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CASE REPORT

# Unusual presentation of vertebral endplate Modic changes in congenital scoliosis associated with pain: a report of two cases

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**Abstract** Congenital scoliosis is not normally associated with pain in young children. We are presenting two cases of young patients with congenital scoliosis and moderate to severe pain. There were no spinal cord abnormalities found in these patients. The magnetic resonance imaging and CT scan revealed disc degeneration with Modic changes at the apex of the congenital scoliosis. We hypothesized that the mechanical instability resulting from poor spinal element formations associated with congenital scoliosis was responsible for causing the disc degeneration and endplate changes. Modic changes have been reported to be associated with pain in degenerative conditions of the spine. Both patients underwent a posterior spinal fusion and instrumentation, which relieved their pain immediately.

**Keywords** Congenital scoliosis · Modic changes · Disc degeneration

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## Case presentation

The first case was a 10-year-old girl presenting with scoliosis and back pain. Her family history was negative for scoliosis. She described daily pain in the upper thoracic area for several months, and found that this pain was related to her activities. She also had night pain but no numbness or weakness in her bilateral lower extremities. The physical examination showed a symmetrical waist, no pelvic obliquity and no midline skin abnormalities. She had hump on right upper thoracic spine while performing Adam forward bending test. She had tenderness with palpation at the upper thoracic spine. Her neurological examination was normal. Radiographic evaluation (Fig. 1) revealed a right thoracic scoliosis of 30° with a hemivertebra at T7 on the right side. Lateral plain radiographs revealed a thoracic kyphosis of 39° and a lumbar lordosis of 46°. However, the remainder of the spine appeared normal, both proximally and distally to the involved area. A left-side bending film was used to evaluate more details at the level of the hemivertebra (Fig. 1c). Computed tomography imaging of the spine was performed, showing fully segmented hemivertebra at T7 with disc space narrowing and sclerosis at the lower endplate of left T6 and upper endplate of left T8 without endplate destruction. The sclerotic changes were located only on the concave side in an asymmetrical distribution (Fig. 2). Magnetic resonance imaging (MRI) of the entire spine was performed as a screening test for neural axis abnormalities, which showed no spinal cord abnormalities. Sagittal views of MRI showed a hypointense signal on T1-weighted, an intermediate signal on T2-weighted at both the lower endplate of left T6 and the upper endplate of left T8 (Fig. 3), which represented Modic changes type 3 [1]. The second case was an 8-year-old

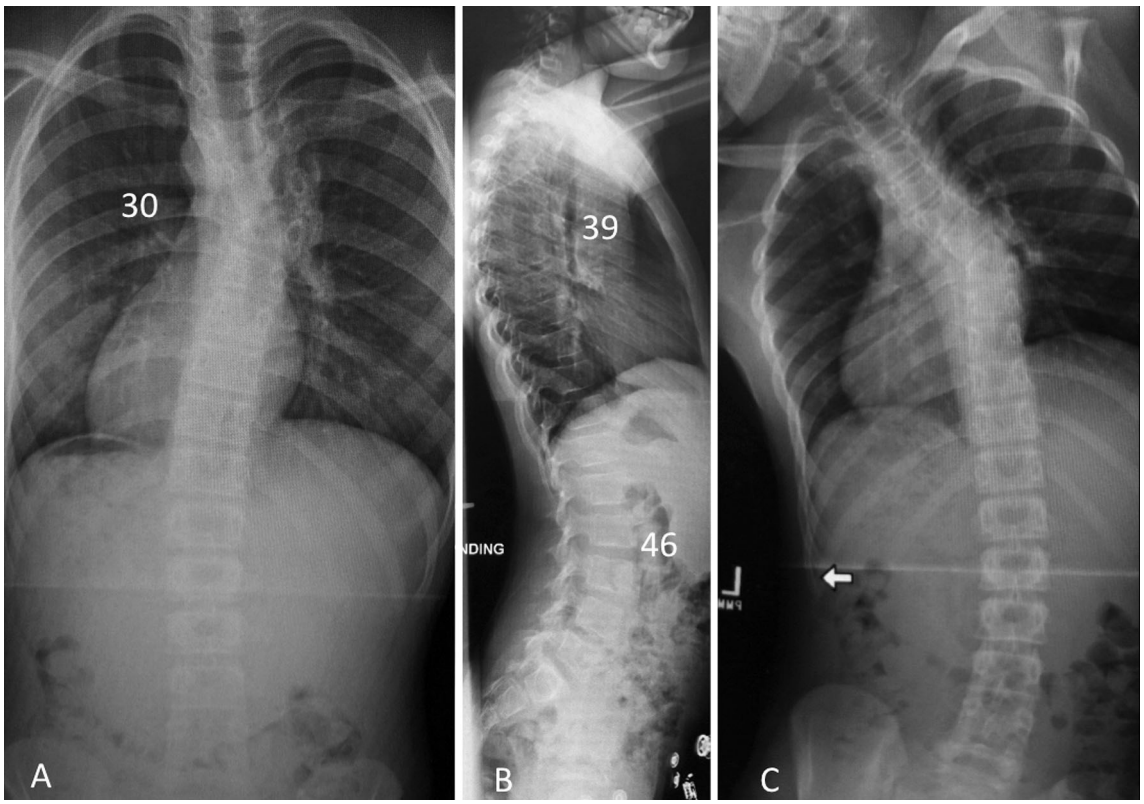


**Fig. 1** Standing AP and lateral radiographs **a**, **b** showed right thoracic scoliosis of 30° due to hemivertebrae at T7 level on the *right side* with normal thoracic kyphosis and lumbar lordosis. *Left-side* bending radiograph **c** showed hemivertebrae at right T7 level

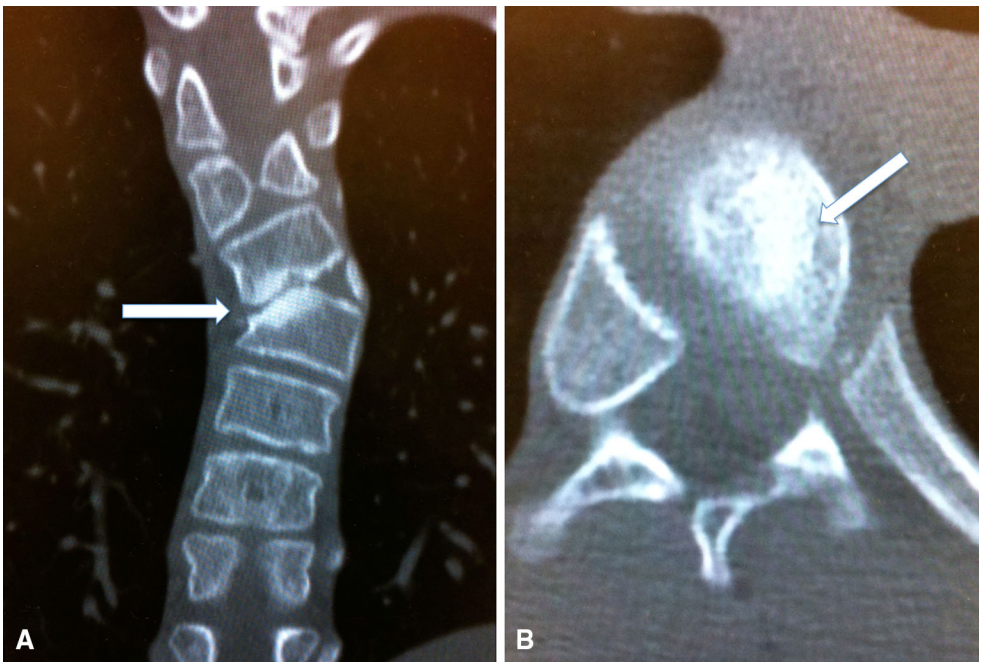


**Fig. 2** Computed tomography of entire spine in coronal and axial views showed fully segmented hemivertebrae at T7 with disc space narrowing and sclerosis (*arrow*) at left lower endplate of T6 and left upper endplate of T8 without endplate destruction. The sclerosis changes were located only on the concave side in asymmetrical distribution

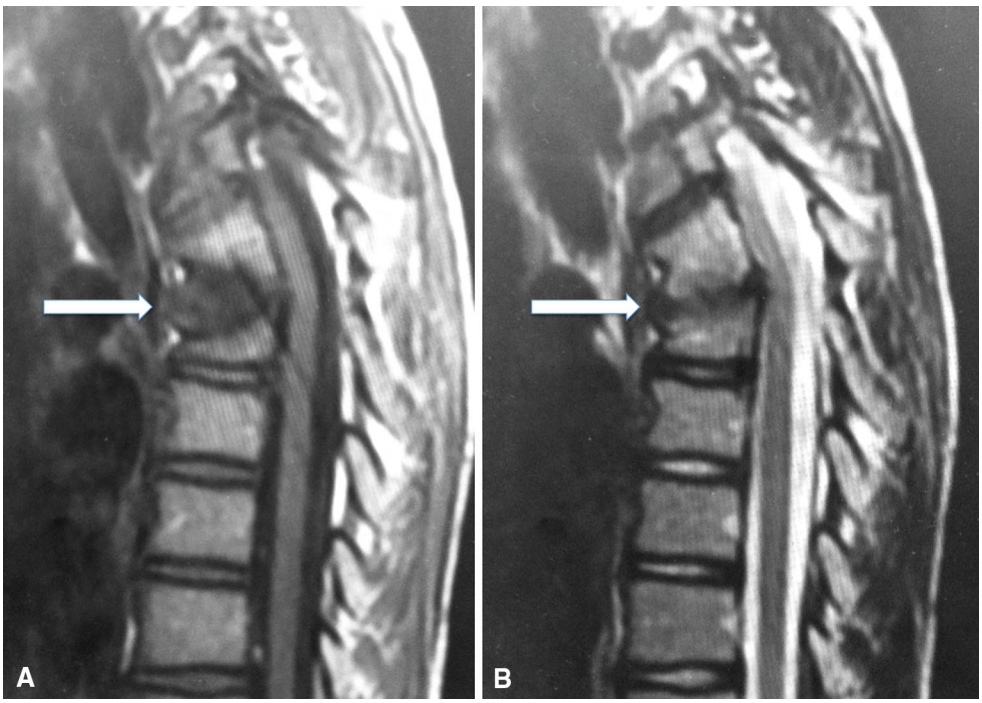


**Fig. 3** On sagittal imaging MRI showed hypointense signal on T1W (**a**), an intermediate signal on T2W (**b**) at the lower endplate of left T6 and upper endplate of left T8 (*arrow*)

girl who also presented in clinic with scoliosis and back pain. She had pain in upper thoracic area during the day and night for 1 year. The pain was also increased with greater physical activity. The physical examination showed a hump on right upper thoracic spine while performing the Adam forward bending test. She had marked tenderness with palpation of the thoracic spine. Her neurological examination did not demonstrate any focal deficits. Radiographic evaluation (Fig. 4a) revealed a right thoracic scoliosis of 48° due to a hemivertebra at T11 on the right side. Computed tomography imaging of the spine was performed which showed fully segmented hemivertebra at T11 with disc space narrowing and sclerosis of the lower endplate of left T10 and upper endplate of left T12 without endplate destruction. The sclerotic changes were located only on the concave side in an asymmetrical location (Fig. 4b). MRI of the spine showed characteristic of type 3 Modic endplate changes similar to the first case. In both cases, a hemivertebrectomy was performed with posterior approach. The hemivertebrae and degenerative discs were removed (Fig. 5). After the operation, the clinical back pain at the thoracic spine resolved. The first patient had more than a year follow-up and the second patient had more than a 4-year follow-up with continued resolution of the pain and return to full physical activities.

## Discussion

Modic changes are signal changes of the vertebral endplate on MRI [1–3]. Changes of the vertebral endplate were first described by Modic et al. and were classified into 3 types [1]. Modic type 1 changes are hypointense signal in T1-weighted sequences and hyperintense signal in T2-weighted sequences. Modic type 1 is associated with fissuring of the cartilaginous endplate and increased vascularity within the subchondral bone on pathological examination which corresponds to the endplate edema. Modic type 2 changes are hyperintense signals in T1-weighted sequences and are either hyperintense or intermediate in T2-weighted sequences that reflect fatty replacement of the adjacent bone marrow. Modic type 3 changes are hypointense signals in T1-weighted sequences and hypointense signals in T2-weighted sequences that are represented in vertebral bodies with sclerotic changes. Modic changes are common in adult spinal degenerative diseases. Most studies have investigated the relationship on Modic changes in chronic low back pain at the degenerative lumbar spinal area. However, no investigation has been reported on the Modic changes in congenital scoliosis at the thoracic area. We report the Modic changes type 3 in 2 girls with congenital scoliosis of the thoracic spine. The Modic changes in our study have asymmetrical distribution that occur only on the concave side of the curve

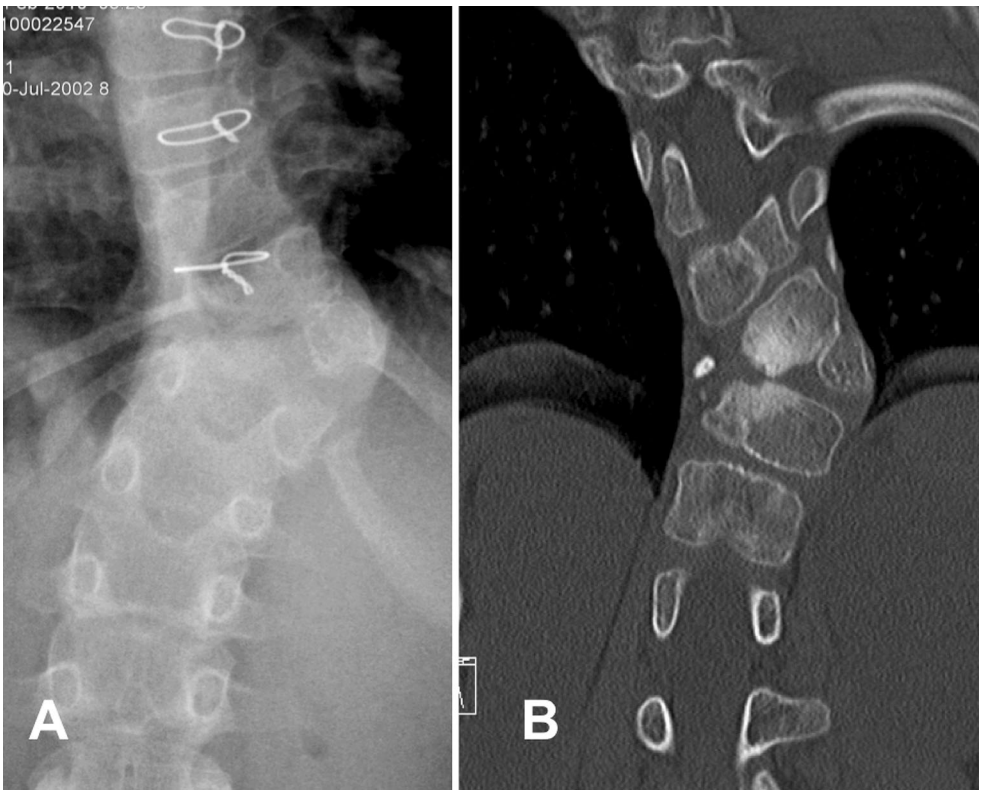


**Fig. 4** Standing AP film (**a**) showed hemivertebrae at right T11, coronal computed tomography (**b**) showed sclerosis at left lower endplate of T10 and left upper endplate of T12 (same characteristic of the first case)

at the apex vertebrae which is characteristically similar to the Modic changes of adult degenerative scoliosis [4]. This characteristic can be explained by the asymmetrical biomechanical loading at the endplate resulted from scoliosis and different type of forces on the endplate (compression force on concave side and tension force on convex side). Pain is typically neither the primary complaint for the patient with congenital scoliosis who seeks treatment, nor the priority for surgeons, when considering operative management because of its rare presentation. In contrast to adult scoliosis, congenital scoliosis is not normally associated with pain [5]. But in our case report we report two cases with relatively minor congenital scoliosis and back pain. Pain in a scoliosis patient can be the result of many pathologies such as disc degeneration, facet arthrosis, muscular imbalance, occult syrinx, spinal cord tumor and neuromuscular disorder [6, 7]. Many studies have also demonstrated that Modic changes are commonly associated with low back pain [1, 3, 8–10]. We hypothesized that the mechanical instability due to the poor spinal element formation leads to increased stress in the disc space leading to degeneration and Modic changes of the endplate. The Modic changes from instability are most likely the cause of pain in these patients with congenital scoliosis. According to the Modic study, the changing signal intensity in MRI reflected a causal process, which was an imbalance of biomechanical stress or intervertebral instability process [8]. Karchevsdy et al. suggested that Modic changes represented a response to degenerative process of vertebral endplate involving the disc degeneration [9]. An increase in the intervertebral shear forces from instability from the congenital scoliosis introduce possibility of endplate trauma and promoted the intervertebral disc and vertebral endplate degeneration [4]. Therefore, we considered instability resulting in endplate degeneration as the cause of pain in these congenital scoliosis patients.

We believe the changes in the endplates are from instability due to lack of proper formation of the spinal structural elements rather than other etiologies such as infection. In one of the cases the patient had cardiac surgery, but never had any indication of infection at the time of surgery or in follow-up. The patient also did not have any constitutional symptoms such as fevers, night sweats, weight loss or severe pain that may suggest a discitis. The MRI also did not have any increase in signal in the disc space to suggest infected fluid as in discitis. We understand that if it is burned out it may not show fluid. We think discitis is unlikely given her clinical history of no infection.

The discs in congenital scoliosis can be formed abnormally. The discs may lack proper growth plates and normal internal architecture that contribute to the cause and progression of the deformity. The instability results from abnormal formation of the spinal elements including the


**Fig. 5** Postoperative film after hemivertebrectomy of T11 from posterior approach

intervertebral discs and bony elements. The instability can create the reaction of the endplates seen in both these cases, which are similar to Modic changes found in degenerative disc disease. The etiology and the start point of the discs to reach the endplate changes are different, but the appearance at the end appears similar.

The postoperative follow-up for one patient is 1 year with complete resolution of pain. The second patient had a 4-year follow-up with complete resolution of pain. The most important point of these two cases is to highlight the etiology of painful small curves in congenital scoliosis may arise from instability resulting in endplate changes that can be helped with stabilization.

## Conclusion

This is the first study that reports the asymmetrical type 3 Modic changes on the concave side of thoracic spine in young girls with pain and congenital scoliosis. The instability resulting in the Modic endplate changes appears to be the cause of pain in these young patients with congenital scoliosis. Stabilization resolved the pain in both cases.

### Compliance with ethical standards

**Conflict of interest** None of the authors has any potential conflict of interest.

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