

Surgical resection of a giant spine aneurysmal bone cyst in a child

Ressecção cirúrgica de cisto ósseo aneurismático vertebral gigante em criança

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ABSTRACT

Introduction: Aneurysmal bone cyst is a benign and destructive bone lesion of unknown etiology. Current approaches to treatment include surgical en bloc excision, curettage, selective arterial embolization and radiotherapy. A giant aneurysmal bone cyst in the lumbar vertebra is reported, along with a brief review of the literature on the topic. **Method:** A 15-year-old boy presents with lumbar pain and a 3 months history of paraparesis. Diagnostic imaging revealed an expansile lytic lesion in the spinous process and lamina of the fourth and fifth lumbar vertebra. Surgical treatment consisted of excisional biopsy and a segmental instrumented posterior fusion from L2-S1. Histopathology was consistent with an aneurysmal bone cyst. After one year, the patient improved his neurological function with only a L5 bilateral paresis. **Discussion:** Treatment of aneurysmal bone cysts of the spine is challenging when it occurs in close proximity to neural and vascular structures. Fusion and stabilization with instrumentation are often necessary. One vertebra is involved in most cases and extensive lesions are uncommon. Complete tumor resection and fusion of lumbar spine, using stabilization with instrumentation provide both cure and preservation of lumbar spine stability. Neurological deficits can be improved by surgical removal of the tumor

Keywords: aneurysmal bone cysts, surgery, spine, tumors.

RESUMO

Introdução: O Cisto ósseo aneurismático é uma lesão óssea benigna e destrutiva de origem desconhecida. Os tratamentos atuais para esta doença incluem a ressecção em bloco, curetagem, embolização arterial seletiva e radioterapia. Um caso de cisto ósseo aneurismático gigante da coluna lombar é relatado. **Método:** Paciente de 15 anos de idade foi admitido com dor lombar e paraparesia com três meses de duração. Os exames radiológicos demonstraram uma lesão lítica expansiva nos

processos espinhosos e lâminas da quarta e quinta vértebras lombares. A cirurgia consistiu da ressecção e instrumentação posterior com fusão de L2 até S1. A histopatologia demonstrou um cisto ósseo aneurismático. Após um ano de pós-operatório, o paciente melhorou a sua função neurológica, permanecendo apenas com uma paresia de L5 bilateral. **Discussão:** O tratamento do cisto ósseo aneurismático é desafiante, principalmente se próximo a estruturas neurais e vasculares. A fusão óssea e estabilização com instrumentação são frequentemente necessárias após a ressecção radical de tumores vertebrais extensos. Uma vértebra geralmente é acometida pelos cistos ósseos e lesões extensas são incomuns. A ressecção completa do tumor e a fusão da coluna lombar com instrumentação permitiram a cura com preservação da estabilidade espinhal. As complicações neurológicas podem ser melhoradas com a retirada cirúrgica do tumor.

Palavras-chave: cisto ósseo aneurismático, cirurgia, coluna vertebral, tumor.

INTRODUCTION

Aneurysmal bone cyst is a benign but locally destructive lesion. This cyst is generally observed in young subjects and in a spinal localization are only 10% of the all cases¹. The lesions are broadly separated into primary and secondary bone lesions². Up to 50% of lesions are superimposed on a preexisting condition and are classified as secondary aneurysmal bone cysts³. Most of these cases are small lesions and only a one segment of the column is affected³. Pain is commonly the initial symptom and the cause of the diagnosis by radiological exams^{2,6,10}. In the literature, there are only case reports and small series of patients, with description of the incidence and the

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surgical techniques for this challenging spinal pathology^{2,6,10}. Fusion and stabilization with instrumentation have often been avoided in children due to concerns over instability following future spine growth. We report the clinical, radiological, and therapeutic aspects of one giant aneurysmal cyst of the lumbar spine, in a child that was operated on by anterior and posterior approaches. The evolution of the case and the current treatment options are discussed.

CASE REPORT

An 8 years-old boy was admitted to neurosurgical evaluation for pain. His history began five months earlier when he suffered a minor trauma during sports practice. He felt a strong pain and his mother noticed a mass at the lumbar region. The child had a progressive loss of movements at the lower limbs with bladder dysfunction and at the same time lost more than 20 kilos. After two weeks, he was unable to walk and the pain was strong enough to keep the child bedridden. Codein and codein-like analgesics were unable to relieve the pain.

A radiographic evaluation showed a destructive lesion of L4 to S1. CT scan and MRI showed a giant aneurysmal bone cyst with intraspinal extension and compression of neural structures (fig. 1 and 2). There were no other tumor and the diagnosis was a primary aneurysmatic bone cyst. Angiography was performed and only small vessels were visualized going to the lesion: embolization was tried but was not achieved.

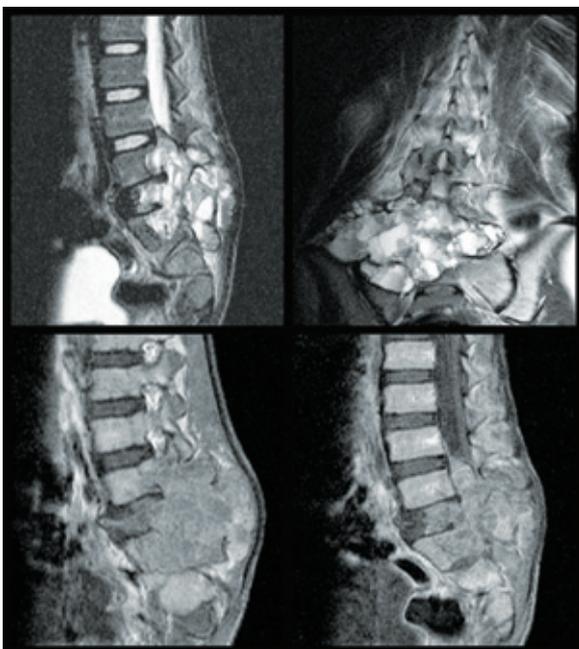


Figure 1. Preoperative MRI showing the extension of the aneurysmal bone cyst

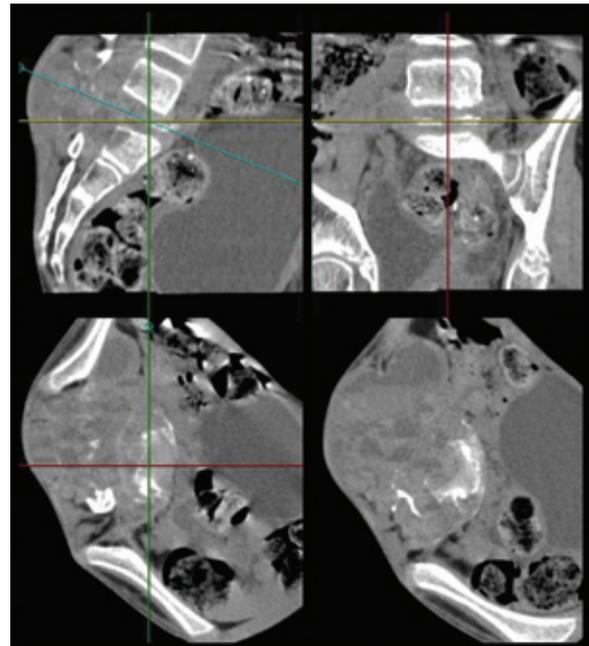


Figure 2. Preoperative CT scan showing the bone destruction.

Surgery was planned to occur in two steps: The first one was an anterior approach with dissection of retroperitoneal space. The spine was localized and the major vessels were dissected out. The lumbar arteries were ligated and an anterior corpectomy at L5, S1 and S2 was performed. Part of the tumor extending outside the spine was resected. One acrylic cage was positioned between L4 to S1 to keep the spine temporarily stable until the next operation. The anterior part of the tumor was excised with extensive intraoperative blood loss (4700ml). The child was kept in bed until his next surgery.

After two weeks, he was submitted to a posterior approach: the rest of the tumor at the posterior vertebral elements and inside the canal was removed. Following excision, transpedicular stabilization of the spinal column was achieved. The acrylic cage was changed by two titanium cages with bone from iliac crest. Transpedicular screws were positioned to fix the spine. This second operation had an intraoperative blood loss superior to 5400ml. Pathologic examination confirmed aneurysmal cyst bone. Total blood loss in the two operations was more than 11000ml.

The patient stayed in hospital for one month and after a rehabilitation program, he was discharged using a thoracolumbar orthosis. He was followed up on an outpatient clinics and after six months he was able to walk without any help and without pain. Strength in the lower limbs improved, with only a S1 paresis, and with full recovery of the neurogenic bladder. Follow up after one year showed total removal of the cyst and complete fusion (fig. 3).

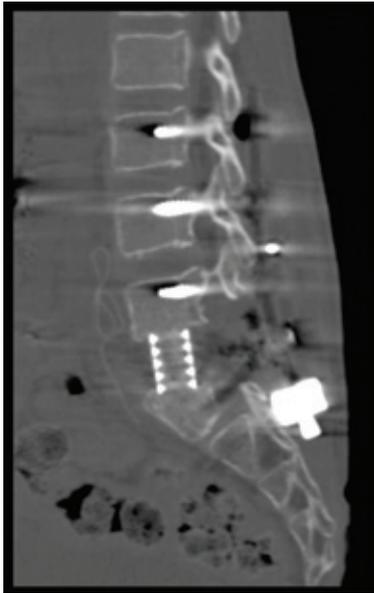


Figure 3. Postoperative CT scan (six months later) shows a complete fusion

DISCUSSION

The term “aneurysmal bone cyst” was first coined by Jaffe and Lichtenstein¹. Benign spine tumors are uncommon if compared to metastasis and multiple myeloma and account for 15% of all primitive tumors of the spine^{2,5}. Aneurysmal bone cyst is a rare spinal disease and occurs usually under the age of twenty, with a slight female predominance, but there are some reports in patients more 50 years-old. Most of the cases are described at the lumbar spine and there are only a few cases in the cervical spine⁵.

Several articles reported on this rare bone lesion, but its pathogenesis remains unclear. The three main hypotheses suggest that the lesion is the result of one of the following: improper repair of a traumatic subperiosteal hemorrhage, a vascular disturbance of the bone, or hemorrhage into a preexisting lesion². These all may make sense, but trauma and vascular occlusion or malformation cannot account for all types of primary aneurysmal bone cyst. The cause appears to be multifactorial¹. Most of the patients are children or young adults, and the initial clinical symptom is pain. Some cases can begin with neurologic deficit if spinal canal or the foramina are early affected. Usually bone lesions are unique but can reach more than one spinal segment⁷.

The diagnosis is done by radiological exams, but CT scan and MRI are the best methods to analyze the spinal cyst, to show

bony walls and liquid-liquid levels suggestive of aneurysmal bone cyst⁹ MRI is better than CT for the evaluation of tumor extension and for the spinal cord compression. Some pictures could be not typical to aneurysmal cyst bone or have one primary bone tumor associated. When CT scan and MRI show characteristic signs of aneurysmal bone cyst, we do not systematically perform a biopsy, if complete removal of the tumor appears possible⁵.

Numerous different treatment options have been used in the management of aneurysmal bone cysts. Surgical treatment can be facilitated by preoperative embolization of the highly-vascular tumor and, in this situation, tumor excision can be achieved without extensive intraoperative blood loss^{4,8}. In the present report, embolization was not achieved, but the first step during the anterior approach was the occlusion of all vessels coming to the cyst from abdominal aorta. This maneuver was very important to keeping blood loss stable during all the surgery. Meyer et al., described one case with a thoracic spine lesion, where the surgical treatment was facilitated by preoperative embolization, with considerable reduction of intraoperative bleeding⁸.

Surgical removal with biopsy is the treatment of choice^{1,2,7}. Reconstruction and stabilization of the spine must be complete using bone graft with or without instrumentation. In case of spinal cord involvement, decompression must be performed rapidly, with the greatest possible tumor removal. Selective arterial embolization can be used as preparation to surgery. Radiotherapy is contraindicated in children, because of the risk of radio-induced tumors. Recurrence is uncommon when complete excision is achieved, with 90% of recurrences occurring within 2 years of treatment³.

The authors conclude that surgery of benign aneurysmal bone cyst constitutes a feasible means of reducing the compression of neural structures despite of intraoperative bleeding. The anterior surgery followed by a posterior approach allowed complete local excision and the reconstructive spinal fixation.

DISCLOSURE

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

REFERENCES

1. Beiner JM, Sastry A, Berchuck M, Grauer JN, Kwon BK, Ratliff JK, et al. An aneurysmal bone cyst in the cervical spine of a 10-year-old girl: a case report. *Spine* 2006, 31:E475-E479.
2. Boriani S, De IF, Campanacci L, Gasbarrini A, Bandiera S, Bigagini R, et al. Aneurysmal bone cyst of the mobile spine: report on 41 cases. *Spine* 2001, 26:27-35.
3. Capanna R, Albisinni U, Picci P, Calderoni P, Campanacci M, Springfield DS: Aneurysmal bone cyst of the spine. *J Bone Joint Surg Am* 1985, 67:527-31.
4. Cory DA, Fritsch SA, Cohen MD, Mail JT, Holden RW, Scott JA, et al. Aneurysmal bone cysts: imaging findings and embolotherapy. *AJR Am J Roentgenol* 1989, 153:369-73.
5. Dekeuwer P, Odent T, Cadilhac C, Journeau P, Langlais J, Padovani JP, et al. [Aneurysmal bone cyst of the spine in children: a 9-year follow-up of 7 cases and review of the literature]. *Rev Chir Orthop Reparatrice Appar Mot* 2003, 89:97-106.
6. DiCaprio MR, Murphy MJ, Camp RL: Aneurysmal bone cyst of the spine with familial incidence. *Spine* 2000, 25:1589-92.
7. Karparov M, Kitov D: Aneurysmal bone cyst of the spine. *Acta Neurochir (Wien)* 1977, 39:101-13.
8. Meyer S, Reinhard H, Graf N, Kramann B, Schneider G: Arterial embolization of a secondary aneurysmatic bone cyst of the thoracic spine prior to surgical excision in a 15-year-old girl. *Eur J Radiol* 2002, 43:79-81.
9. Pennekamp W, Peters S, Schinkel C, Kuhnen C, Nicolas V, Muhr G, et al. Aneurysmal bone cyst of the cervical spine (2008:7b). *Eur Radiol* 2008, 18:2356-60.
10. Perlmutter DH, Campbell S, Rubery PT, Vates EG, Silberstein HJ: Aneurysmal bone cyst: surgical management in the pediatric cervical spine. *Spine* 2009, 34:E50-E53.
11. Tatter SB, Borges LF, Louis DN: Central neurocytomas of the cervical spinal cord. Report of two cases. *J Neurosurg* 1994, 81:288-93.

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