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Case Report

Management of aggressive recurrent thoracic spine aneurysmal bone cyst in a 7-year-old male: A case report and review of the literature

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ABSTRACT

Background: Spinal aneurysmal bone cysts (ABCs) are rare, histologically benign tumors with aggressive behavior, which may cause bone and soft-tissue destruction, particularly affecting neural elements. Management of these tumors, including treatment modalities and follow-up protocols, remains challenging.

Case Description: A 7-year-old boy presented with chest wall pain persisting for two months before admission, accompanied by progressive mono paresis lasting ten days before admission. Myelopathy signs were evident during the examination. Imaging confirmed a multicystic lesion at the T6 level involving the posterior elements of the vertebra, with significant cord compression. Due to deteriorating neurological function, he underwent urgent laminectomy and neural decompression, followed by subtotal tumor resection. Postoperative histopathological examination confirmed the diagnosis of an ABC, and the patient experienced significant neurological recovery. However, after 21 days, the patient was readmitted to the emergency department with severe paraparesis. Magnetic resonance imaging revealed rapid growth of the residual tumor, leading to cord compression. He underwent aggressive total tumor resection, T6 vertebral body corpectomy, and fixation with pedicle screws and cage insertion. Following the second surgery, prompt neurological recovery occurred.

Conclusion: This rare case report emphasizes the importance of a close follow-up protocol for spinal ABCs in the pediatric population. It highlights the challenges in managing these tumors and the need for vigilant monitoring to detect and address rapid recurrences.

Keywords: Aneurysmal bone cyst, Follow-up, Pediatric, Thoracic spine

INTRODUCTION

Aneurysmal bone cyst (ABC) tumors are rare bone lesions, representing about 1.4% of all primary bone tumors and 15% of all primary spine tumors.[7,9,48] In the spine, the thoracic and lumbar regions are the most common sites of involvement.[18,29] The most common presenting symptoms are nocturnal pain, followed by paresthesia in the lower extremities and radicular pain in the lower extremities, chest wall, and abdomen.[7,11,27] Acute onset of paraplegia has also been reported.[11,35]

ABCs are histologically benign; however, due to the possibility of rapid growth and destruction of adjacent bones and soft tissues, they can show aggressive behavior.[43] ABC tumors may lead to spinal cord compression without complete vertebral body collapse.[9,11] Histologically, they are composed of large cavities containing blood.[7,41] Both computed tomography (CT) scans and magnetic resonance imaging (MRI) are helpful for diagnosis of ABCs.[11,41] Extent of the lesion can be well assessed by MRI due to better contrast resolution.[6,41] In MRI, a multiloculated lytic eccentric expansile lesion with multiple internal septations and fluid-fluid levels is characteristic of ABC tumors.[9,23,41]

Treatment options for spine ABC tumors include total excision, radiotherapy, and embolization.[10,11,16,35] *En-bloc* resection is associated with a lower risk of recurrence; however, particularly in the spine region, *en-bloc* resection is not always achievable due to proximity to neural elements.[4,17,24,28,37,42,44] Laminectomy/decompression and subtotal tumor resection are associated with a high rate of recurrence. (42.3% vs. 8.3% in cases of complete resection).[37] Data regarding the success rate of any of the therapeutic modalities are scarce. Therefore, there is still no standard specific treatment protocol for the spinal ABCs.[16]

Due to the uncertainty of the treatment protocol and the high probability of recurrence, close follow-up has been suggested as an irreplaceable part of any therapeutic strategy in every patient with ABC. However, it is still not clear what the appropriate interval for postoperation follow-up is. In this report, we present a case of thoracic spine ABC tumor in a 7-year-old male with aggressive behavior, which recurred 21 days after primary decompression.

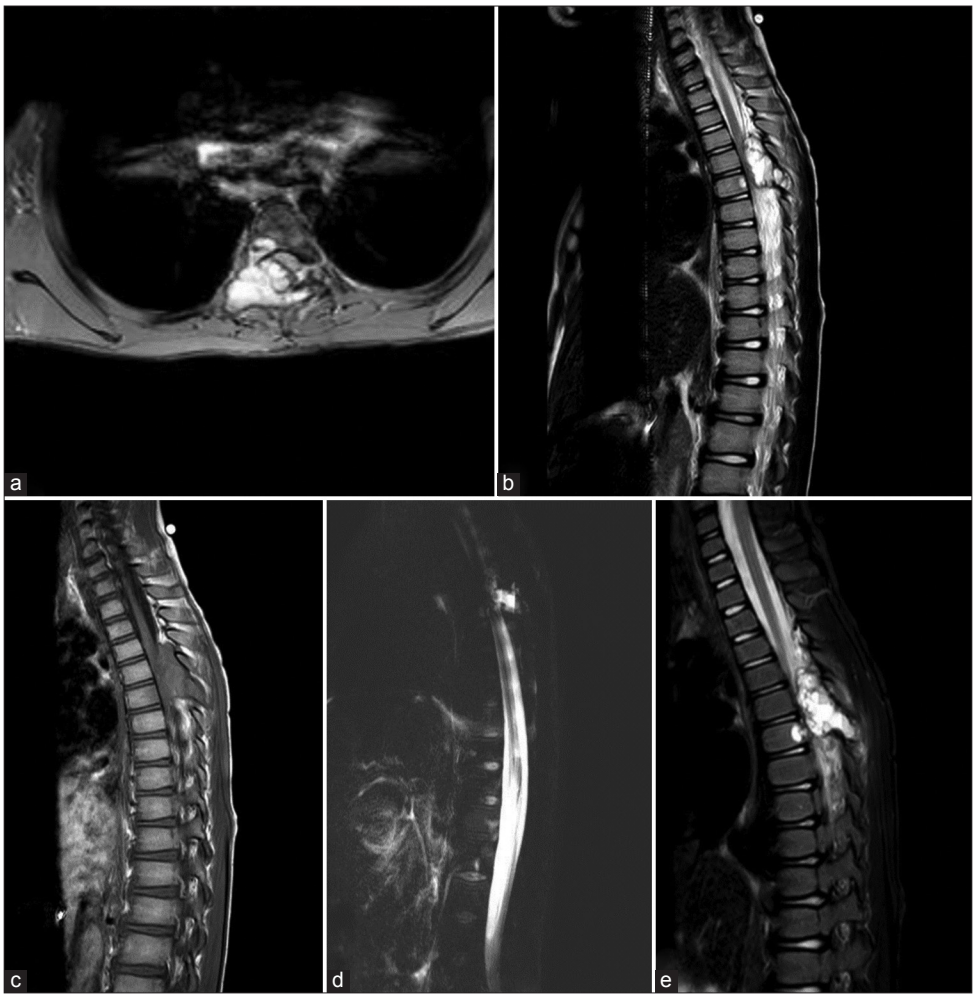

Figure 1: Preoperative magnetic resonance imaging of the patient: (a) Axial T2 view, (b) sagittal T2 view, (c) sagittal T1 view, (d) myelogram, and (e) short tau inversion recovery (STIR) view showed expansile and heterogeneous cystic bone lesion with fluid signal intensity (T1: Hypointense with T2 and STIR hyperintense) and thin internal septation and cord compression at the T6 vertebra.

CASE PRESENTATION

A 7-year-old boy with no prior medical history presented at the emergency department with acute left lower extremity monoplegia and sensory loss below the T6 dermatome, which had been ongoing for ten days before admission. Notably, there was no bowel or bladder dysfunction. On physical examination, spasticity in his lower limbs and reduced sensation to pinprick, temperature, and vibration/position below the T6 dermatome were evident. He had been experiencing chest wall pain for the past three months, particularly worsening at night. There was no history of trauma or recent infections reported. All laboratory tests were within normal ranges.

An MRI scan revealed a cystic tumor with internal septations involving the spinous process, lamina, pedicle, and body of the T6 vertebra [Figure 1]. Urgent surgical decompression was performed, and the tumor was subtotally resected. Histopathological findings were consistent with a diagnosis of ABC. Postoperatively, the patient experienced significant neurological improvement. One day after the operation, he could ambulate with assistance. The patient was discharged four days post-surgery. A follow-up MRI showed minimal lesion residue with slight thecal sac compression [Figure 2]. In this regard, approximately 5–10% of tumor residue was observed in the MRI, primarily concentrated in the anterior part (posterior aspect of the vertebral body). In the initial operation, the approach involved incomplete intralesional excision, encompassing laminectomy with posterior tumor resection. However, the tumor involving the pedicle and vertebral body was not resected during the initial procedure.

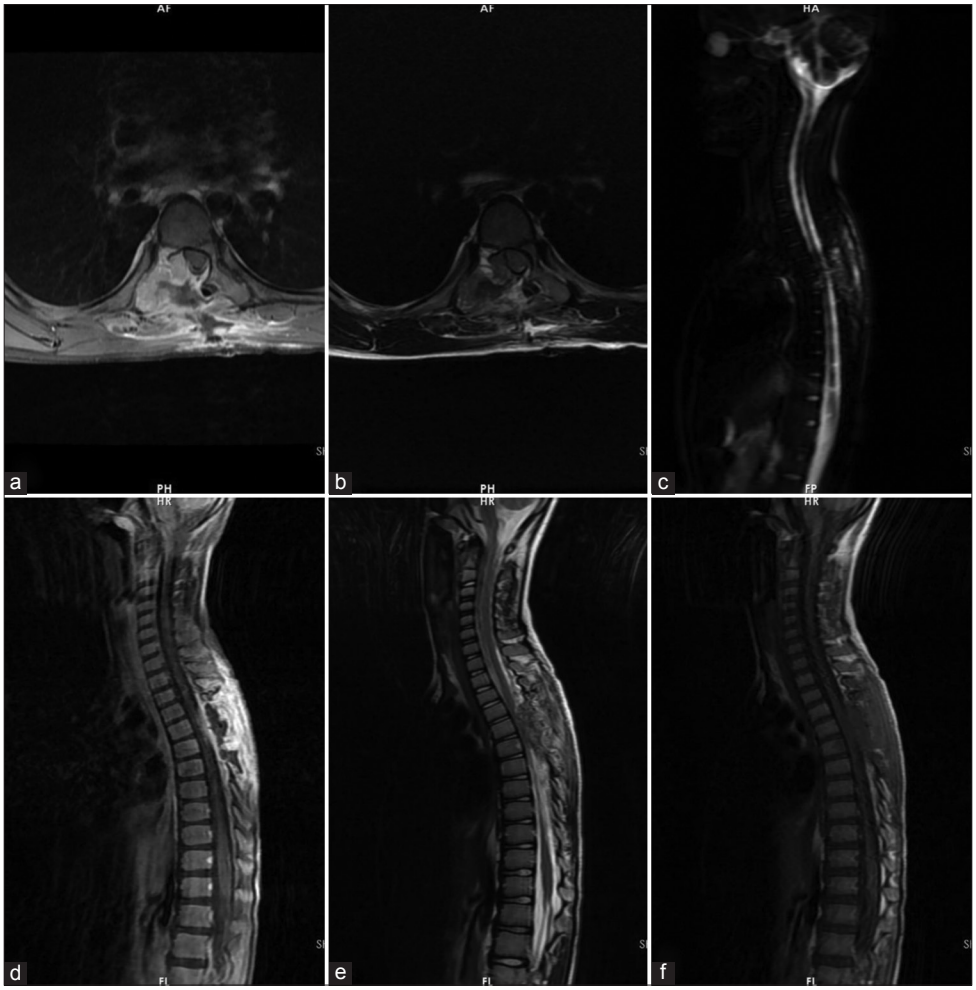

Figure 2: Follow-up magnetic resonance imaging study of the patient, ten days after the first surgery: (a) T1 with contrast axial view, (b) T2 axial view, (c) myelogram, (d) T1 with contrast sagittal view, (e) T2 sagittal view, and (f) T1 sagittal view showed an expansile and heterogeneous cystic bone lesion with fluid signal intensity (T1: Hypointense with T2 and short tau inversion recovery (STIR) hyperintense) and thin internal septation and cord compression at the T6 vertebra. As shown above, laminectomy and cord decompression were performed. Tumor remnants at the pedicle and posterior parts of the body can be seen. The spinal cord is decompressed relatively.

Twenty-one days after the operation, the patient returned to the emergency department with acute and rapidly worsening severe paraparesis that had started three days prior. The motor strength in both lower extremities was 1/5. MRI revealed rapid tumor growth and significant spinal cord compression [Figure 3]. An angiogram demonstrated large vessels supplying the tumor, leading to embolization performed before surgery. The patient was scheduled for surgery, and T6 posterior vertebral column resection with posterior T3–T9 pedicle screw fixation was carried out [Figure 4]. Considering the resection of the cyst wall and all grossly abnormal tissue, along with the *en-bloc* resection of the T6 vertebra after removing the superior and inferior intervertebral discs, we have determined that a complete resection was achieved.[20] One day after the surgery, the patient regained the ability to move his legs. Postoperative MRI scans confirmed total tumor resection. The patient can now walk independently one month after the surgery.

DISCUSSION

ABCs are rare, with an annual prevalence of 0.32/100,000 young population.[50] They are more prevalent in the first two decades of life.[26] Historically, these tumors were believed to result from trauma or increased venous blood flow.[25] However, today, due to molecular findings such as the TRE17/USP6 gene (abnormalities of the short arm of chromosome 17), ABC tumors are considered neoplasms.[3,4,31,33,49] Spinal ABC tumors can present with nocturnal pain. Patients may also experience paresthesia, paresis, or radicular pain extending to the lower extremities, abdomen, or chest wall.[7,11,27]

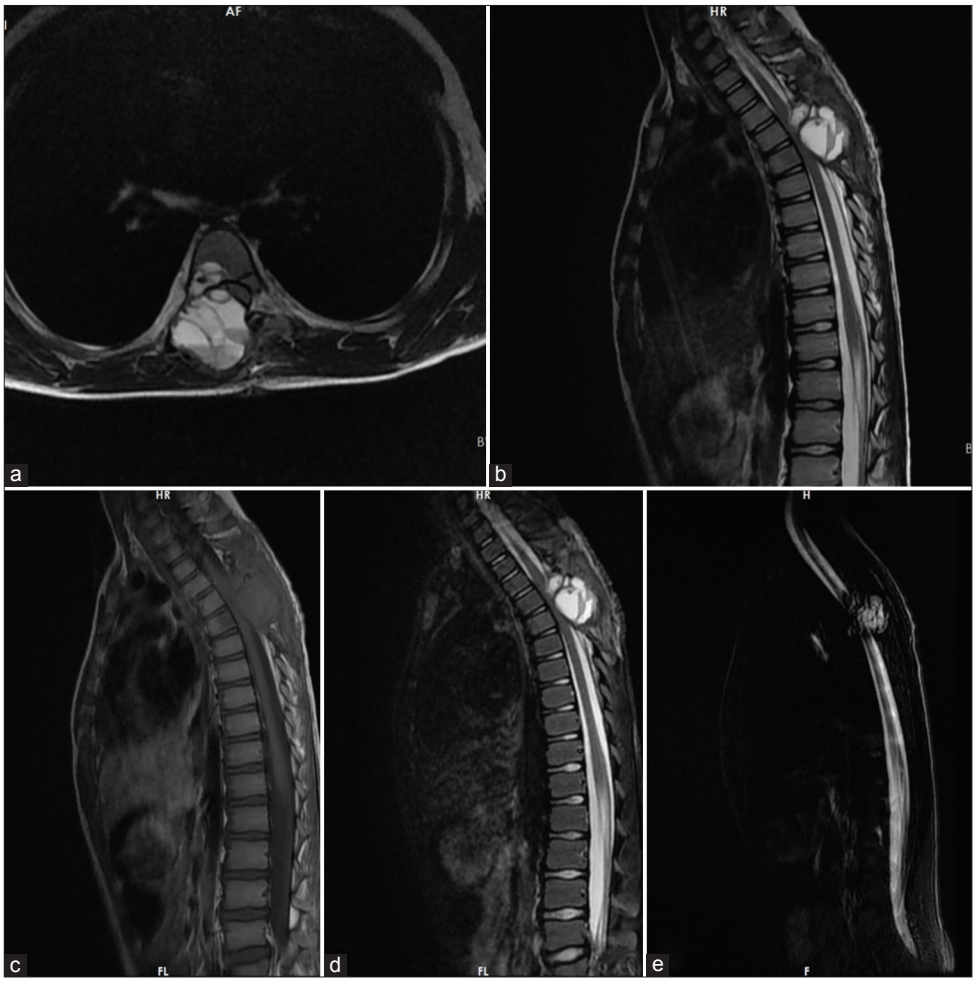

Figure 3: Magnetic resonance imaging study of the patient before the second surgery (21 days after the first surgery): (a) T2 axial view, (b) T2 sagittal view, (c) T1 sagittal view, (d) sagittal short-tau inversion recovery (STIR) view, and (e) myelogram showed an expansile and heterogeneous cystic bone lesion with fluid signal intensity (T1: Hypointense with T2 and STIR Hyperintense) and thin internal septation and fluid-fluid level and significant cord compression at the T6 vertebra. As seen above, the tumor remnant grew rapidly, leading to cord compression.

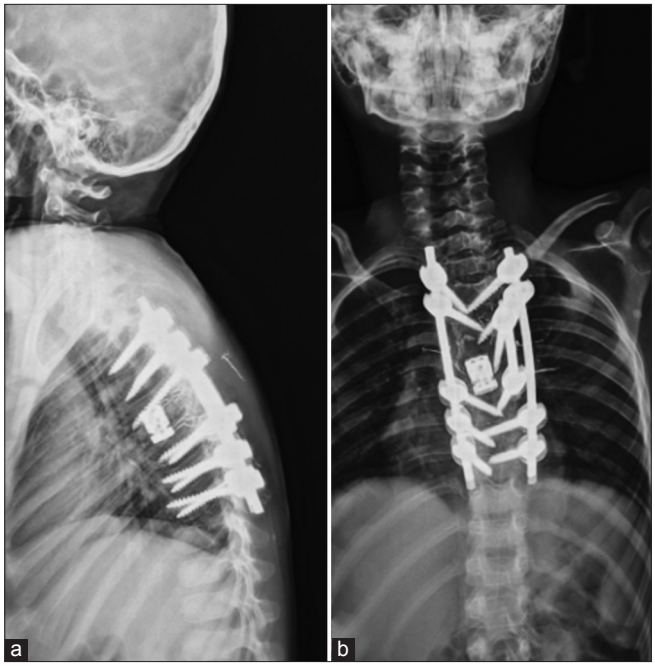

Figure 4: Postoperative plain radiography of the second surgery (a) Lateral view and (b) AP view demonstrated T6 posterior vertebral column resection with posterior T3–T9 pedicle screw fixation.

The rapid growth of a spinal ABC tumor or vertebral body collapse may cause acute paraplegia due to spinal cord compression.[1,4] Although MRI and CT scans are helpful diagnostic methods, the definitive diagnosis is confirmed through percutaneous or open biopsy.[30] The best treatment option for ABC tumors is still debated.[1] *En-bloc* resection results in the lowest recurrence rate and is the preferred treatment for patients with neurological deficits.[4,8,22,28] Nevertheless, recurrence despite complete resection has been reported.[18] En bloc resection is not always achievable, and partial excision is associated with a high recurrence rate.[4,22,37] Radiotherapy may cause secondary malignancy[35] and, in some cases, kyphosis.[17] Therefore, the use of radiotherapy is limited to inoperable lesions.[35] Preoperative embolization is recommended to reduce blood loss during surgical excision.[2,35] Other treatment options, such as injection of hydroxyapatite osteoconductive cement,[18] denosumab,[38] and doxycycline[44] have been mentioned in the literature. Furthermore, intraoperative adjuvant therapies, such as high-speed burr, argon beam coagulation, phenol, and cryotherapy with curettage, have demonstrated a lower recurrence rate.[13,36,39,40,45] It is worth noting that while adjuvant therapies have improved

Table 1: Previous reports of spinal aneurysmal bone cyst tumors in children who presented with neurological deficits and underwent surgery.

Authors/year	Case presentation	Level	Treatment protocol	Postoperative outcome	Follow-up duration
Nisson *et al.*/2019[32]	13 Y/O (F) with complete paraplegia	T4	T1–T7 fixation and complete spondylectomy and cage insertion	The patient regained full strength in both lower extremities	9 m
Alanazi *et al.*/2022[1]	7 Y/O (M) with paraplegia and sensory level at T3	T3	Spinal cord decompression, posterior spinal instrumentation, and complete tumor resection	Full recovery in sensory and motor functions	7 m
Mehta *et al.*/2017[30]	16 Y/O (M) with acute paraplegia	T5	360° decompression and insertion of the expandable cage and pedicle screw fixation	Complete rehabilitation of sensory and motor function	18 m
Frassanito *et al.*/2019[18]	15 Y/O (M) with acute severe paraparesis	T7	1st surgery: Subtotal resection and spinal cord decompression 2nd surgery: total resection 3rd surgery: Percutaneous osteoconductive cement injection 4th surgery: Re-injection of percutaneous cement	Fully neurological recovery	24 m
Codd *et al.*[11]	8 Y/O (M) with progressive low back pain	L3	1st: Embolization before surgery 2nd: Both a retroperitoneal and a posterior approach to the lumbar spine and complete tumor resection with Vertebral body excision	Low back pain-free	3 m

Table 2: Previous reports on recurrence of aneurysmal bone cyst.

Author/Year	Number of cases	Tumor location	Treatment modalities	Recurrence time interval
Szendröi *et al.*/1992[46]	68	Humerus Tibia Pelvis Femur	1. Curettage 2. *En bloc* resection 3. Segment resection	Mean: 14 months
Vergel *et al.*/1992[48]	238	Femur Tibia Pelvis Spine	1. Curettage 2. Curettage with adjuvant therapy 3. Wide excision with or without radiotherapy 4. Wide amputation with radiotherapy	4 months to 2 years
De Kleuver *et al.*/1998[14]	31	Spine	1. Curettage 2. *En bloc* resection/subtotal resection 3. Curettage with radiotherapy 4. Radiotherapy 5. Selective arterial embolization	1–21 months
Gibbs *et al.*/1999[19]	40	Extremities	1. Curettage 2. Excision	2–24 months
Boriani *et al.*/2001[7]	41	Spine	1. Curettage 2. Resection 3. Curettage with radiotherapy 4. Selective arterial embolization	1–12 months
Papagelopoulos *et al.*/2001[34]	40	Sacrum Pelvis	1. Excision-curettage 2. Intralesional excision	<18 months
Cottalorda *et al.*/2005[12]	21	Femur Tibia humerus Clavicle Fibula radius Pelvis Calcaneus	1. Curettage 2. Resection 3. Injection of fibrosing agent 4. Injection (Methylprednisolone acetate)	3–13 months
Mankin *et al.*/2005[28]	150	Tibia Femur Pelvis Humerus Spine	1. Curettage 2. *En bloc* resection 3. Intralesional resection	0.3–3 years
Başarir *et al.*/2007[5]	56	Humerus Femur Fibula	1. Intralesional excision 2. Radiotherapy 3. Intralesional excision with adjuvant therapy	6–50 months (Mean 15 months)
Hauschild *et al.*/2016[21]	28	Long bones Spine Pelvis	1. Curettage 2. Embolization 3. Watch and wait after the biopsy	6–31 months
Frassanito *et al.*/2019[18]	1	Spine	1. Subtotal resection followed by total resection	1.3 months after subtotal resection 3 months after total resection
Jahangiri *et al.*/2024	1	Thoracic Spine	1. 1st surgery: Subtotal resection 2. 2nd surgery: Selective arterial embolization with total resection (PVCR and posterior fusion)	21 days after the first surgery

local control rates, their availability is limited to some specialized centers, particularly in developed countries.[47] Table 1 summarizes previous reports of spinal ABC tumors in children who presented with neurological deficits and underwent surgery.

Here, we present the case of a 7-year-old male who exhibited progressive sensorimotor symptoms and experienced significant improvement after the initial spinal cord decompression surgery. While the postoperative MRI study, conducted ten days after the surgery, showed significant cord decompression, the

patient was advised to undergo another imaging study 3 months later and return for the next visit. However, the patient returned to the ER with paraparesis 21 days after the initial operation. As shown in Table 2, there is no consensus regarding the intervals for follow-up in patients with spinal ABCs who undergo surgery. The aggressive behavior of the lesion observed in our patient has not been previously documented in the literature. Introducing this case may be valuable in establishing an imaging follow-up strategy for patients with spinal ABCs who have undergone subtotal resection.

Reviewing the literature, we can find that recurrences have been observed to occur within a range of 1–50 months after treatment. Moreover, as reported by Hauschild *et al.*, 90% of recurrences took place between 6 and 12 months after the initial treatment.[21] Hauschild *et al.* recommended routine follow-up protocols involving clinical and MRI examinations at 3, 6, 12, 18, and 24 months, followed by yearly check-ups up to the 5th year.[21] Codd *et al.* suggested close follow-ups every three months following treatment.[11] Desai *et al.* suggested MRI studies regularly for all patients who experienced symptoms in favor of tumor recurrence.[15] According to the literature, the shortest time interval from tumor treatment to recurrence is one month. In this case, we presented that tumor recurrence occurred even in <1 month. Therefore, it appears that a 3-month interval from the time of treatment to the first follow-up visit may not be efficient, and a shorter interval could be considered. However, determining the exact follow-up interval requires further study.

It is worth noting that in the presented case, the tumor residue in the vertebral body grew rapidly, leading to cord compression and neurological deficits. It is hypothesized that involvement or the presence of anterior parts may be associated with an increased probability of symptoms or a higher rate of recurrence, which is consistent with the previous studies.[11,30,32] The optimal treatment regimen for spinal ABC is still unclear.[1] Therefore, vertebral body involvement should be considered as one of the significant factors in the treatment plan. However, further studies are required to determine the exact association.

CONCLUSION

ABC tumors, historically considered benign, have the potential to exhibit aggressive behavior. The optimal treatment approach remains uncertain, and the recurrence rate is relatively high. Therefore, close follow-up is strongly recommended. The current follow-up protocol suggests the first visit should occur three months after the initial treatment. However, in cases of subtotal spinal tumor resection, it appears that closer follow-up may be necessary. Furthermore, additional studies are needed to establish a clearer association between vertebral body involvement and the treatment protocol.

Ethical approval

Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript, and no images were manipulated using AI.

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