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Case Report

Osteoid osteoma presenting as thoracic scoliosis

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Abstract

BACKGROUND CONTEXT: Osteoid osteoma of the thoracic spine is relatively uncommon and is often difficult to diagnose, especially when patients do not complain of pain.

PURPOSE: This study aims to describe an unusual case of scoliosis caused by osteoid osteoma of the thoracic spine that was challenging to diagnose.

STUDY DESIGN/SETTING: A case report of a 12-year-old girl who presented with scoliosis caused by osteoid osteoma of the thoracic spine without apparent pain was carried out.

METHODS: Diagnosis of the lesion was made using computed tomography (CT) and magnetic resonance imaging as well as the Scoliosis Research Society-22 (SRS-22) patient-based questionnaire.

RESULTS: A preoperative CT myelogram revealed a mass lesion in the lamina of the 10th thoracic vertebra that was considered to be osteoid osteoma. This diagnosis was histologically confirmed following tumor excision. The patient's spinal deformity and SRS-22 scores were both improved at 5 months postoperatively.

CONCLUSIONS: Osteoid osteoma of the thoracic spine may present as non-painful scoliosis. Tumor resection is effective. Clinicians should bear this uncommon lesion in mind during recommended CT examination before scoliosis surgery.

Keywords: Osteoid osteoma; Scoliosis; Spinal deformity; Tumor excision; Vertebral rotation; CT myelogram

Introduction

Osteoid osteoma was first described by Jaffe in 1935 [1]. It is a benign reactive bone lesion that most frequently occurs in male adolescents between 10 and 20 years of age [2,3]. The predominant cause of painful scoliosis in adolescents is osteoid osteoma of the spine [4]. Approximately 10%–25% of osteoid osteoma cases occur in the spine [5–9]: 60% of tumors are found in the lumbar spine, 27% in the cervical spine, 12% in the thoracic spine, and 2% on the sacrum. There is a very strong correlation between the neoplasm and scoliosis because two-thirds of cases manifest as painful scoliosis [10]. However, osteoid osteoma is commonly missed in a large, albeit undetermined, number of patients [11] because the lesion is often recognized only after months or years of ineffective bracing [12].

Osteoid osteoma of the thoracic spine is relatively uncommon. It is often difficult to diagnose, especially when the patient does not complain of pain. We herein describe the clinical outcome of a girl who presented with scoliosis caused by osteoid osteoma of the thoracic spine that was detected by preoperative computed tomography (CT).

Case report

A 12-year-old girl was suspected to have scoliosis in a routine school health screening in May 2013. Because her condition worsened despite the use of a brace, she visited our institution for scoliosis correction surgery in April 2014. Her height was 154 cm and her weight was 40 kg at presentation. She had pervasive developmental disorders along with an expressive language impediment, although her intelligence quotient score was average. She possessed no family history of scoliosis. She complained of no additional

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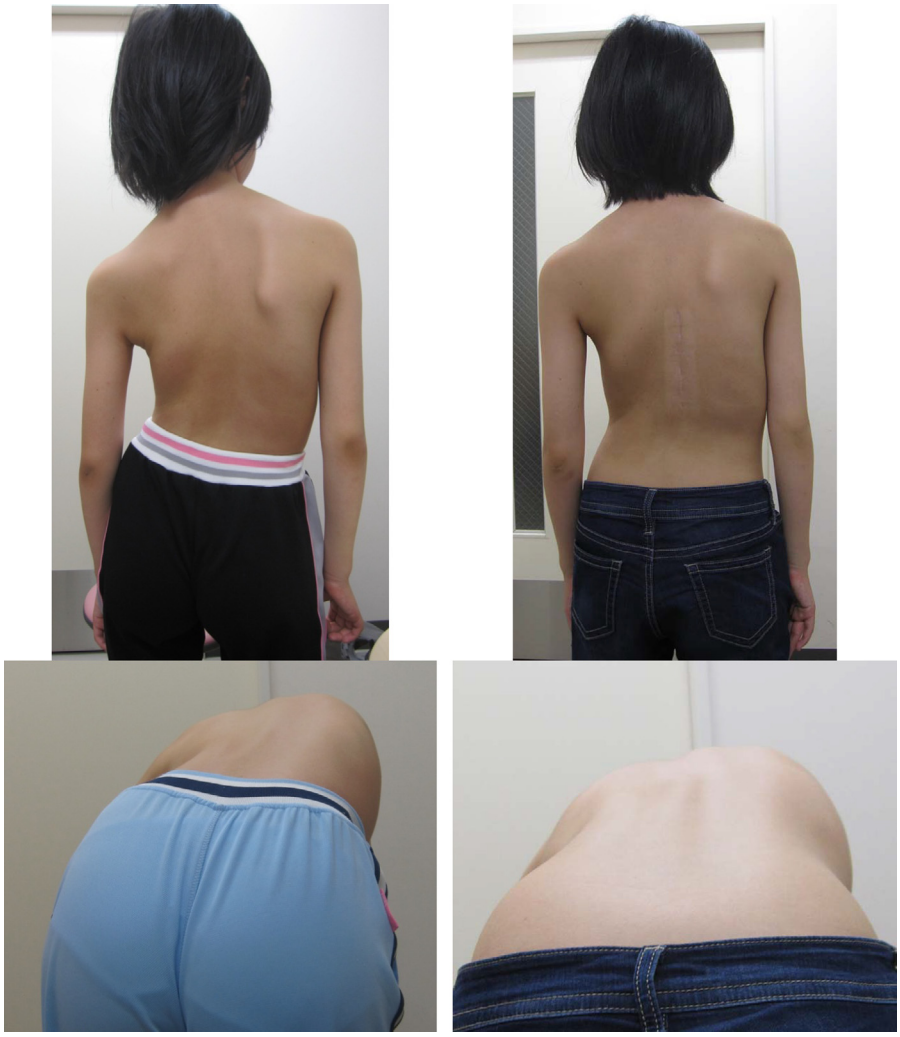

Fig. 1. (Left) Apical trunk rotation (ATR) was 19° as determined by a scoliometer. (Right) ATR had improved to 9° at 5 months postoperation.

symptoms, including back pain. Physical examination revealed right shoulder elevation. Apical trunk rotation, as determined by a scoliometer, was 19° (Fig. 1, Left). We observed no neurologic deficits. Whole spine radiographs revealed a Lenke-type 1AN curve with a Cobb angle of 45° from T5 to L2 (main thoracic) and 28° from L2 to L5 (lumbar) (Fig. 2, Left). Risser grade was 2.

Magnetic resonance imaging of the whole spine did not reveal any evidence of Arnold-Chiari malformation, syringomyelia, or dural ectasia (Fig. 3). As we had planned to perform posterior correction surgery for the patient's scoliosis, a CT examination was performed after myelography, which demonstrated a mass lesion in the lamina of the 10th thoracic vertebra with a visible nidus (Fig. 4). A re-examination of the MRI results verified this finding. The patient's Scoliosis Research Society-22 (SRS-22) patient questionnaire scores for function, pain, self-image, mental health, and subtotal were 4.5, 5.0, 2.4, 4.0, and 3.9, respectively. This enabled a revised diagnosis of osteoid osteoma, and the lesion was surgically excised. Histologic tissue findings confirmed our diagnosis (Fig. 5).

Postoperative brace therapy improved the patient's spinal deformity. Her shoulder imbalance was diminished and her apical trunk rotation had improved to 9° at 5 months after surgery (Fig. 1, Right). At 5 months postoperatively, Cobb angle from T5 to L2 was ameliorated at 30° (Fig. 2, Right) and SRS-22 scores for function, pain, self image, mental health, subtotal, satisfaction, and total had improved to 4.8, 5.0, 3.4, 4.6, 4.5, 4.0, and 4.4, respectively.

The patient and her family have provided consent for the data in this case to be published.

Discussion

Osteoid osteoma is a benign osteoblastic neoplasm that is characterized by a well-demarcated core of <1 cm and a distinctive surrounding zone of reactive bone formation [13]. Although the lesions may occur anywhere in the cortex or

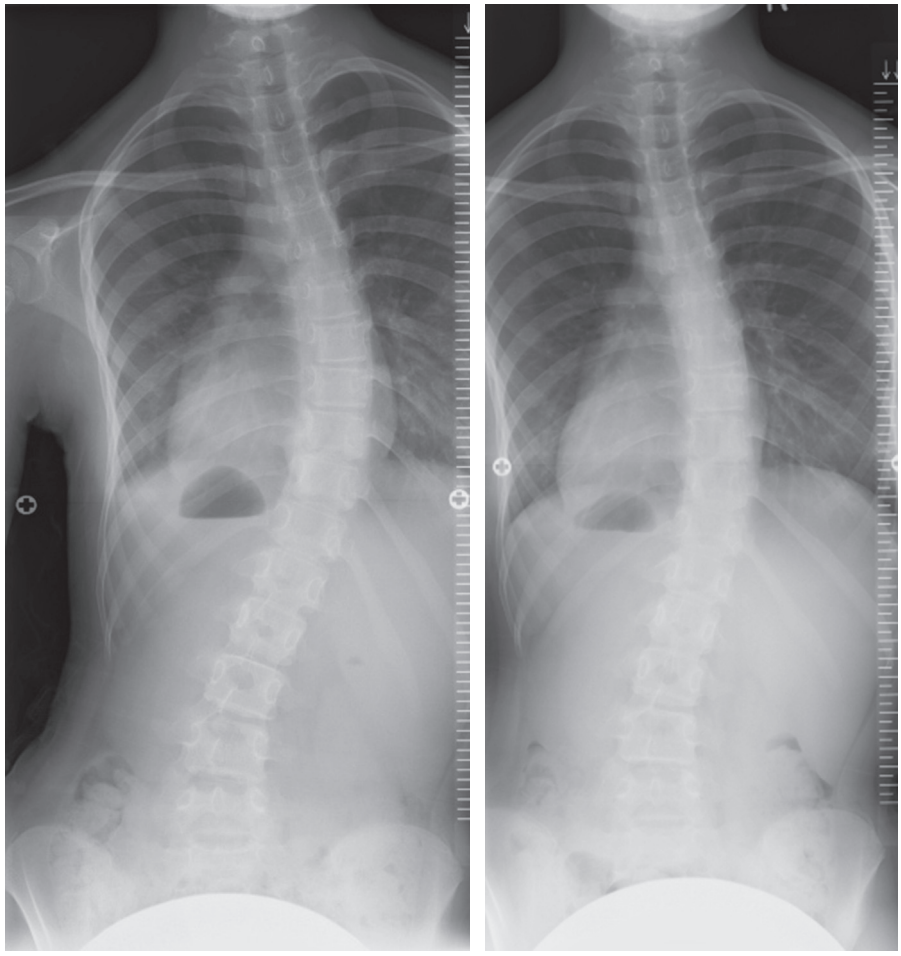

Fig. 2. (Left) Whole spine radiographs showed a Lenke type 1AN curve with a Cobb angle of 45°. (Right) Cobb angle had improved to 30° at 5 months postoperation.

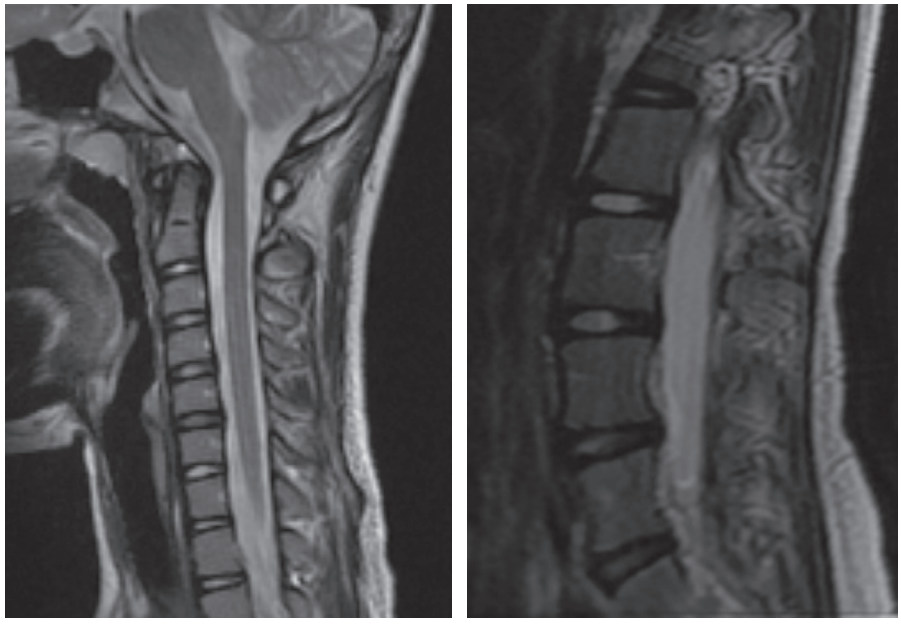

Fig. 3. MRI of the whole spine showed no signs of Arnold-Chiari malformation, syringomyelia, or dural ectasia.

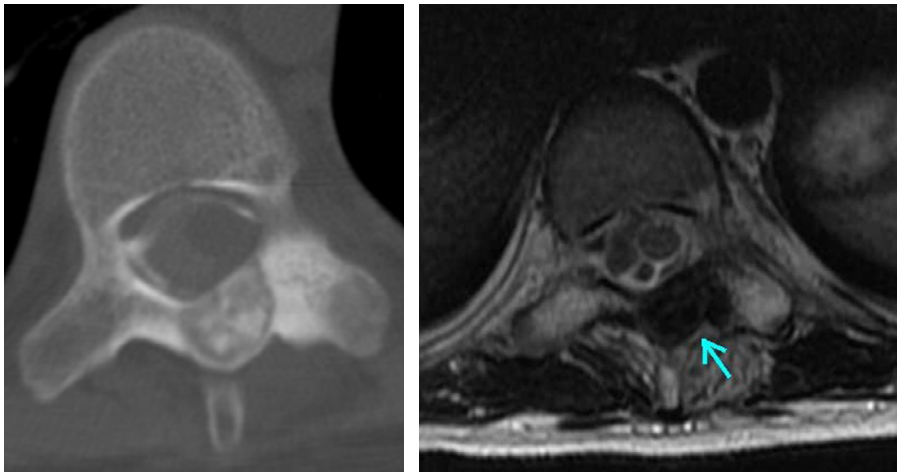

Fig. 4. CT revealed a mass lesion in the lamina of the 10th thoracic vertebra. The lesion had a visible nidus (arrow).

medulla of the skeleton, they usually affect the lower extremities. Approximately 10%–25% of osteoid osteoma cases manifest in the spine [5–9], with a tendency to involve the posterior elements that range from 70% to 100% [14–16]. At an incidence of only 12% of spinal cases, osteoid osteoma of the thoracic spine is relatively rare [10].

Pain is the primary symptom of initial and recurrent disease [17], although reports of osteoid osteoma without the presence of pain exist [18]. In this case report, the SRS-22 questionnaire before surgery revealed a pain score of 5.0. It is difficult to diagnose osteoid osteoma without the presence of pain.

We first became aware of the osteoid osteoma lesion in a CT image. Indeed, preoperative CT is indispensable for not overlooking the presence of osteoid osteoma. Furthermore, 3D-CT revealed that the spinal curvature in this patient had less rotation than that in typical adolescent idiopathic scoliosis (AIS) (Fig. 6), which confirmed the utility of CT before possibly unnecessary scoliosis surgery. Surgical removal of the lesion improved the patient's deformities and survey scores. Retrospective examination of preoperative radiographs showed that pedicle width on the convex side was wide in the apex region. Because the pedicle width on the concave side is narrow due to spinal rotation in AIS patients, pain-related scoliosis may be considered when no vertebral rotation is evident.

Scoliosis with early age onset, long C curve, and left thoracic curve is often associated with a Chiari malformation or syringomyelia [19], whereas a short, sharp, angular curve is usually present in cases of neurofibromatosis type 1 [20]. Meanwhile, scoliosis in Marfan syndrome tends to exhibit longer kyphosis extending through the thoracolumbar junction [21]. Careful examination of curve types is essential for

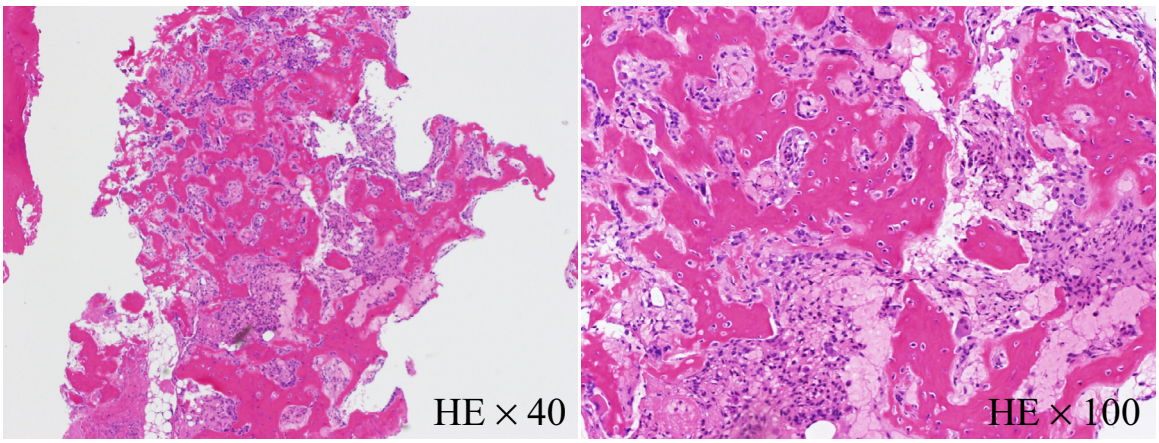

Fig. 5. Histologic examination of the resected tumor with hematoxylin and eosin (HE) staining showed calcific and non-calcific osteoid tissue, which confirmed the diagnosis of osteoid osteoma.

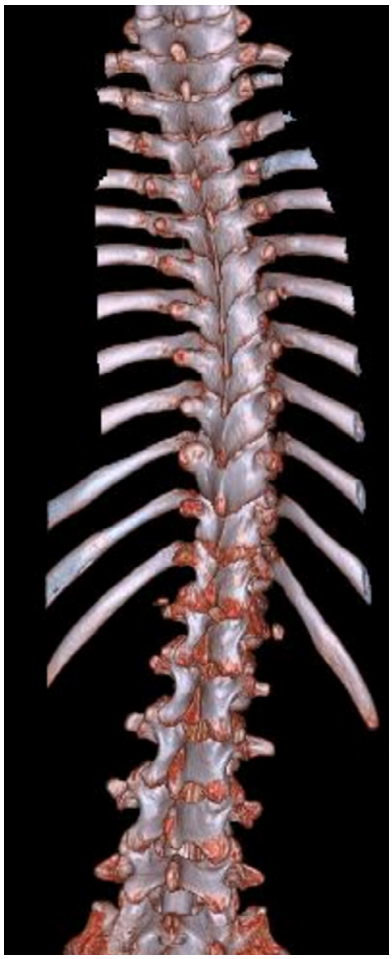

Fig. 6. 3D-CT revealed that the spinal curvature in the patient had less rotation than that in a typical adolescent idiopathic scoliosis.

the correct diagnosis of AIS. A limitation of this case report is a short follow-up period.

Conclusions

We encountered an unusual case of osteoid osteoma of the thoracic spine presenting as scoliosis that was difficult to diagnose. We successfully treated the patient by tumor excision, but continued follow-up is needed. Clinicians should bear in mind the possibility of osteoid osteoma before scoliosis surgery, even in patients without apparent pain, and carefully examine CT findings for evidence of tumor involvement.

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