Clinical Case Report

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Revision surgery for curve progression after implant removal following posterior fusion only at a young age in the treatment of congenital scoliosis

A case report

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Abstract

Rationale: Congenital scoliosis due to a hemivertebra creates a wedge-shaped deformity, which progresses and causes severe spinal deformities as an individual grows. The treatment of congenital scoliosis focuses on early diagnosis and appropriate surgical management before the development of severe deformity.

Patient concerns: We report the case of a 4-year-old male child with a left thoracolumbar scoliosis of 27° (T10-T12) due to a T11 hemivertebra who was treated by posterior fusion and pedicle screw fixation at the age of 4 years. The implant was removed due to pain secondary to implant prominence after 4 years without definitive revision surgery, which led to significant progression of the scoliosis, to 50°. The indication for posterior vertebral column resection (PVCR) is a congenital spinal deformity with a curve magnitude greater than 30° with fast progression. This includes documented progression of the curve by more than 5° in a 6- month period, failure of conservative treatment, or both.

Outcomes: The patient underwent PVCR of the T11 hemivertebra. Nine years after the revision surgery with PVCR, the patient showed satisfactory results and his spine was well balanced.

Lessons: This case shows that removal of an implant that was not the only cause of curve progression at a young age may lead to progression of scoliosis and, therefore, should be avoided unless it is absolutely necessary.

Conclusion: Congenital scoliosis due to a hemivertebra at a young age could be treated by hemivertebra resection or anterior and posterior epiphysiodesis as definitive surgical treatment. The patient was eventually treated with PVCR, which achieved satisfactory correction without curve progression in a long-term follow-up.

Abbreviations: PVCR = posterior vertebral column resection, TLSO = thoraco-lumbo-sacral orthosis.

Keywords: congenital scoliosis, hemivertebra, implant removal, posterior fusion, posterior vertebral column resection

1. Introduction

Congenital scoliosis results from the abnormal formation or segmentation of vertebral elements that leads to asymmetric growth of the spine.[1] A complete unilateral failure of formation creates a hemivertebra, which may cause a wedge-shaped deformities during further growth.[2–6]

Even though many cases of hemivertebra do not require early surgical treatment, instrumented posterior spinal fusion is one of the surgical treatments for congenital scoliosis at a young age.

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D-GC and J-HY contributed equally to this study.

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However, classical early surgical treatment of a hemivertebra at a young age may include: complete resection of the hemivertebra with fusion, or anterior and posterior epiphysiodesis, if surgically indicated.[7–16]

Determining the appropriate timing and type of surgery is important but difficult, because natural evolution of a hemivertebra does not always lead to severe deformities and worsening of spinal deformities can occur during the pubertal growth spurt, usually in the form of a curve that is more severe than the local malformation.

Posterior vertebral column resection (PVCR) in pediatrics has been reported as the curative treatment of congenital scoliosis with successful results.[7,8] Herein, we report a case with long-term follow-up of PVCR for curve progression after implant removal following instrumented posterior spinal fusion in congenital scoliosis at a young age.

2. Case report

This is a case report of a male child who was born by vaginal delivery in 1992, following a full-term pregnancy. The patient's mother noticed that he had an asymmetry in his back at the age of 2 years, and was referred to our hospital by a private physician after a radiograph showed congenital anomalies in the thoracolumbar spine.

On clinical examination, the patient was noted to have left thoracolumbar scoliosis with segmental kyphosis but no complain of any subjective symptoms including pain. The results of physical and neurological examinations were otherwise within normal limits. Associated malformations were ruled out a clinical work-up. Thorough physical examination including investigation of foot or leg asymmetry, craniofacial malformation, and cardiac and urinary malformations was performed, with unremarkable findings. At the first visit to our hospital, the patient was fitted with a thoraco-lumbo-sacral orthosis (TLSO). At the 6-month follow-up visit, the curve had progressed more than 5°.

The patient had left thoracolumbar scoliosis of 27° (T10–T12) due to a T11 hemivertebra (Fig. 1A). Because of the known progression of this vertebral anomaly, surgery was recommended. At the age of 4 years, the patient underwent posterior fusion with pedicle screw fixation to correct the deformity with convex side compression, from T10 to T12 (Fig. 1B), without hemivertebra resection or anterior and posterior epiphysiodesis. The preoperative degree of scoliosis (27°) was maintained after surgery (Fig. 2A). The patient had no subjective symptoms and did well after surgery. However, 3 years after surgery, he started to have pain secondary to implant prominence and his parents elected to have the implant removed. At the age of 8 years, he underwent implant removal without hemivertebra resection or epiphysiodesis (Fig. 2B).

He had a left thoracic curve of 31° after implant removal, and the curve and hump started to progress as he grew. At the age of 14 years, 6 years after implant removal, he had left thoracic scoliosis of 50° and a left thoracolumbar hump of 17 mm (Fig. 3 A and B), but no complaints of subjective symptoms such as pain. He underwent a PVCR at T11 and posterior fusion with pedicle screws from T8 to L1 (Fig. 3C). The scoliosis improved postoperatively and was maintained at 9 years post-PVCR (Fig. 4A and B).

Two weeks after PVCR, the patient was mobilized with a localizer cast that he wore for 3 months; after that, he used a TLSO for 6 months.

3. Discussion

A hemivertebra should be treated at the earliest age possible, before the deformity increases and structural differentiation takes place in adjacent spinal segments, if surgery is indicated.[3,10,11] Surgical procedures may include: in situ fusion, anterior and posterior fusion with or without instrumentation, combined anterior and posterior convex hemiepiphysiodesis or hemiarthrodesis, hemivertebra excision and fusion, or PVCR.[12–16] The best permanent corrective surgery may be removal of the hemivertebra. Recently, posterior hemivertebra resection or PVCR in growing children has been reported with satisfactory results.[7–9,14–16]

This case illustrates what not to do in the treatment of congenital scoliosis, which is what caused significant curve

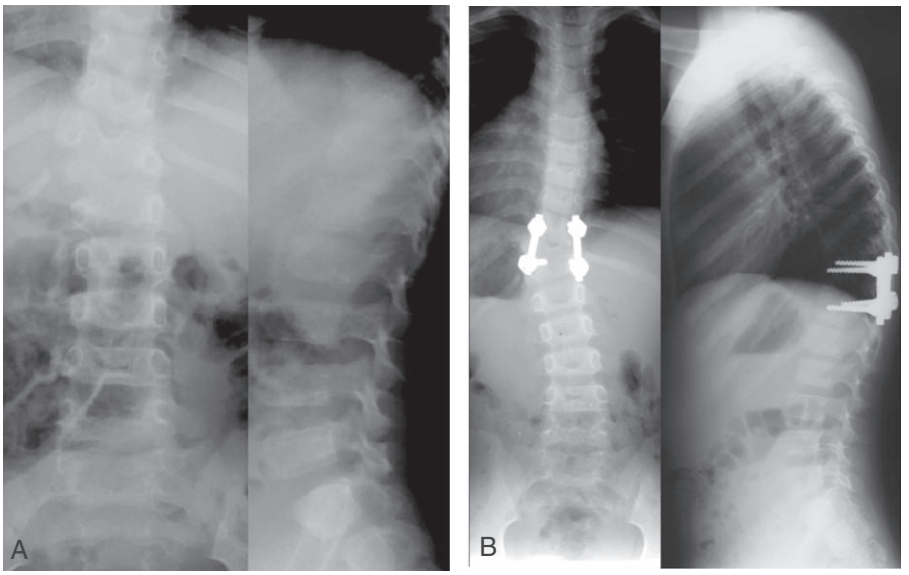

Figure 1. (A) Radiographs taken before the initial surgery showing congenital hemivertebra at T11 with 27° left thoraco-lumbar scoliosis. (B) Radiographs taken 1 year after the initial surgery showing that the main curve of scoliosis was 29°.

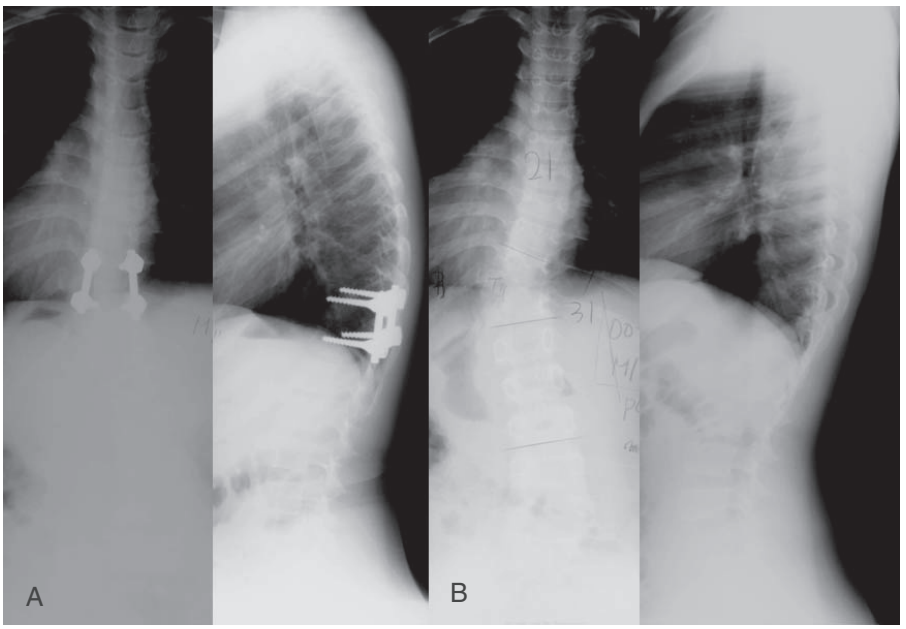

Figure 2. (A) Radiographs taken before implant removal showing that the main scoliotic curve was maintained at 28° at a 4-year follow-up. (B) Six-month follow-up radiographs after implant removal showing that the main scoliotic curve was maintained at 31° without curve progression.

progression. Preoperative computed tomography clearly showed that a fully segmented hemivertebra was causing the patient's condition. There was also curve progression greater than 5° during the 6-month follow-up at a young age, and we decided to conduct an operation even though the patient had a relatively small Cobb angle of 27°, which might be considered excessively aggressive treatment. While growth arrest or hemivertebrectomy would be operative options in this circumstance, these procedures

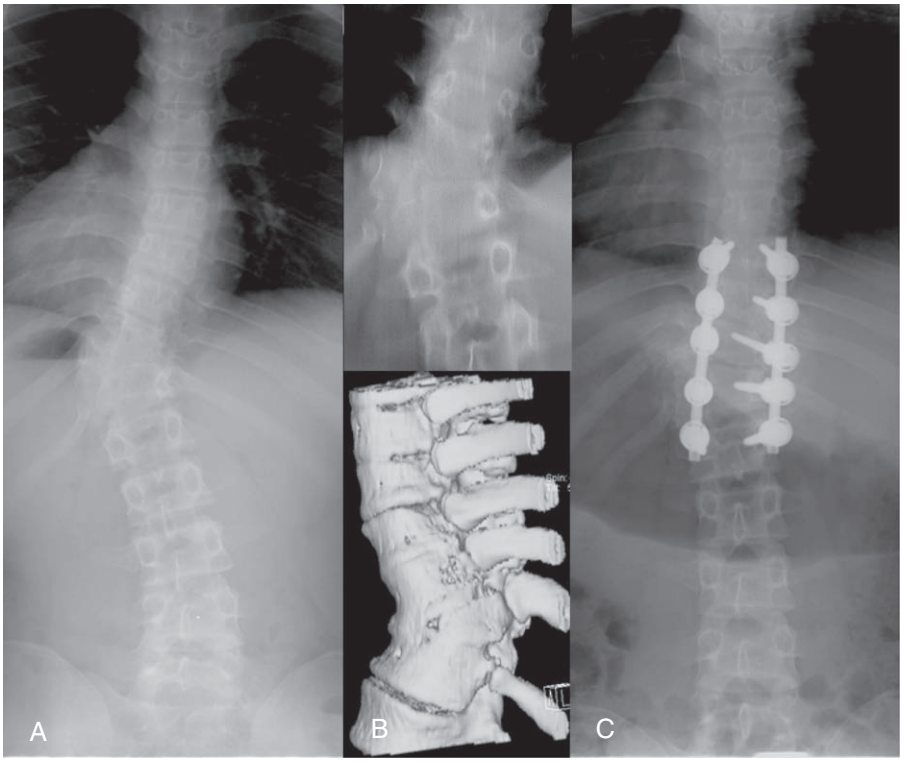

Figure 3. (A) Four-year follow-up radiographs after implant removal showing that the main scoliotic curve had deteriorated to 50° and the segmental angle of kyphosis was 23°. (B) Four-year follow-up 3-dimensional computed tomography (CT) after implant removal. (C) Immediate postoperative radiography after revision surgery showing that the main scoliotic curve had improved to 21° with segmental kyphosis of −3°.

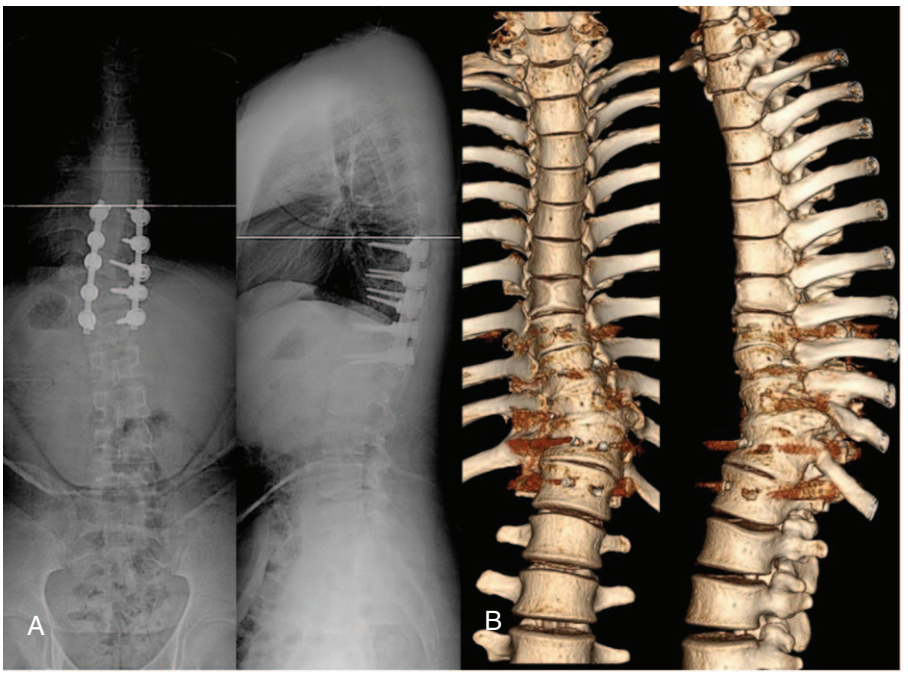

Figure 4. (A) Follow-up radiographs taken 9 years after revision surgery showing that the main scoliotic curve was well maintained at 23° with a segmental angle of kyphosis of −1°. (B) Nine-year follow-up 3D reconstruction CT scan after revision surgery.

were not done on 4-year-old children in 1996. In other words, the first operation should have been a hemivertebral resection or at least an anterior–posterior epiphysiodesis, not simply a posterior segmental fusion, because remnant hemivertebral segments can grow and lead to increasing deformity. Indications for surgery must be determined mainly on the basis of trunk imbalance from a large curve induced by the vertebral malformation, not only on the Cobb angle of the short curve around the hemivertebra. Spinal deformities can worsen during pubertal growth, usually taking the form of a wider scoliosis curve than the local malformative curve. It was expected that initial surgery consisting of only posterior fusion was appropriate for a patient with a Cobb angle of 27° and a spine that seemed relatively well balanced. However, optimal initial surgery would have achieved arthrodesis and bilateral posterior osteosynthesis with initial resection of the hemivertebra or correction of the deformity.

At the age of 8 years, 4 years after the initial surgery, the implant was removed because of pain caused by metal prominence. Implant removal in individuals who are still growing should be avoided unless it is absolutely necessary.[17–19] The spinal implant needs to be removed in patients who develop pain, metal irritation, or infection. Disadvantages and/or complications due to implant removal are rarely reported, especially in young children.[19] If removal of the implant, which was not the only cause of curve progression, had not been performed, it was possible that there would have not been deterioration of deformity in our patient. At the time of removal of the posterior instrumentation at the age of 8 years, a hemivertebrectomy or PVCR should have been simultaneously considered to achieve a sagittal and coronal balance of the spine. However, that extensive surgery was not carried out in the 8-year old child as there was a high risk of complications such as excessive blood loss and neurologic complications.

For 6 years after implant removal, deformity progression occurred; the condition deteriorated as the patient passed through his adolescent growth spurt. The patient was treated eventually with PVCR, which achieved satisfactory correction without curve progression in a total 19-year follow-up. In addition, cosmetic concern such as kyphoscoliosis-related deformities plays an important role in a patient's mental health and satisfaction with surgical outcomes.

There is a limitation to our study: most cases differ greatly even though they may present with similar symptoms, so we are unable to draw definite conclusions from a single case report. However, there are very few previously published case reports on long-term follow-up after PVCR for curve progression after implant removal following instrumented posterior fusion in congenital scoliosis at a young age, so this report is valuable.

In summary, our case showed that removal of an implant which was not the only cause of curve progression at a young age may lead to curve progression and, therefore, should be avoided unless it is absolutely necessary. Congenital scoliosis due to hemivertebra at a young age could be treated by hemivertebra resection or anterior and posterior epiphysiodesis as definitive surgical treatment. Our patient was treated with PVCR, which achieved satisfactory correction without curve progression in a long-term follow-up.

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