

CASE REPORT

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# Unusual presentation of aneurysmal bone cyst with scoliosis: a case report

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## Abstract

**Background:** Aneurysmal bone cysts are benign bone tumors that not uncommonly involves the spine. However, this involvement can cause scoliosis, albeit rarely. This report focuses on the importance of proper management for complete tumor resection to prevent recurrence and spinal deformity.

**Case presentation:** A 12-year-old Middle Eastern boy, with a history of T11 aneurysmal bone cyst resection and bone grafting carried out at another hospital, presented with spine deformity of 4 months' duration. The deformity was not associated with pain or neurological deficit. A whole-spine magnetic resonance imaging with contrast confirmed the recurrence of the aneurysmal bone cyst. Posterior spinal instrumentation with corpectomy of T11 was then performed, and confirmed with histopathology the recurrence of aneurysmal bone cyst. Two years post-corpectomy, deformity correction was done from T5–L4.

**Conclusion:** Management of aneurysmal bone cysts requires meticulous planning and full excision to prevent recurrence, especially in the growing spine. If neglected, it can cause major spinal deformities and cord compression, which places a medical burden on the patient and family. To avoid such complications, treating aneurysmal bone cysts along with scoliosis correction can prevent deformity progression.

**Keywords:** Aneurysmal bone cyst, Scoliosis, Vertebral column resection

## Background

Aneurysmal bone cysts (ABC) are benign aggressive bone lesions that account for 1.5–6% of primary bone tumors [1]. More than half of these tumors affect young patients under 20 years of age [2]. These tumors can occur in the spine in 6–20% of cases and specifically 34% in the thoracic spine [3]. Scoliosis and kyphosis manifest in roughly 10–15% of the cases [3].

Van Arsedale first described aneurysmal bone cysts as ossifying hematomas in 1893, and in 1942 the term ABC began to be used [4]. The pathophysiology of ABCs

is debated, but most authors suggest that they develop owing to intercellular edema caused by the primary lesion expanding the surrounding loose stroma and permitting rupture of vessels into the microcysts under hemodynamic pressure, thereby causing a blood-filled cyst [5]. We present a case of a child who presented with symptoms of painless scoliosis with recurrence of the ABC. Subsequently, the patient underwent corpectomy and reconstruction with cage and bone graft. At follow-up 2 years post-surgery, the deformity had worsened and required long spinal fusion and deformity correction from T5–L4.

## Case presentation

A 12-year-old Middle Eastern boy, medically free, presented at our spine clinic complaining of spinal deformity for around 4 months. The deformity was not associated

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with pain or neurological symptoms. The patient provided a history of an aneurysmal bone cyst of T11 vertebra that had been resected with bone graft augmentation and posterior instrumentation T10–T12 at a different hospital 2 years ago. The diagnosis of ABC was confirmed by histopathology. There was no family history of malignancy or scoliosis.

#### Clinical findings

The examination was performed at the time of the initial presentation. It revealed mild scoliotic deformity upon Adam's forward bend test with full spinal range of

motion. There was no neurological deficit. Hips and feet were unremarkable.

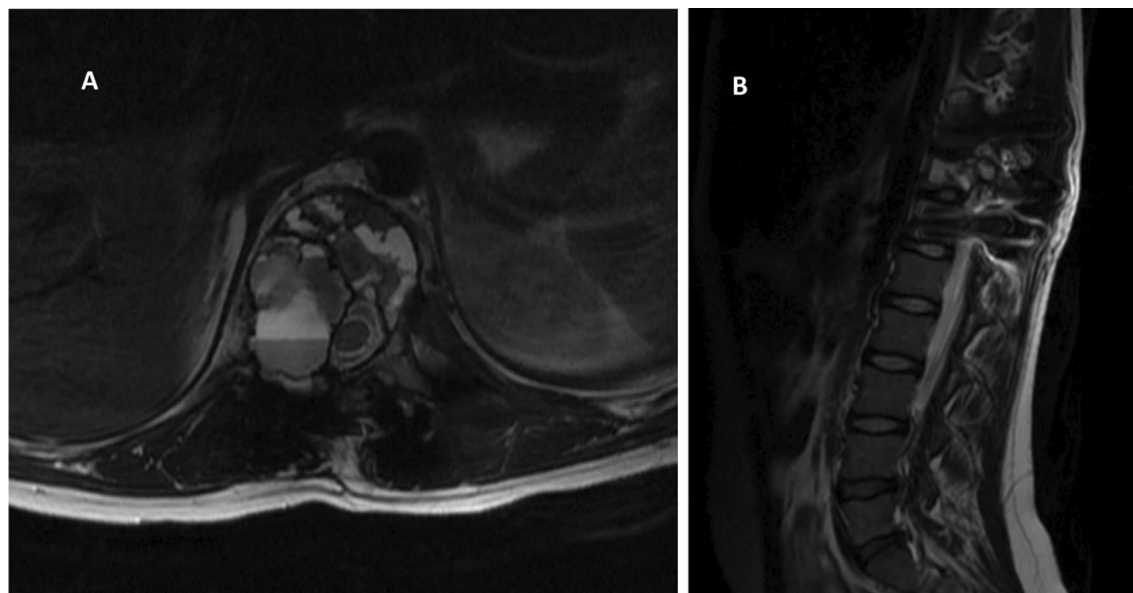
A full spine X-ray confirmed the finding of left scoliotic deformity with a Cobb angle of 24 along with posterior spinal instrumentation from T10–T12 with an open triradiate cartilage Rieser 0 (Fig. 1A, B). A full-spine magnetic resonance imaging (MRI) showed recurrence of the aneurysmal bone cyst (Fig. 2A, B). A diagnosis of juvenile idiopathic scoliosis was made.

#### Therapeutic intervention

Six weeks after presentation, the patient was admitted for surgery. Prior to surgery the patient was taken for



**Fig. 1** A, B Scoliosis X-ray A AP, and B lateral showing posterior spinal instrumentation from T10 to T12 with an open triradiate cartilage Reiser 0 along with left scoliotic deformity with Cobb angle of 24



**Fig. 2** **A, B** MRI **A** axial and **B** sagittal showing Expansile bone lesion with fluid–fluid level at T11, keeping with ABC causing moderate compression fracture with extension into posterior elements causing severe right foraminal stenosis and mild cord compression

embolization, but following an angiogram, no embolization was performed in view of the mild vascularity and the origin of posterior spinal artery from the same level. The procedure consisted of revision posterior spinal fusion and instrumentation, T9–L1 (5 levels), laminectomy, T10–T12 (2 levels), open biopsy T11, tumor resection, navigation-assisted T11 revision of corpectomy, and anterior fusion T11 with cage (Fig. 3A, B). The final pathology result confirmed the recurrence of ABC (Fig. 4). Upon follow-up, we noticed that the child had developed juvenile idiopathic thoracolumbar scoliosis, which increased to 11 degrees (Fig. 5A, B). One year post-surgery, an MRI confirmed no recurrence of ABC (Fig. 6A, B). Two years post-surgery, scoliosis had reached 30 degrees (Fig. 7A, B). Regarding growth stage, the child was at Risser stage 0. A trial of brace treatment was carried out, but unfortunately the child was not compliant with the brace. An MRI done at that time showed no recurrence. Hence the decision was made to proceed with scoliosis correction and extend the fusion to T5 proximally and to L4 distally. At 1-year follow-up, scoliosis x-ray (Fig. 8A, B) showed maintenance of scoliosis correction without implant complications, and a final MRI was done which showed no recurrence. The patient believed that the treatment had improved his quality of life and increased his satisfaction.

## Discussion

We present a case of an aggressive aneurysmal bone cyst of the thoracic spine that presented with juvenile scoliosis and showed recurrence of the ABC; MRI confirmed the diagnosis, along with a histopathological examination. The patient underwent posterior spinal fusion and instrumentation of five levels from T9 to L1 via a one-stage posterior and anterior approach, decompression, open biopsy, complete tumor resection, and anterior fusion of T11. Post resection of the tumor, the initial scoliosis was resolved. Upon follow-up, there was no recurrence of the tumor; however, there was progression of scoliosis. The patient was treated with one-stage long spinal fusion and deformity correction from T5 to L4 via an anterior and posterior approach. To our knowledge, this is the first case in literature that reports progressive spinal deformity along with complete tumor resection that was treated with a one-stage anterior and posterior approach. One of the most important factors to prevent tumor recurrence is the extent of tumor resection. Zenonos et al. reported that of 14 patients, two showed recurrence owing to incomplete tumor resection [6]. Recurrence of ABC is around 10–44%, and 90% recur within 2 years [7]. Rahimizadeh et al. published a case in 2013 of ABC at C7 level, where the patient underwent a one-stage anterior and posterior approach and showed no recurrence after 1 year [8].

ABC usually presents in the second decade of life. It accounts for approximately 1.5–6% of all primary bone tumors, and 15–20% of all primary spine tumors [1][1].

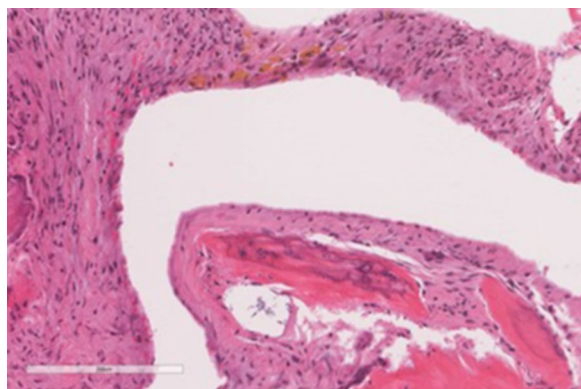


**Fig. 3** A, B scoliosis X-ray **A** anteroposterior (AP) and **B** lateral, 4 months post first surgery showing T11 resected vertebral body with cage placement, thoracolumbar fixation rods and screws, with a scoliosis measure of 6.5°

The areas of predilection are the femur, tibia, humerus, spine, pelvis, ribs, and small bones of the hands and feet [10]. Most commonly, the posterior elements of the spine are involved [11]. Some studies report more incidence in the lumbar spine [12, 13]. Others report more incidence in the cervical and thoracic spine [14, 15], or equal incidence in the thoracic and lumbar regions [6, 16].

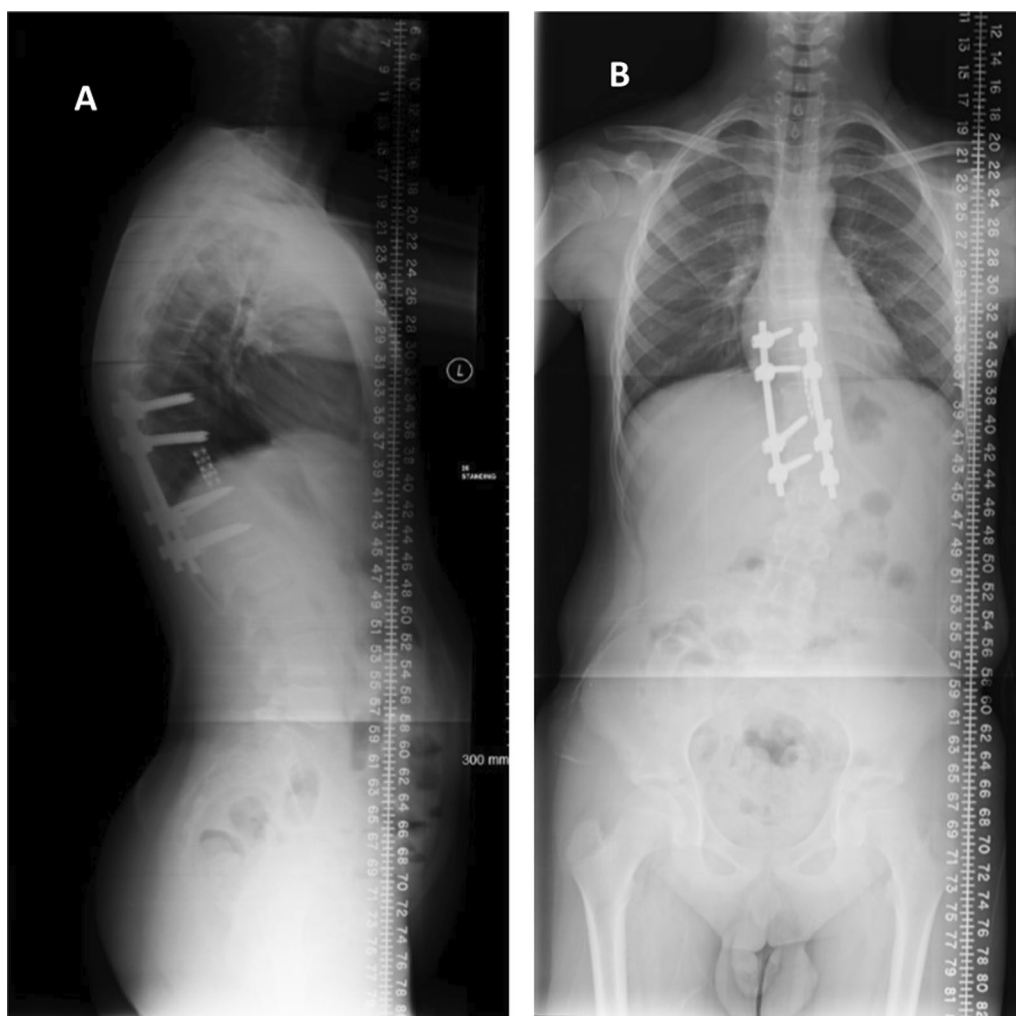
Patients present with neurological symptoms in 60–70% of cases [17]. Many neurological signs and symptoms may be noted, ranging from backache and radiculopathy to paraplegia depending on the level. Acute spinal cord compression may occur if there is a break in the posterior cortex of the body [18]. Scoliosis and kyphosis are noted in 10–15% of cases owing to back pain, spasms, or





**Fig. 4** Photomicrograph (haematoxylin–eosin-stained  $\times 20$ ) of the tumor showing fragments of bone tissue cystic space walled by fibroblastic tissue, area of hemorrhage, multinucleated giant cells, fibrosis, and hemosiderin deposition

weakness [3]. Konbaz et al. mentioned that coexistence of tumor with scoliosis is present in the literature [19]. Many techniques should be used to arrive at the diagnosis and for preoperative planning. X-ray shows eccentric, osteolytic, expansive, and trabeculated lesions with fine-walled cystic cavities. Loss of cortical contours can mimic a malignant lesion [20]. A spine X-ray should be performed as an initial workup examination, although it does not reveal the presence of ABC [21]. CT scans also show internal septation and eggshell appearance with calcified rim, as well as fluid level [22], but these findings were not exclusive. For fluid, the patient should stay 10 minutes in position to obtain enough separation of the materials of different attenuation. CT was also needed for preoperative fusion and instrumentation planning of pedicles and transverse processes [23]. MRI allows for



**Fig. 5** A, B Scoliosis X-ray A AP and B lateral, 1 year post first surgery showing T11 resected vertebral body with cage placement thoracolumbar fixation rods and screws with S-shape scoliosis Cobb angle of  $11^{\circ}$

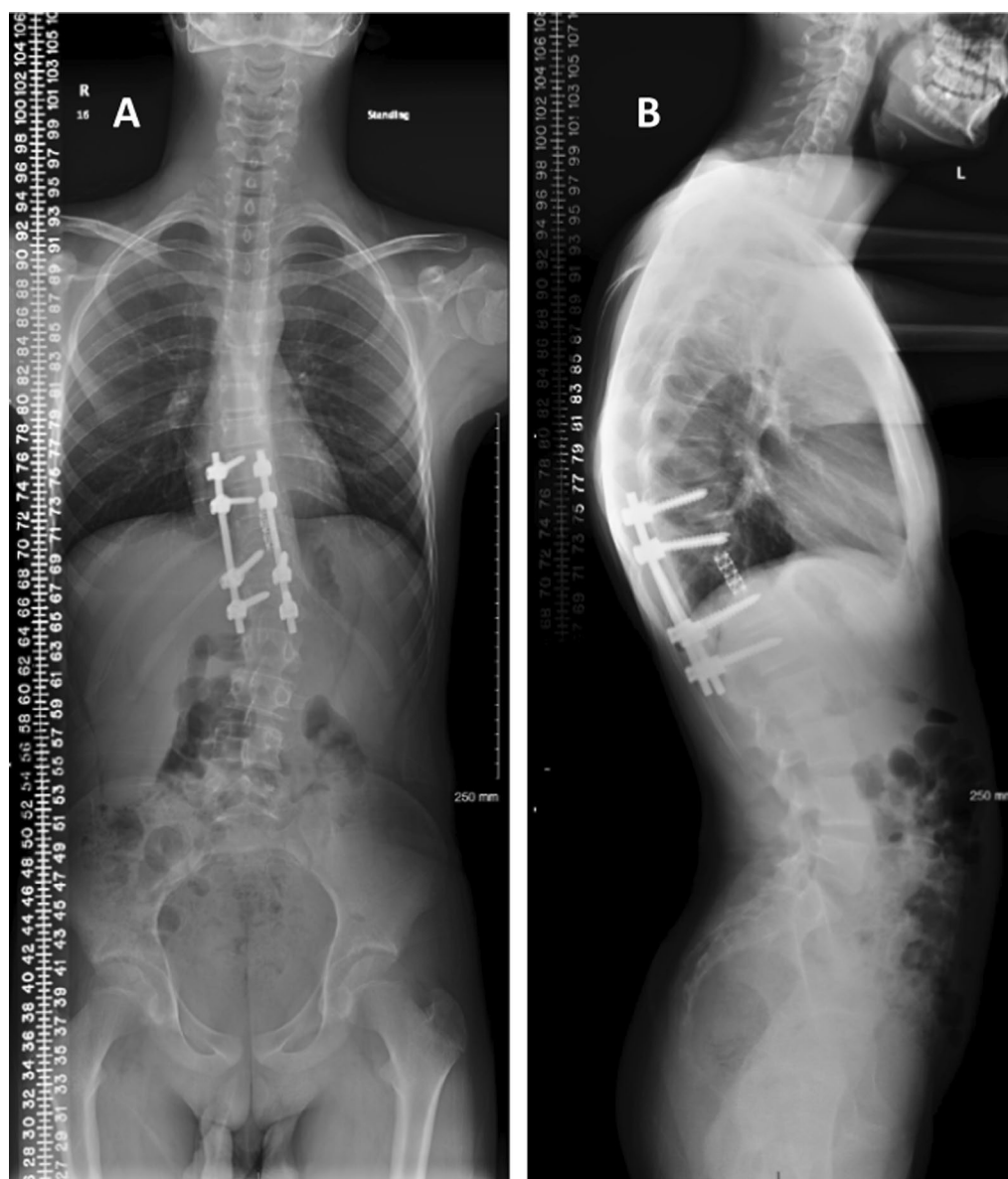


**Fig. 6** **A, B** MRI **A** axial and **B** sagittal showing partially resected T11 with presence of a vertical mesh cage in place with no evidence of residual or recurrent aneurysmal bone cysts

visualizing a well-defined lesion with lobulated contours and liquid inside it. Internal septa showed a decrease in the signal, presumably owing to the presence of fibrous tissue. Liquid characteristics of ABC are better explored in MRI [24]. Kransdorf et al. used MRI even for follow-up as it is superior to biplanar radiographs [7]. Bone scintigraphy showed increased uptake of radionuclides in the peripheral area of the lesion in most cases [21]. In some patients, the radiological findings are inconclusive, so an open biopsy is necessary to establish the definitive diagnosis because it can determine grading and signs of tumor malignancy [25]. The character of ABC histology is cavernous channels surrounded by a spindle cell stroma with osteoclast-like giant cells and osteoid production [26]. Differential diagnoses are simple bone cysts, hyperparathyroidism brown tumor, giant cell tumor, chondrosarcoma, osteosarcoma, and Ewing's sarcoma [25]. At the microscopic level, findings are dense and cellular composition of the cyst, containing plump stromal cells, multinuclear giant cells, and thin-walled blood vessels, or are in the form of preponderantly fibrous tissue with enlarging vascular spaces [27].

Treatment of ABC is also controversial. Because of their unique anatomical structure and function, there are special considerations when managing ABCs of the spine. Treatment options are curettage with or without bone grafting, complete excision, arterial embolization, intralésional drug injections, and radiation [22]. Curettage alone

has shown highly variable recurrence rates, reaching 59% in some studies [28]. Vertebroplasty or kyphoplasty can be used after curettage to reinforce bone defects [29]. Radiotherapy is recommended in inoperable cases [14]. Arterial embolization can be used as an adjunct to surgery, but it has also been employed as a primary treatment in ABC lesions that are difficult to access when surgery is not feasible [18]. According to Park et al., treatment of ABC is usually surgical, but embolization and radiotherapy can be used without surgery when it will cause significant morbidity plus medical management with denosumab [30]. Regarding our patient, his initial surgery at a different hospital was not done appropriately. Therefore, he presented with recurrence of tumor and scoliosis. The second surgery, which we carried out, achieved complete tumor resection and stabilization; however, we did not address his scoliosis. We had to carry out a third procedure to address his scoliosis with deformity correction and fusion. What is unique about our case is that post-tumor resection, the patient continued to grow and his scoliosis progressed; however, there was no tumor recurrence. This young patient had to undergo three spine surgeries in the space of almost 4 years. To achieve complete tumor resection and prevent recurrence, careful planning in the index surgery with meticulous tumor resection along with spinal reconstruction and fusion is extremely important and might have spared the patient all these sequelae. The literature lacks a well-structured protocol in such cases as to

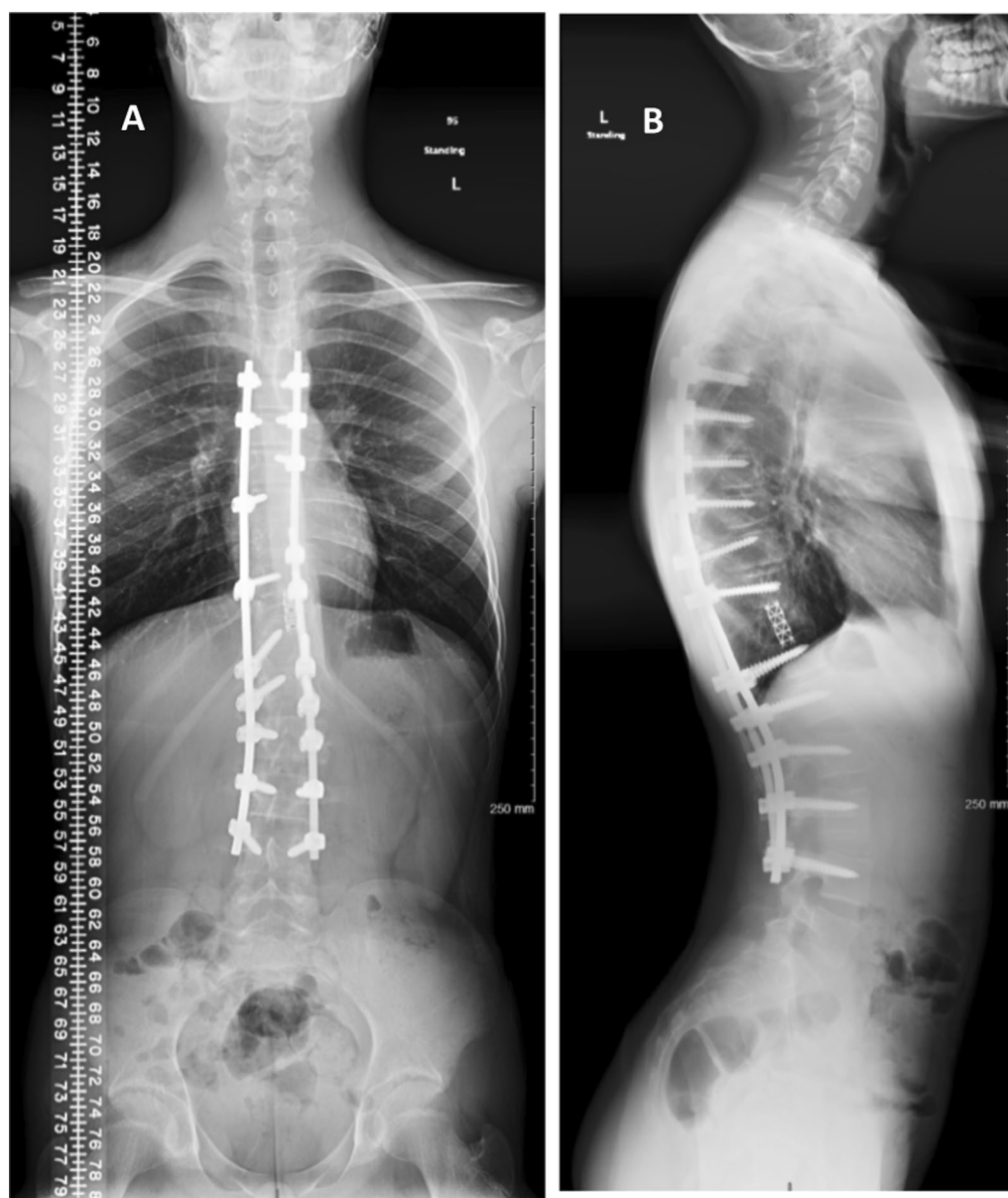


**Fig. 7** A, B Scoliosis X-ray **A** AP and **B** lateral, 2 years post first surgery showing T11 resected vertebral body with cage placement thoracolumbar fixation rods and screws with S-shape scoliosis with Cobb angle of 30°

whether the tumor alone is treated or both the tumor and spinal deformity are treated at the same time, especially for a patient who is still growing. However, in young patients with a growing spine it is important to preserve the alignment of the spine and allow natural development until growth is completed [31]. We believe more cases need to be published to establish a clear guideline for proper management of such cases and to avoid future complications.

### Conclusion

Recurrent ABC can be challenging to manage as it can result in spinal deformity, instability, and surgical impairment, posing a significant burden on the child and their family. Although in cases of tumor coexisting with scoliosis the usual recommendation is to manage the tumor first, we believe that discussing the option of managing both problems at one stage is a valid consideration. Therefore, identification of



**Fig. 8** **A, B** Scoliosis X-ray **A** AP and **B** lateral, 1 year post revision surgery showing T11 resected vertebral body with cage placement, and posterior spinal fixation from T5 to L4 with scoliosis improvement and no complication is seen

an appropriate index treatment that decreases recurrence rates and complications is crucial for the treatment of ABC.

#### Abbreviations

ABC: Aneurysmal bone cyst; MRI: Magnetic resonance imaging.

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#### Author contributions

All authors were involved in drafting, editing, and approving this manuscript. Drs. Yousef and Hisham were involved in the literature review, writing, reviewing the first draft, and providing the figures with full description of the findings for the manuscript. Dr. Faisal was involved in critical editing and revising the first and final draft of the manuscript. Drs. Sami, Majid, and Fahad were involved in revising and approving the final draft. All authors were approving the final manuscript. All authors read and approved the final manuscript.

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### Availability of data and materials

All data generated or analyzed during this study are included in this published article and its supplementary information files.

### Declarations

#### Ethics approval and consent to participate

Case report was ethically approved by Imam Abdulrahman Bin Faisal University IRB committee with an IRB number PGS-2022-01-104. The patients' Guardian (Father) provided informed consent for his son's case to participate.

#### Consent for publication

Written informed consent was obtained from the patient's legal guardian for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

#### Competing interests

The authors declare that they have no competing interests in this section.

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