

Original article

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Surgical treatment of aneurysmal bone cysts of the lumbar spine: case report

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Abstract

Introduction Pediatric and adolescent patients with aneurysmal bone cyst of the spine are normally treated by orthopaedic, neuro surgeons and oncologists. There is no consensus on the strategy and volume of preoperative and surgical treatment of the condition. There is a higher risk of pathological fracture and secondary neurological complications, tumor recurrence if open surgery is not performed in time. A second surgical intervention can hardly be avoided in such cases. **The objective** was to report the result of surgical treatment of a patient with aneurysmal bone cyst of the spine. **Material and methods** Outcome of a patient with an aneurysmal bone cyst of the L3 vertebra is presented. The technique of using therapeutic punctures as a preoperative preparation to be followed by an open surgical treatment is described. Absence of complaints and tumor recurrence, stability of the metal construct and bone fusion were the evaluation criteria. **Results** The patient was followed for 17 months after open surgery and CT scans showed no recurrence and demonstrated bone fusion at a long term. **Discussion** Many aspects of the course of the disease are to be considered in the treatment of children and adolescents with aneurysmal bone cyst of the spine. In our opinion, the step in the treatment of aneurysmal bone cyst of the spine should be considered Puncture for therapeutic and diagnostic purposes is to be performed to reduce pathological process is reduced, and the procedure to be followed by surgical intervention to remove the tumor. The strategy of surgical treatment would rely on impaired supporting columns of the spine, the degree of neural structure compression and a secondary spinal deformity. The need and extent of metal fixation is to be evaluated, potential vertebral growth identified, expediency of bone grafting after tumor removal, radical excision of the pathological focus to be considered. **Conclusion** The clinical case showed an effective combination of surgical techniques in the treatment of an aneurysmal bone cyst of the spine. Preoperative therapeutic puncture of the cyst allowed for reduction of the pathological process (decreasing pressure inside the cyst, thickening the cortical layer of the cyst and delimiting the tumor from the surrounding tissues), decreased volume of surgical intervention and blood loss due to a decreased activity of the pathological process compared to cases of untimely performed open surgery. Metal fixation in combination with bone auto- and allograft was practical for bone fusion.

Keywords: aneurysmal bone cyst, spine, bone graft, tumor, spine surgery

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INTRODUCTION

Aneurysmal bone cysts (ABC) account for 1 to 6 % of bone tumors with an incidence of 1.4 to 3.2 new cases per 1 million population per year [1-8]. ABC commonly affects flat human bones, pelvis and metaphysis of long bones. Aneurysmal bone cysts of the spine are reported to occur in 8-30 % of cases, they account for 15 % of all tumors and tumor-like diseases of the spine [1, 2, 3, 9, 10]. The pathology is more common in patients aged 12 years and over with a slight predominance in female patients [1, 2, 3, 11]. According to Russian and foreign authors, the lumbar spine is affected in 40-45 % of cases, the cervical spine involved in 30 %, and the thoracic spine impaired in 25-30 % [1, 2, 3, 12]. The search for PubMed publications between 1961 and 2022 using the keywords: aneurysmal bone cyst, spine, children revealed 246 articles reporting treatment of aneurysmal bone cysts of pediatric spine of various localization. Most of the papers were case reports with no treatment approaches discussed in several contributions.

Etiology, pathogenesis. Aneurysmal bone cyst is a benign neoplasm characterized by local bone swelling and cystic cavities limited by fibrous septa and containing bloody fluid with the cellular structure being represented by proliferated fibroblasts with giant cells such as osteoclasts and histiocytes [1, 12, 13, 14]. The etiology of primary aneurysmal bone cyst is associated with upregulation of the ubiquitin-specific protease (USP6) gene in the majority of cases [1, 2, 3, 4, 14, 15]. There are two types of ABC: primary (de novo) accounting for 70 %, and secondary ABC featuring multiple hemorrhages and resulting from cystic restructuring of a giant cell tumor, osteoblastoma, chondroblastoma or fibrous bone dysplasia [1, 16]. Despite the primary benign nature, ABC can be locally aggressive and can develop to Enneking grade III [1, 2, 17] with an exophytic spread tendency. With the tumor localized in the spine, compressive myelopathy is common and necessitates combined treatment.

Clinical manifestations. Pain is the most common symptom in primary tumors of the spine and encountered in 84 % of patients seen for the first time [1, 17]. The duration of symptoms largely depends on the aggressiveness of the tumor, and it can play the role in differential diagnosis. Pathological vertebral fractures and subsequent complications can result from ABC. The patient may have a palpable mass,

with ABC being located in the posterior supporting column. Scoliotic or kyphotic deformity in ABC can be caused by impaired shape of the affected segments and a pronounced pain syndrome leading to antalgic deformities.

The objective of the work was to report the result of surgical treatment of a patient with an aneurysmal bone cyst of the spine.

MATERIAL AND METHODS

The study included the data of an 11-year-old female patient with an aneurysmal bone cyst of the L3 vertebra treated with staged therapeutic punctures as a preoperative preparation followed by an open surgery. The evaluation criteria included regression of the clinical manifestations, absence of tumor

recurrence, stability of the metal construct and formation of bone fusion.

The authors' team obtained consent from the patient and her parents for the publication of anamnestic data, the results of examinations and the treatment, and intraoperative photographs.

RESULTS

An 11-year-old patient first seen in July 2019 presented with pain in the lumbar spine radiating to the left leg, scoliotic curvature of the thoracolumbar spine deviated to the right. The patient sought medical assistance at the place of her residence for impaired posture one month prior to her visit to the Priorov National Medical Research Center of Trauma and Orthopaedics (PNMRC TO). Conservative treatment with massage and physiotherapy was recommended for her. She developed greater pain as a result of the therapy and complained of pain in her leg a month later.

CT and MRI performed during her next visit showed a large pathological formation of the L3 vertebra and a lytic defect at the root of the arch and the body with a fluid level. An aneurysmal bone cyst of the L3 vertebra, spinal canal stenosis and chronic lumbodinia were diagnosed based on imaging findings. Physical examination performed on admission in July 2019 showed that the patient could ambulate independently in the clinical department, without additional support, limping on her left leg. There was swelling visualized in the lumbar spine on the left at the levels of L2, L3 vertebrae being painful on palpation. There were limited movements in the lumbar spine being painful in extreme positions. Right-sided scoliosis of the lumbar spine was identified. Another CT and MRI examinations performed at the PNMRC TO demonstrated a large formation (up to 64 mm in diameter) at L3 occupying the entire left side of the posterior and middle supporting columns (according to F. Denis) and about 10 % of the vertebral body with lateral stenosis of the

spinal canal at the level of the tumor. Delimited cavities with a liquid component were determined in the cyst. The cortical bone was weakly expressed on CT scan (Fig. 1).

A therapeutic and diagnostic puncture was produced at the first stage and the surgical material was used for post-mortem examination to verify the diagnosis. Intraoperative pressure inside the cyst was 342 mm H₂O. A 0.9 % NaCl solution was used to wash the cyst cavity until it was free of blood impurities and then it was treated with an aminocaproic acid solution to reduce the activity of the process. Pathomorphological assessment demonstrated an aneurysmal bone cyst. The patient reported a decreased pain in the lower back next day after the procedure and pain radiating to the left lower limb relieved.

Repeated therapeutic punctures were performed with an interval of 11/2 months. Dark blood of a liquid consistency was obtained during therapeutic punctures. The blood volume obtained during the first puncture was 120 ml decreasing by an average of 15-20 ml with subsequent punctures. The pressure inside the cyst as the main criterion evaluated during therapeutic punctures decreased to 73 mm H₂O during the final manipulation. Pain relief, complete regression of neurological symptoms and decreased cyst activity persisted after the third manipulation. MRI demonstrated decreased stenosis of the spinal canal, number of cavities and the volume of the liquid component at 11/2 months. CT scans showed signs of partially repaired vertebral bone tissue, solidified cortical bone, appearance of osseous septa in ABC) at 11/2 months (Fig. 2).

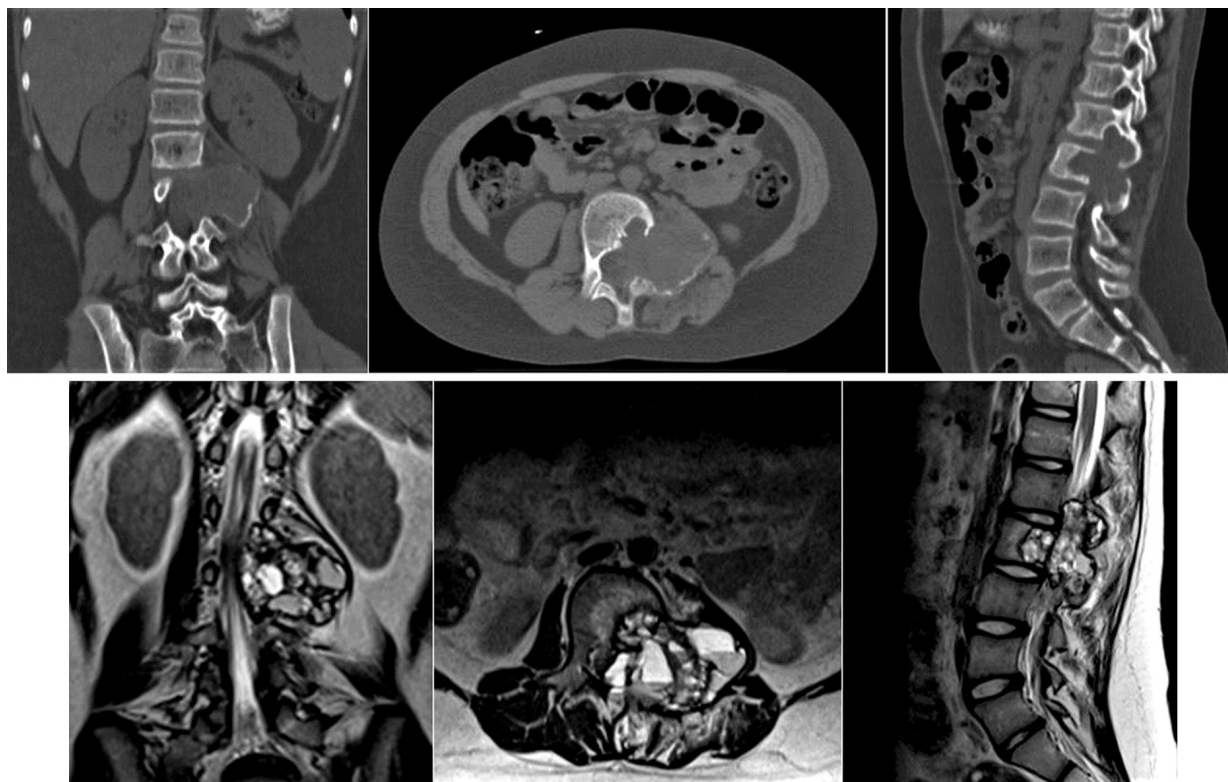


Fig. 1 MRI and CT scans of the lumbar spine on admission

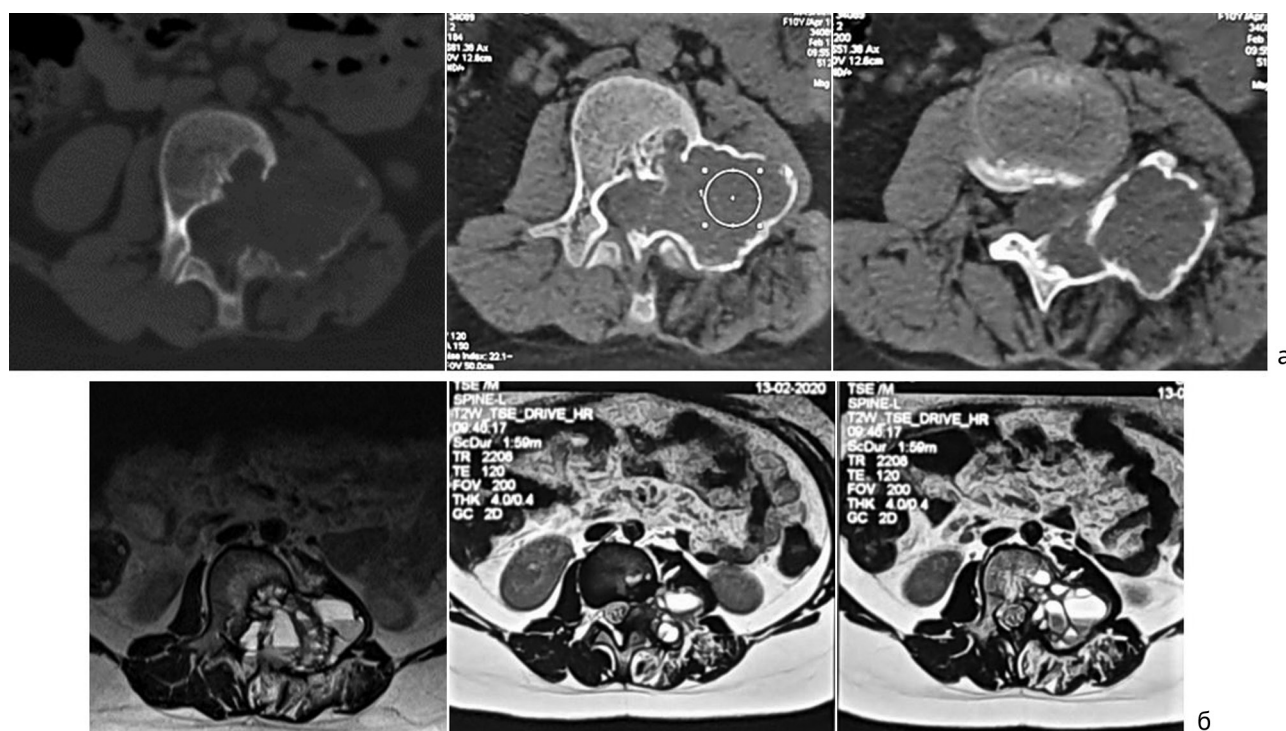


Fig. 2 CT and MRI scans of the cystic area before treatment (a) and after three therapeutic punctures (b)

The second stage included an open surgical intervention of transpedicular fixation at the level of L2, L3, L4 on the right and L2, L4 on the left, L3 hemilaminectomy on the right, removal of aneurysmal cyst membranes, decompression of the spinal canal, combined plasty of the

bone defect using auto- and allografts. The operating time was 2 hours 15 minutes. Intraoperative blood loss was 240 ml (Fig. 3).

The patient was encouraged to ambulate on the 2nd day using a semi-rigid orthosis fixing the lumbar spine.

Case report

The pain completely relieved on the 5th day. No signs of relapse were detected at 11/2-month follow-up. CT examination showed stable transpedicular fixation. The bone fused with the allograft integration at 9 months, and the bone restored at the resection site with the "bone

shape memory" type (Fig. 6). Considering the patient's age and absence of substantial vertebral growth of the column in Risser Stage 4 there were no indications for the removal of the metal construct at the time of the last follow-up.

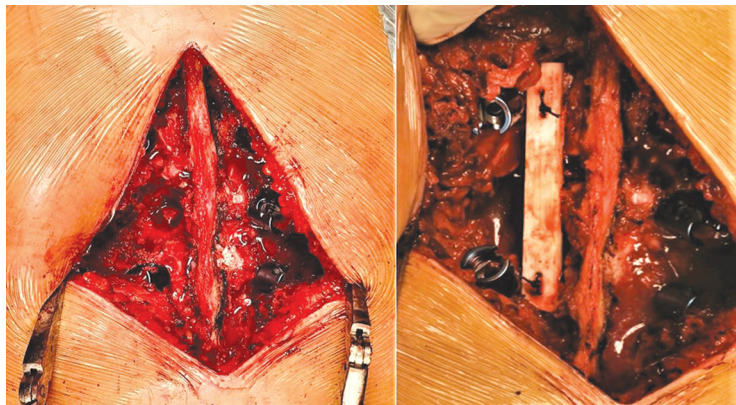


Fig. 3 Surgical stages

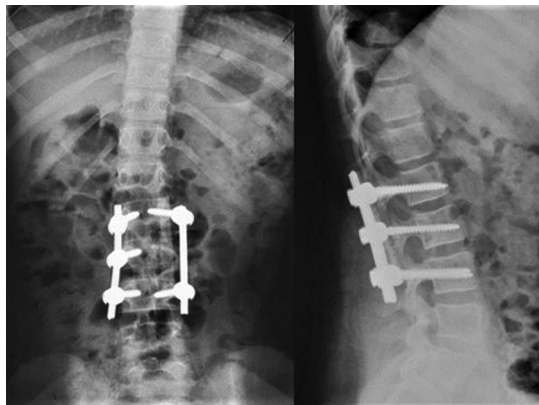


Fig. 4 Two radiographic views of the spine two days after surgery

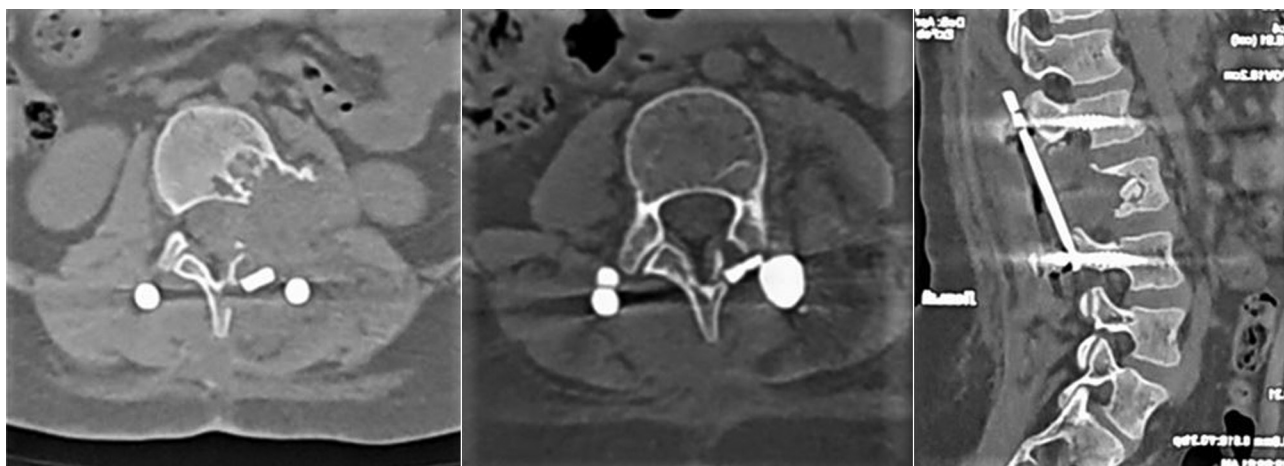


Fig. 5 CT scans performed three days after surgery



Fig. 6 CT scan of the surgical site at 18 months

DISCUSSION

Treatment of ABC of the spine in children and adolescents suggests considerations of many aspects in the course of the disease. The choice of treatment

strategy is at the intersection of three specialties: orthopaedics, oncology, neurosurgery. Surgical orthopaedic treatment would rely on the volume of bone

tissue damage, secondary deformity, if any, the length of metal fixation to be calculated for adequate spine stabilization and the possibility and volume of bone grafting. Neurosurgical consideration would include the involvement of neural structures in the tumor process and stenosis of the spinal canal. Oncology aspects would include considerations of the process staging, conservative reduction of the tumor activity and planning the possibility of ablative removal of the neoplasm. Treatment for ABC includes removal of the tumor with or without bone grafting and metal fixation, embolization, radiation therapy or a combination of the above. [12, 19, 20, 21]. The literature also describes vertebroplasty with bone cement but the method is not commonly used. The choice of the optimal strategy for the treatment of ABC of the spine remains controversial. Marginal resection and debridement of the pathological focus is a surgical priority. Stabilization of the spine using transpedicular fixation has shown high efficiency in modern orthopaedics providing spinal stability and preventing or correcting curvature [19, 22, 23]. Puncture therapeutic and diagnostic treatment can be considered as the first step in management of ABC of the spine. The treatment method facilitates the assessment of the tumor activity by measuring the pressure inside the cyst which

can measure 400-450 mm H₂O at first punctures, and collection of material for histological examination. Cyst puncture ensures relief of the pressure inside the cyst, thereby reducing the intensity of pain and neurological symptoms, if any. A 0.9 % NaCl solution is used to wash the cyst cavity during puncture treatment to be followed by washing with an aminocaproic acid solution or other hemostatic agent. The treatment method can be used for quite a long time with long-term results showing high efficiency in the form of complete closure of the cyst or transition to the repair stage [1, 10, 22]. The case report has shown that puncture treatment allows for an open surgical intervention to be produced with a small intraoperative blood loss due to a decreased cyst activity after staged therapeutic punctures, safer and optimal removal of the tumor membranes due to compacted cyst membranes being demarcated from the spinal canal ad oculus, implantation of metal constructs and combined bone grafting. Each case of an aneurysmal bone cyst requires a differentiated treatment approach, which depends on the volume of the pathological focus, the extent of damage to the supporting columns of the spine, the presence of spinal canal stenosis and neurological symptoms, the activity of the pathological process and a secondary spinal deformity [9, 10, 15].

CONCLUSION

The clinical example presented showed the effectiveness of staged preoperative puncture treatment of an aneurysmal bone cyst followed by open resection of the affected area in combination with transpedicular stabilization and bone grafting. The results demonstrated that puncture treatment of an aneurysmal cyst facilitated a reduced risk of severe bleeding, reduced pressure inside

the cyst stimulating bone repair that resulted in lower intraoperative risks of open surgery and recurrence, and improved postoperative outcome. The lack of a larger sample of patients and a limited follow-up period are limitations for analyzing the effectiveness of the combined technique and substantiating the algorithm for surgical treatment of ABC patients.

Conflict of interest Not declared.

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