Percutaneous treatment of symptomatic aneurysmal bone cyst of L5 by percutaneous injection of osteoconductive material (Cerament)

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SUMMARY

We present a case report of a 33-year-old woman with back pain for several months which was resistant to medical treatment. Thoracolumbar MRI and multidetector CT showed an aneurysmal bone cyst intersecting the body and pedicles of L5. Minimally invasive treatment was performed with percutaneous injection of osteoconductive cement (Cerament) to induce sclerosis and bone remodeling of the bone cyst lesion with an analgesic effect. Before treatment, spinal angiography was performed to exclude arterial afferents. No bone biopsy was done. Under general anesthesia and fluoroscopic guidance, a first vertebroplasty was performed by a bilateral transpedicular approach using the osteoconductive cement followed 2 months later by a second treatment with CT-fluoro-guided direct injection of Cerament. No complications occurred during the procedure. At 4 and 6 months follow-up the MRI/CT showed sclerotic bone remodeling of the walls of the aneurysmal cyst with clinical improvement.

deformity, disruption of growth plates and, rarely, neurologic symptoms (depending on the location) or pathologic fracture.^{3,4}

The treatment of spinal ABCs is controversial. If symptomatic, the gold standard treatment is open curettage which carries a risk of surgical complications (intraoperative hemorrhage or postoperative hematoma, infection) and a local recurrence rate of 20–30% complicated by kyphoscoliosis and compression of the spinal cord and nerve roots.⁵ Other strategies have included radiation therapy, endovascular treatment and sclerotherapy. Direct intralesional injection of a sclerotic agent is a therapeutic option, sometimes combined with surgical or endovascular treatment, especially for large and resistant lesions.⁶

This report describes the minimally invasive treatment of ABC with percutaneous injection of osteoconductive cement (Cerament) in order to induce bone sclerosis and remodeling of the cyst with a good analgesic effect, without any major or minor complications.

BACKGROUND

We describe a case report of the mini-invasive percutaneous treatment of a symptomatic aneurysmal bone cyst (ABC) by injection of osteoconductive cement (Cerament) under CT or fluoro guidance. The procedure resulted in complete pain resolution and sclerotic bone remodeling of the walls of the aneurysmal cyst with periosteal new bone formation circumferentially followed by gradual recalcification within the cystic cavity.

CASE PRESENTATION

The ABC is a benign lesion involving virtually any type of bone, most often the tibia (24.7%) or femur (17.3%) followed by the upper extremities (10%) and the pelvis (9%).¹ About 14% of all ABCs occur in the spine, with those in the cervical spine making up only 2%. Predominantly affecting children (60% of patients aged <20 years) and with a preponderance for females, its etiology is still unknown.² The ABC is a typical pseudotumoral hyperemic-hemorrhagic expansive osteolytic lesion with a thin wall containing blood-filled cystic cavities with typical imaging patterns. Although benign and generally asymptomatic, the ABC can be locally aggressive causing extensive weakening of the bony structure and impinging on the surrounding tissues. However, malignant transformation is extremely rare and most are asymptomatic. They are occasionally found during routine examination but sometimes, due to their expansive nature, they can cause pain which is resistant to continuous medical treatment, swelling,

TREATMENT

A 33-year-old woman with back pain resistant to medical treatment for several months was admitted to our neuroradiology service. The thoracolumbar MRI (sagittal T1W, T2W, STIR, T1W and axial and sagittal T1W fat-saturated with contrast element) integrated with multidetector CT multiplanar reconstructions showed a typical ABC intersecting the body and pedicles of L5 (figures 1A–F and 2A–D). Because there was no neurologic compromise and no spine instability, surgical excision and associated spine fixation was deemed unnecessary. The more traditional interventional radiology procedure of particle embolization was also excluded because no arterial feeders were present. After orthopedic and neurosurgical consultations, a minimally invasive percutaneous treatment using an osteoconductive cement injection was proposed in order to obtain an analgesic effect and for vertebral compression fracture by cementoplasty, adding a sclerotic and bone remodeling effect of the bone lesion.

Under general anesthesia and fluoroscopic guidance, a first cementoplasty was performed by interventional neuroradiologists using the transpedicular bilateral approach. The osteoconductive cement (10 mL) was distributed into the vascular cysts using a 16G needle (figure 3A–C). No bone biopsy was performed. A visual analogue scale (VAS) and the Oswestry Disability Index (ODI) were evaluated before and 48 h and 2–6 months after treatment. There were no complications during the procedure (figures 4A–D and 5A–E). The absorbed radiation

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Figure 1 A 33-year-old woman with back pain for several months, resistant to medical treatment. Sagittal T2W (A), T1W (B), STIR (C) and axial T2W (D) MRI showed multiple cystic spaces involving the L5 body containing fluid-fluid levels due to blood products. Sagittal (E) and axial (F) T1W with contrast element showed enhancement at the periphery and septae between cysts.

Figure 2 Axial and sagittal multi-detector CT with multiplanar reconstruction of soft tissue (A, B) and bone windows (C, D) showing balloon-like expansile remodeling extending into the vertebra body of L5 with focal cortical destruction and fluid-fluid levels caused by hemorrhage and blood product sedimentation.

Figure 3 (A) Lateral fluoroscopy control of the patient in prone position showing the needles positioned in the L5 body by a bipeduncular approach. (B) Lateral and (C) posteroanterior fluoroscopy control showing distribution of Cerement in the L5 body without leakage.

dose during the first procedure with fluoroscopy guidance was 618.2 mGy for a total of 5.1 min of fluoroscopy time; while during the CT procedure was 4148 mGy-cm.

OUTCOME AND FOLLOW-UP

Symptoms improved within 4 months after the procedure with a good analgesic effect. The pretreatment VAS score was 4 points, while the ODI score was 26 points. At 4 and 6 months follow-up the MRI and CT controls showed partial sclerotic bone remodeling of the walls of the aneurysmal cyst with complete clinical improvement of the patient (4-point reduction in VAS evaluation and ODI decreased to 0; figures 6A–D and 7A–E). The osseous response was periosteal new bone formation circumferentially followed by gradual recalcification within the cystic cavity (figure 8A–D).

DISCUSSION

ABC is a pseudotumoral hyperemic-hemorrhagic expansive osteolytic bone lesion that most often affects individuals during

their second decade of life and may occur in any bone in the body. ABCs consist of blood-filled spaces separated by fibrous tissue containing multinucleated giant cells. This finding is demonstrated by CT and MRI by separation of the blood content forming levels of different fluid densities. However, the fluid-fluid signal is not exclusive to ABCs and can be seen in other bone lesions such as chondroblastoma, giant cell tumor and osteoblastoma. Histologically, the cyst appears like a ‘blood-soaked sponge’ containing blood with walls consisting of histiocytes, giant multinucleated cells, thin capillary vessels, granules of hemosiderin and immature bone trabeculae. The septations of the lesion are hypervascularized, and venous injection of contrast media on CT or MRI can help to differentiate the thin smooth septa of ABCs from other tumors which have nodularities. Selective angiography can sometimes show the blood supply to these vascular lesions and may occasionally reveal arteriovenous shunts.^{4–6}

Treatment remains controversial, especially because of the benign and asymptomatic nature of these lesions. However, they

Figure 4 (A–D) Follow-up CT control at 2 months showing a thin subcortical sclerotic layer around the cyst walls.

Figure 5 (A–C) Under CT-fluoro control with the patient in the prone position, Cerament was injected directly into the aneurysmal cysts. (D) Sagittal and (E) axial multidetector CT with multiplanar reconstructions after treatment showing distribution of the cement in the bone cysts.

can become symptomatic, ranging from pain resistant to medical treatment to swelling or onset of neurological problems due to weakening and destruction of bone, especially when the spine is involved.

Surgical treatment is considered to be the first choice and includes curettage and wide resection with intralesional excision or 'en bloc' excision and spinal fixation, but it is complicated by the risk of significant blood loss, postoperative spinal deformity, axial deformity, postoperative hemorrhage or postoperative recurrence which occurs in 20–50% of cases.^{6–8} Radiotherapy, which was used in the past, is contraindicated because of the risk of sarcoma induction, gonadal damage and growth plate disruption.⁶ Percutaneous intralesional injections offer the least invasive initial therapeutic option, with more invasive surgical or endovascular strategies reserved for resistant lesions. If arterial feeders are present, superselective embolization by a sclerotic agent (eg, polidocanol, Ethibloc, Glubran, Onyx) with or without surgical excision has been reported to achieve an analgesic effect in up to 80% of patients.^{2,7–9}

In our case report, we have chosen to use a bioactive, osteoconductive element – Cerament–already used for the treatment of vertebral compression fracture with vertebroplasty in Europe. It has the CE mark but it hasn't the FDA approval yet. The rationale of this choice was the young age of the patient and to

obtain an antalgic effect combined with the bioactive propriety of the cement to lead an remodelling bone response. Blood lacunar cysts are lower resistance bone zones accompanied by pain syndrome and a higher incidence of vertebral fractures. Cerament consists of resorbable calcium sulfate (60%) and hydroxyapatite (40%) with osteoconductive properties; the hydroxyapatite acts as a slow or non-absorbable framework which slows down the absorption rate of calcium sulfate and, at the same time, acts as an osteoconductive template for new bone ingrowths.¹⁰ Unlike the PMMA that we generally use in elderly patients affected by porotic or neoplastic VCF to stabilize the trabecular micro-movements of the fractured vertebra with pain relief effect, the action of the new bone-substitute Cerament lies in specific biomechanical and biological properties to support the spinal column associated to antalgic effect. Using pmma, the patient does not have type of osteointegration of the injected material. Considering the age of the patient, the target was to obtain a stability process thanks to Cerament. Once deposited into a load-bearing environment, it is able to withstand cyclic and static complex loading patterns. It has the ability to be resorbed at a rate equal to new bone ingrowth, achieving complete bone remodeling and healing while being able to tolerate the motion/load stresses usually borne by the spine without changing the spinal biomechanics. The

Figure 6 Sagittal T2W (A), T1W (B), axial T2W (C) and axial STIR (D) showing the sclerotic effect of bone cysts at 2 months follow-up.

Figure 7 Axial (A, B), coronal (C) and sagittal (D, E) multidetector CT with multiplanar reconstructions showing sclerotic bone remodeling of the walls of the aneurysmal cyst with periosteal new bone formation circumferentially followed by gradual recalcification within the cystic cavity.

Figure 8 Sagittal (A–C) and axial (D) multidetector CT with multiplanar reconstructions showing sclerotic bone remodeling of the aneurysmal cyst with periosteal new bone formation circumferentially followed by recalcification within the cystic cavity at 6 months follow-up.

mechanical properties, the low stiffness of the device and the bone remodeling processes decrease the shear stresses at the border between the bone and the cement. Compared to Cerament, the PMMA has not bioactive properties, it is inert and non-absorbable cement and it produces only an antalgic effect with good outcome without guaranteeing or combining the bone sclerotic reaction as reported by Guarnieri *et al.*⁶ Contrary to other case reports in which authors used a direct intralesional injection of calcitonin and methylprednisolone or demineralized bone matrix mixed with bone marrow concentrate in order to obtain complete sclerosis and ossification of the cyst over a period of 6 months before surgery, we obtained a clinical improvement within 4 weeks with periosteal new bone formation circumferentially followed by gradual recalcification of the cystic lesion cavity.^{7 11}

Clayer performed aspiration and injection with calcium sulfate in 15 patients unsuccessfully treated by mini-invasive methylprednisolone injection. The calcium sulfate cement was reabsorbed completely within 8 weeks, with periosteal new bone formation circumferentially followed by gradual opacification of the cystic cavity. Complete relief of symptoms at 4 weeks was reported by all patients.¹²

Learning points

- ▶ Aneurysmal bone cysts are benign pseudotumoral hyperemic-hemorrhagic expansive osteolytic bone lesions.
- ▶ Management remains controversial, especially because of their benign and asymptomatic nature.
- ▶ Surgical treatment is considered to be the first choice with a high rate of complications.
- ▶ Mini-invasive percutaneous injection of osteoconductive cement into cyst spaces is possible obtaining an antalgic effect combined with the bioactive propriety of the cement to lead an remodelling bone response like for the vertebroplasty of vertebral compression fracture.

CONCLUSIONS

Percutaneous treatment of spinal ABCs with osteoconductive cement offers a relatively safe early clinical and radiological response and a low complication rate for the primary treatment of ABC.

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