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TECHNICAL NOTE

Successful NBCA embolization of a T2 aneurysmal bone cyst

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Surgically accessible aneurysmal bone cysts (ABC) have traditionally been treated with curettage. Selective arterial embolization was initially proposed as a preoperative adjuvant to reduce peroperative bleeding. Currently, the role of embolization has been extended to the definitive treatment of aneurysmal bone cyst of the spine in children, as well as to other locations in the skeleton. The authors describe the technique in a 15-year-old girl with a T2 aneurysmal bone cyst. Digital subtraction angiography was performed for tumor vascular mapping, followed by selective arterial embolization with N-butyl 2 cyanoacrylate (NBCA). Because of persistent local pain, repeat embolization was done at 8 months. Pain relief and progressive ossification of the lesion were now observed. At 4-year follow-up, the patient was asymptomatic, with complete ossification of the lesion. Selective arterial embolization (SAE) is a minimally invasive, safe and effective procedure for the permanent occlusion of the pathological feeding vessels of spinal ABC. It should be considered as the treatment of choice for lesions difficult to access with surgery, especially in young patients. Careful pre-embolization vascular mapping of the lesion, operator's experience and use of NBCA are the keys to success.

Keywords : aneurysmal bone cyst ; spine ; selective embolization ; NBCA.

INTRODUCTION

Aneurysmal bone cyst (ABC) is a lytic hemorrhagic benign lesion, which represents 1% of all primary bone tumors (*6*). The term aneurysmal bone cyst was initially introduced by Jaffe and Lichtenstein (*17,21*). ABC is classified as primary if there are no associated lesions, and secondary when associated with other bone tumors, such as giant cell tumor, chondroblastoma, osteoblastoma or fibrous dysplasia (*20*). ABC typically occurs in the first two decades of life. Any bone may be affected, the most common sites being the pelvis, the spine and the metadiaphyses of the long bones. The spine may be affected in 6% to 22% of the cases (*10,24*).

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According to Enneking's staging system (*13*), ABC may range from stage 1 (latent) to 3 (aggressive). Progression may be very rapid ; spontaneous regression has been reported after a fracture or an open biopsy (*6*).

Traditional treatment for ABC is curettage of surgically accessible lesions (*2,6*). Selective arterial embolization (SAE) was initially proposed as a preoperative adjuvant to reduce peroperative bleeding. Currently, the role of SAE has been extended to the definitive treatment of ABC of the spine in children (*4,25*), as well as for other locations in the skeleton (*33*). This article presents an adolescent with a T2 ABC, successfully treated by SAE with N-butyl 2 cyanoacrylate (NBCA). It emphasizes the emerging therapeutic indications of spinal ABC.

HISTORY

A 15-year-old girl presented to the authors' institution with a 6-month history of swelling at the base of the neck. She reported a minor local trauma some months before. Physical examination showed a well-defined tender soft-tissue mass at the cervicothoracic junction, without any local inflammation or neurological involvement of the extremities. Radiographs showed a T2 lesion (Fig. 1a). Computed tomography (CT) demonstrated a large osteolytic lesion, originating from the pedicles, laminae and spinous process of T2, and extending to C4 and T5 (Fig. 1b-d). Magnetic resonance (MR) imaging showed a multicystic mass with fluid-fluid levels. CT-guided biopsy was performed ; histological examination of the biopsy specimen confirmed the diagnosis of ABC. Given the young age of the patient and the location of the lesion, selective arterial embolization was recommended.

The femoral artery was catheterized under general anesthesia, using the Seldinger technique. Digital subtraction angiography (DSA) of the spine showed the pathological feeding vessels and the vasculature of the cyst itself (Fig. 2). The feeding vessels were catheterized with a 4 French diagnostic catheter and a microcatheter. Then the embolic agent NBCA, mixed with a 33% solution of lipiodol ultrafluid, was injected, sandwiched with a 5% glucosate solution to prevent polymerization with blood. Three feeding vessels were selectively embolized. Control angiography showed complete occlusion of the feeding vessels. Patient's recovery was uneventful.

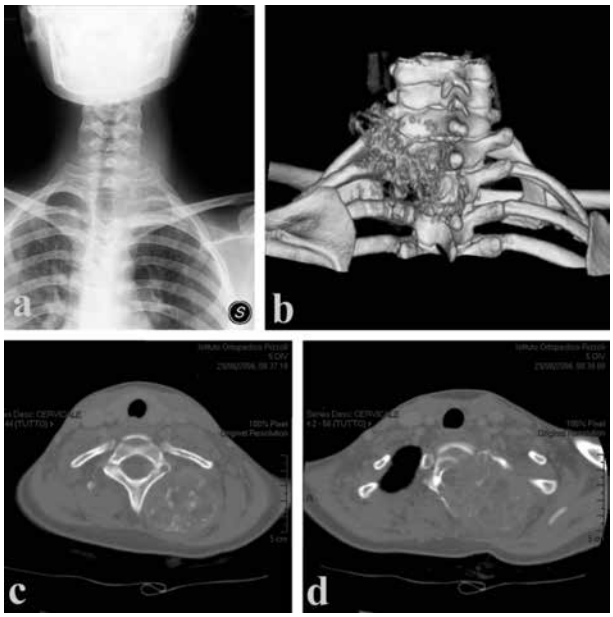

Fig. 1. — (a) AP radiograph. (b) CT three-dimensional reconstruction. (c) and (d) osteolytic expansile lesion involving pedicles, laminae, and spinous process of T2, extending to the paravertebral area C4-T5.

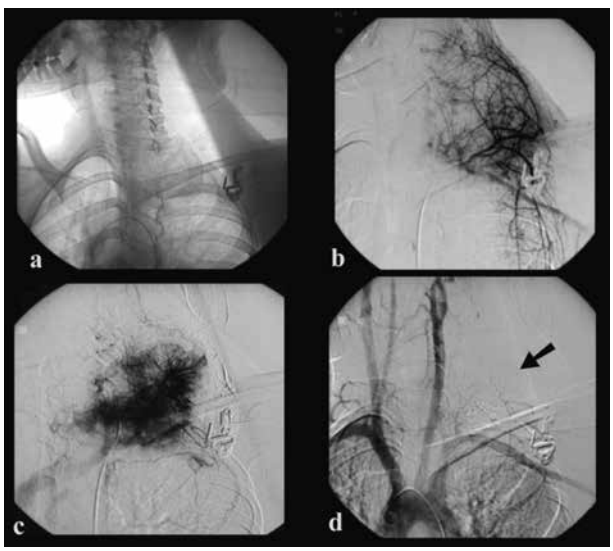

Fig. 2. —First embolization. (a) transfemoral catheterization. (b) and (c) selective catheterization and NBCA embolization. (d) post-embolization angiography shows complete occlusion of the pathological vessels (*arrow*).

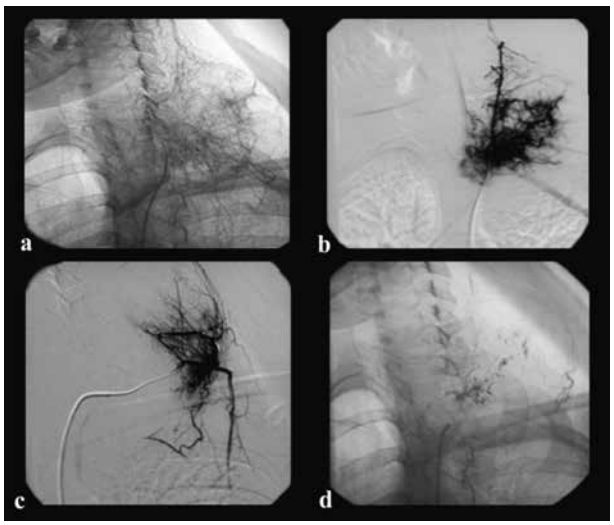

Fig. 3. — Second embolization. (a) persistent vascularization. (b) and (c) selective catheterization and NBCA embolization of two pathological feeding vessels. (d) post-embolization angiography shows complete occlusion.

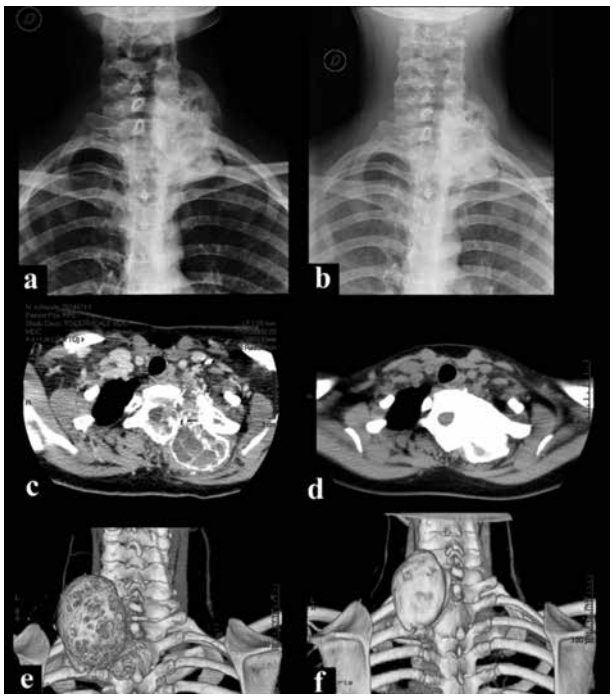

Fig. 4. — (a) and (b) plain radiographs, 1 and 2 years post-embolization : progressive ossification. (c) and (d) axial CT at 1 and 4 years : ossification. (e) and (f) three-dimensional CT at 1 and 4 years : ossification.

At follow-up the patient experienced mild local pain. Because of continuous pain, repeat angiography was performed 8 months after the initial procedure. It showed persistent pathological vascularization of the lesion, and two feeding vessels were embolized (Fig. 3). The pain disappeared completely. Radiographs showed progressive homogeneous trabecular bone formation. Four years after embolization, the patient was asymptomatic ; imaging showed complete ossification of the lesion without evidence of local recurrence (Fig. 4).

DISCUSSION

Spinal ABC is not common (*6*). The lumbar spine is most frequently affected, followed by the thoracic and cervical spine and the sacrum. Although benign, ABC can be locally expansive and destructive ; it can result in pathological spinal fractures and neurological complications (*5,6,7,8,15,25*). Early symptoms are pain and swelling ; the mean duration of symptoms prior to diagnosis is 2 years. Occasionally, imaging may show vertebra plana (*8*), acute vertebral collapse (*15*), rarely scoliosis or kyphosis (*25*), or compression of the spinal cord (*5,7*). CT and MRI show an expansive lytic lesion, and fluid-fluid levels (*19,23,29*). The differential diagnosis includes osteoblastoma, simple bone cyst, giant cell tumor, Ewing's sarcoma and telangiectatic osteosarcoma (*20,23*).

The optimal treatment of spinal ABC is controversial. Traditional treatment included curettage with or without bone grafting, or en bloc excision, often associated with peroperative hemorrhage, post-laminectomy kyphosis and instability (*3,20, 31,37*). Therefore, SAE has been recommended for lesions with definite diagnosis of ABC in patients without neurological deficit and/or instability (*4*). In contrast, curettage has been recommended for patients with pathological fracture or vertebral body location and neurological deficits, while en bloc excision with or without reconstruction has been recommended for ABC in the posterior spinal elements (*4*).

Initially, preoperative SAE has been used as a surgical adjuvant procedure to reduce peroperative bleeding. However, with the improvement of endovascular techniques, the role of SAE has rapidly been extended to the definitive treatment of ABC of

Table I. — Studies about tumors treated with NBCA (N-butyl 2 cyanoacrylate) embolization

Author	Age/gender	Tumor/location	Treatment	Follow-up	Results
Syal	20/m	Hemangioma Mandible, pelvis, skull	Embolization + 5 weeks of radiotherapy	6 months progression	Cessation of bleeding and
Schirmer	49-66/ 2m/3f	Renal cell carcinoma metastasized to spine	Embolization + decompression	8 months	Reduction of peroperative bleeding
Liebig	68/f	Colon cancer metastasized to spine	Embolization + excision	Not available	Reduction of peroperative bleeding
Cotton	55-71 2m/2f	Hemangioma Spine	Embolization + percutaneous injection of methyl-methacrylate and NBCA + excision	4 months bleeding	Reduction of peroperative
Kaneko	10/f	Hemangioma Mandible	Intralesional injection	3 years	Local control
Mindea	17/f	Recurrent giant cell tumor cervical spine	Embolization + resection	2 years	Reduction of vascular supply. Control of disease
Rossi	3.3-60.8 20m/16f	Aneurysmal bone cysts Spine and skeleton	Embolization	0.9-5 years	Local control
Marushima	12/m	Aneurysmal bone cyst T9	Embolization	3 years	Local control
Authors' case	15/f	Aneurysmal bone cyst T2	Embolization	6 years	Local control

the spine (*1,12,14,36*) and of ABC in other sites (*7*). The advantages are : less invasiveness, lower cost, easiness, and repeatability, compared to surgery (*4,33*). The available embolic agents are : 1. coils (*12*), which are more appropriate for preoperative embolization of ABC, 2. particles such as polyvinyl alcohol (PVA), and 3. liquid materials such as NBCA used in the current study (*33*). The advantages of NBCA are radiopacity with lipiodol, and permanent occlusion of the feeding vessels, in other words a lower risk of recanalization (*26,30*). Few studies have reported on SAE with NBCA for bone tumors (Table I) (*9,18,22,25,27,33,34,35*). De Cristofaro *et al (11)* and Mohit *et al* (*28*) reported ossification and reduction in size of ABC, 6 to 24 months after SAE with PVA. In the current study progressive ossification of the lesion was observed from the first year after embolization ; complete ossification without evidence of local recurrence was observed at the 4-year follow-up. A possible complication of SAE is spinal infarction due to anastomosis of pathological tumor vessels with the artery of Adamkiewicz in the thoracic spine or with the vertebral arteries in the cervical spine (*32*). Careful vascular mapping with pre-embolization angiography is paramount to avoid such complications (*16, 28,32*).

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