 degrees left thoracic curvature was detected in the first brother and 10 degrees left thoracic scoliosis was revealed in the other.

In the case report made by Cho *et al.* [3] in 2018, they mentioned patients with three different identical twins. They detected scoliosis due to hemivertebra in both of the first twins. In the second twin siblings, there was thoracic scoliosis developed due to the hemivertebra level seen at the T10-T11 level. In contrast, no curvature was observed in the scoliosis graph of the other sibling. One of the third twins had congenital scoliosis due to left T9 hemivertebra and right L4 hemivertebra and they found that the other sibling was healthy.

Limitations: this case study represents one pair of identical twins.

Conclusion

In this case report, we demonstrated the development of congenital scoliosis due to multiple vertebral abnormalities in one sibling of the identical twins and the presence of adolescent idiopathic scoliosis in the other. In the literature review, when congenital scoliosis was detected in one of the identical twins, it was observed that the other sibling had scoliosis mostly. This situation could often be explained by genetic background. In our case, congenital scoliosis in the thoracic region due to multiple vertebral anomalies was found in the low birth weight twin. At the same time, the other sibling had adolescent idiopathic scoliosis developing in the lumbar region. We concluded that twin 1's multiple vertebral anomalies might result from low birth weight.

Competing interests

Both authors declare no competing interests.

Authors' contributions

Both authors have contributed to this manuscript. They have read and agreed to the final manuscript.

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We thank our patients for accepting to participate in this case report study.

Figures

Figure 1: twin 1 and twin 2 were evaluated together

Figure 2: Adams Forward Test

Figure 3: anteroposterior (A) and lateral (B) x-ray images of twin 1

Figure 4: Cobb angle of twin 1 was 44.4 degrees of thoracic scoliosis

Figure 5: Cobb angle of twin 2 was 13.8 degrees of lumbar scoliosis

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Figure 1: twin 1 and twin 2 were evaluated together

Figure 2: Adams Forward Test

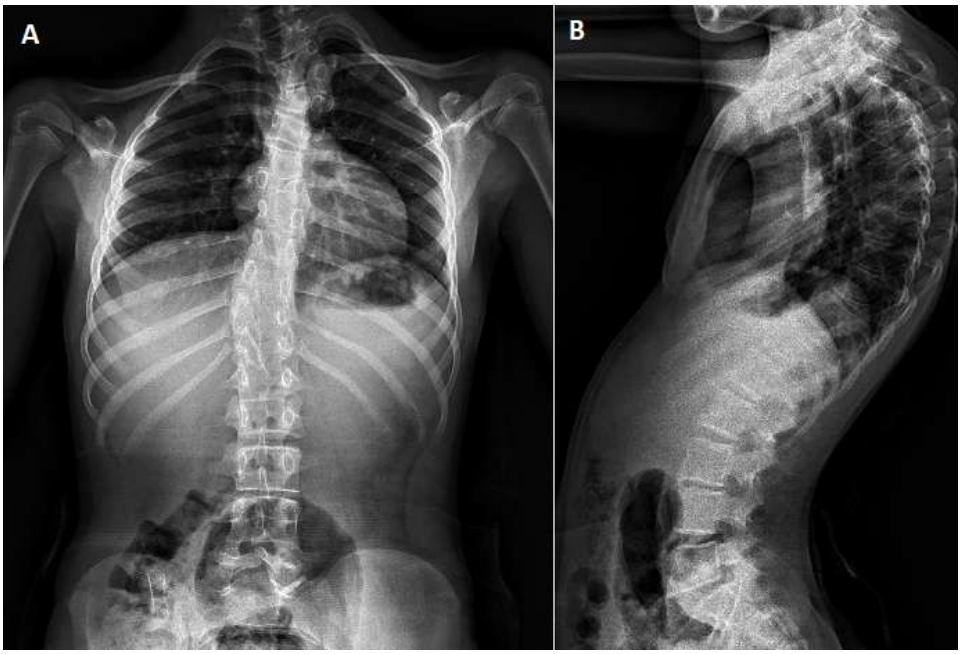

Figure 3: anteroposterior (A) and lateral (B) x-ray images of twin 1

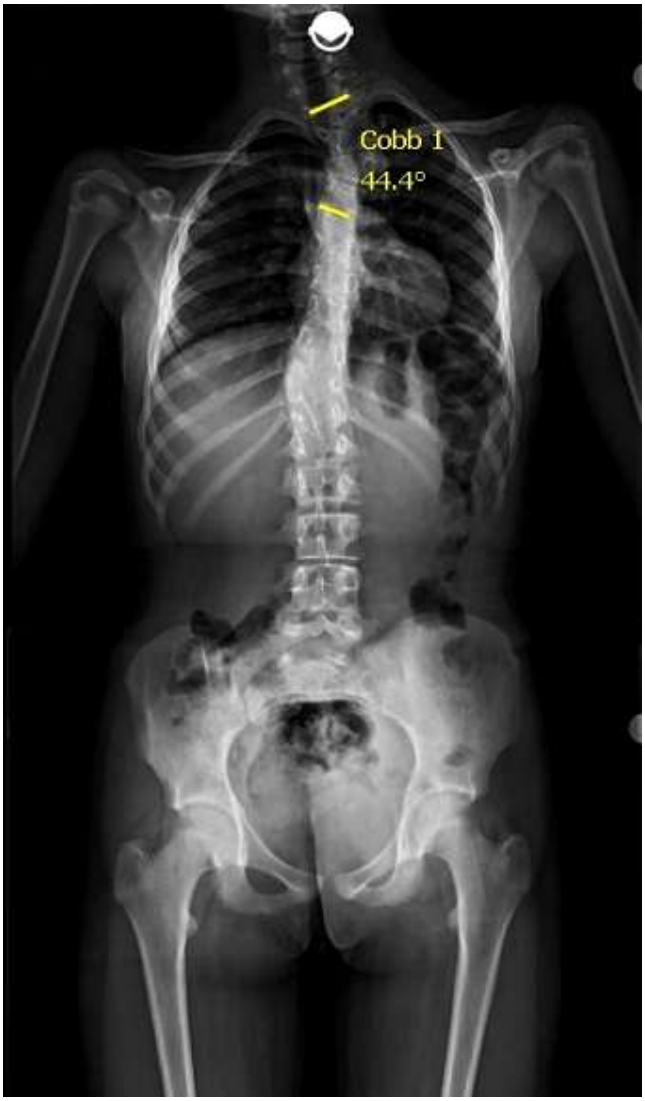

Figure 4: Cobb angle of twin 1 was 44.4 degrees of thoracic scoliosis

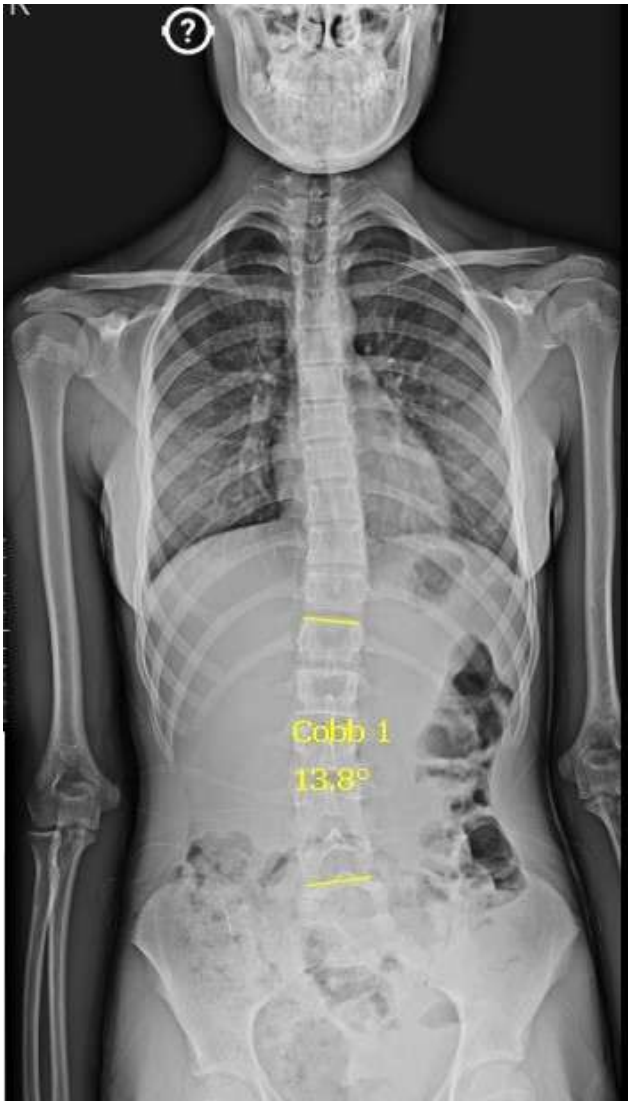

Figure 5: Cobb angle of twin 2 was 13.8 degrees of lumbar scoliosis