

ANEURYSMAL BONE CYST AT C2

Imaging evaluation after intralesional injection of calcitonin and methylprednisolone

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Abstract – Objective: To demonstrate imaging findings during the follow-up of patients with aneurysmal bone cyst (ABC) of C2 treated with intralesional injection of calcitonin and methylprednisolone. **Method:** Three patients with ABC of C2 were treated percutaneously with intralesional injection of calcitonin and methylprednisolone. All the patients were females, with ages varying from 6 to 13 years. All of them presented with cervical masses, but without neurological symptoms. **Results:** Imaging follow-up with CT and plain films showed progressive ossification and reduction of the blood-filled cavities with volume reduction of all lesions. No recurrence was noted during the follow-up. **Conclusion:** Intralesional injection of calcitonin and methylprednisolone is a safe treatment option for cervical ABC. The CT scan is especially valuable as a guide of injection site and for the follow-up of ossification of the treated ABCs.

KEY WORDS: aneurysmal bone cyst, calcitonin, methylprednisolone.

Cisto ósseo aneurismático em C2: avaliação por imagem após injeção intralesional de calcitonina e metilprednisolona

Resumo – Objetivo: Demonstrar os aspectos de imagem no acompanhamento de pacientes submetidos ao tratamento de cisto ósseo aneurismático com injeção intralesional de calcitonina e metilprednisolona. **Método:** Três pacientes com cisto ósseo aneurismático em C2 foram tratados com injeção intralesional percutânea de calcitonina e metilprednisolona. Os três pacientes eram do sexo feminino com idades variando de 6 a 13 anos. Os três pacientes apresentavam massa cervical sem sintomas neurológicos. **Resultados:** O acompanhamento por imagem com tomografia computadorizada e radiografia simples demonstrou ossificação progressiva e redução das cavidades preenchidas por sangue, com redução do volume das lesões. Não foi percebida recidiva durante o acompanhamento. **Conclusão:** A injeção intralesional de calcitonina e metilprednisolona é uma opção de tratamento segura para o cisto ósseo aneurismático cervical. A tomografia computadorizada é especialmente útil para orientar o sítio da punção e para o acompanhamento da ossificação dos cistos ósseos aneurismáticos tratados.

PALAVRAS-CHAVE: cisto ósseo aneurismático, calcitonina, metilprednisolona.

Aneurysmal bone cysts (ABC) are relatively rare lesions that occur more commonly among patients in the first and second decades, especially in females. Characteristically, the ABC has many cavities full of blood. Most of the lesions occur at long bones, vertebrae and flat bones. The cervical vertebrae involvement is uncommon^{1,2}. When seen in this location, the lesion involves initially the posterior arch and pedicles, with bone destruction and cor-

tical expansion. Occasionally, the ABCs compress the spinal cord and nerve roots.

Unlike many surgical treatments performed to deal with vertebral ABC percutaneous intralesional injection of calcitonin and methylprednisolone is a less aggressive but effective approach³⁻⁶. This technique stimulates the cyst ossification, with new cancellous bone formation and reducing angiogenesis⁷.

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We present the imaging follow-up of three patients with ABC in the posterior arch of C2, who were treated with percutaneous intralesional injection of calcitonin and methylprednisolone.

METHOD

This study evaluated a small series of patients with ABCs in the posterior arch of C2, who were treated with intralesional injection of calcitonin and methylprednisolone. All the patients signed informed consent and the Institutional Review Board of the Hospital approved the study. The patients underwent clinical and imaging investigation, including plain films, CT scans and also magnetic resonance imaging (MRI) in one case. The histological diagnosis of ABC was defined after surgical biopsy. The intralesional injection of calcitonin and methylprednisolone was performed between 2 and 6 times (mean of 4 times), with doses of 200 UI and 120–160 mg, respectively. The follow-up was

done with clinical and imaging examinations, ranging from 2 to 18 months (mean of 7.7 months).

RESULTS

Case 1

A six-year-old female patient was referred to our institution with diagnosis of ABC defined in another hospital. She had the first medical consultation 3 years before, and the main complain was reduced cervical mobility, which was treated initially as torticollis, without any radiological study. The symptoms did not improve, and when she was 4-years old a cervical spine CT scan showed a lytic expansible lesion involving the body and the posterior arch of C2 (Fig 1A), with fluid-fluid level content. This finding was also demonstrated in the MRI as hyperintense and isointense fluid-fluid levels. The plain film showed the “bubble” aspect of the lesion involving the vertebral

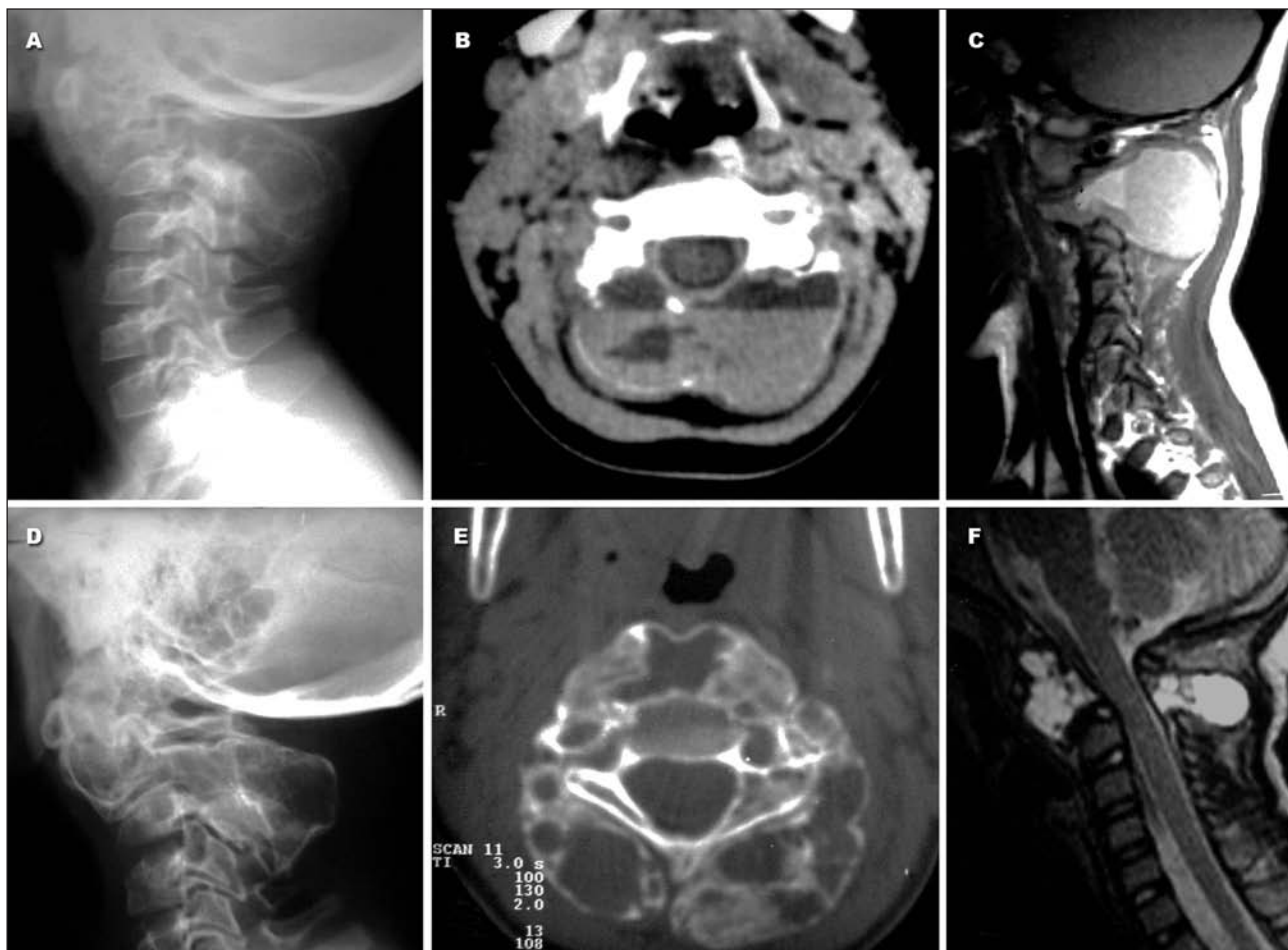


Fig 1 (Case 1). (A) The plain film of the cervical spine shows the “bubble” aspect of an ABC involving the vertebral body and the posterior arch of C2. Notice that the outer cortex is barely seen due the eggshell thin characteristic. (B) CT image at the level of C2 shows an expansible lytic lesion of the posterior arch with fluid-fluid levels. (C) The sagittal T1-weighted MRI shows the hyperintense fluid-fluid level inside the expanded cavity. (D) The follow-up plain film performed one year later after the infiltration with methylprednisolone and calcitonin shows mild shrinkage of the lesion and the thickening of the bone wall. (E) The follow-up CT scan shows the intense ossification of the lesion and reduction of cavities filled with blood. (F) The sagittal T2-weighted MRI shows shrinking hyperintense lesion and the sparing of the vertebral canal.

body and the posterior arch of C2 (Fig 1B). The plain film and the MRI performed at our institution one year later showed significant reduction of the lesion size, although remaining with areas of high-signal on T2-weighted images (Fig 1C). Subsequently, a percutaneous infiltration of 200 IU of calcitonin and 120 mg of methylprednisolone were infused in the lesion. The injection was performed inside the operation room and under general anesthesia to prevent patient movements during the procedure. She completed 4 sessions within 6 months and underwent the follow-up CT that showed intense ossification of the le-

sion with lesser cavities filled with blood. She abandoned the treatment for 18 months, returning two years later, when she received another infiltration. The follow-up CT showed that the lesion was completely ossified (Fig 1D). MRI showed shrinking hyperintense lesion and sparing of the vertebral canal (Fig 1F).

Case 2

A 13-year-old girl presented with complains of cervical spine deformity. The mother noticed the lesion one year before while bathing her. The cervical radiographs showed

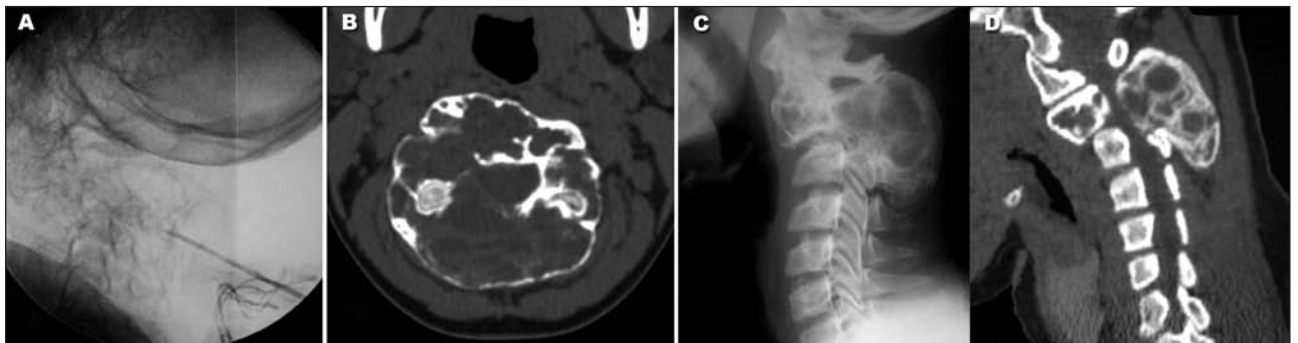


Fig 2 (Case 2). (A) Plain film of the cervical spine showing the needle inside the lesion that is involving the posterior arch of C2. (B) The CT scan demonstrates the expansive lytic lesion involving the body and posterior arch of C2, which presents fluid-fluid levels. The plain film (C) and sagittal CT scan (D) performed one year later show the shrinkage of the lesion and the ossification of its walls. Notice the reduction of size of the cavities.

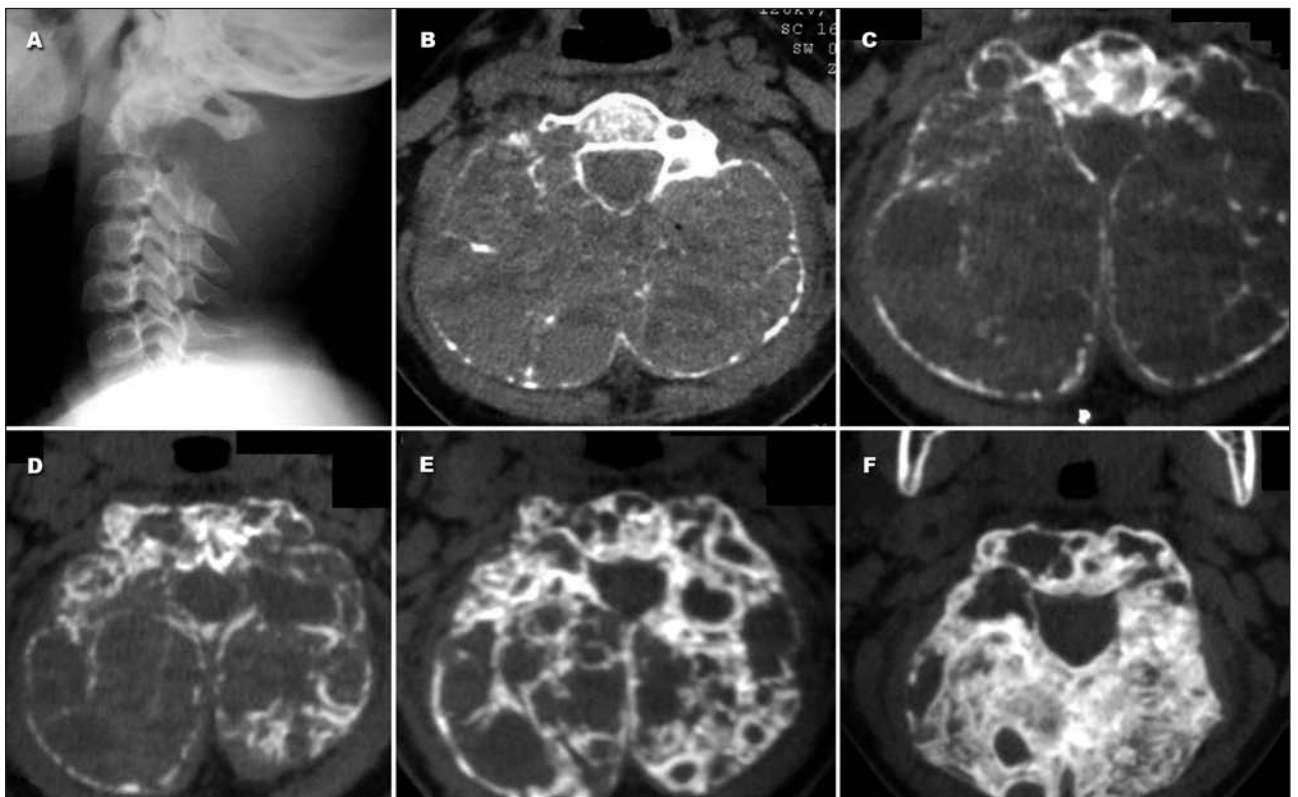


Fig 3 (Case 3). Plain film (A) of the cervical spine shows a soft-tissue mass at the topography of the C2 posterior arch, which is not seen. The CT scan (B) shows the egg-shell-thin cortex and the fluid-fluid levels. Notice the sparing of the cervical canal. The CT scans during the one year long follow-up (C-F) show the progressive thickening of the outer cortex and concentric occupation of the cavities with bone until almost complete ossification of the lesion.

an expansible lytic bone lesion at the posterior arch of C2. The CT scan corroborated the findings and demonstrated the characteristic fluid-fluid levels inside the lesion. She underwent a surgical biopsy that confirmed the diagnosis of aneurysmal bone cyst. At the same time, the first intralesional infiltration of 200 IU of calcitonin and 160 mg of methylprednisolone was performed. Two months later another infiltration was done, and the plain films and the CT showed that the cyst ossified intensely, with volume reduction (Fig 2).

Case 3

An 11-year-old girl present with a 5-month history of cervical pain, associated with growing volume in the posterior region of the neck. The cervical radiographs showed a lytic expansible lesion involving the body and especially the posterior arch of C2, which presented multiple fluid-fluid levels at the CT scan (Fig 3A). The surgical biopsy defined the diagnosis of aneurysmal bone cyst. At the same procedure, 200 IU of calcitonin and 120 mg of methylprednisolone were injected inside the lesion. This treatment was repeated twice with one-week interval, and then repeated monthly three times. Three months after the last injection, the patient referred significant pain reduction, and the CT scan showed the lesion intensely ossified and volume reduction (Fig 3).

DISCUSSION

Aneurysmal bone cysts are lesions containing thin-walled cystic cavities filled with blood. The ABC occurs more frequently among female patients of the first and second decades. Most of the lesions involve long bones, vertebrae and flat bones¹. In the spine the ABCs occur more frequently at the thoracic and lumbar segments, being rare in the cervical spine. In this study, we evaluated three cases of ABCs at the C2 level, showing the satisfactory imaging evaluation after treatment with intralesional injection of calcitonin and methylprednisolone.

The ABC affects initially the posterior arch and pedicles of the vertebrae, with bone destruction and cortical expansion. In some cases, it may compress the spinal cord and neural roots. The clinical presentation of vertebral ABCs is varied, and patients may present local deformity, movement limitations, muscular stiffness, torticollis, local heat, and occasionally, murmur over the affected region^{4,7-9}. In our study, the lesion affected mainly the posterior arch, but also the vertebral body. In addition, none of our cases presented significant neurological symptoms, even when there was reduction of the vertebral canal size and change of the vertebral axis.

The main pathological characteristic of the ABC is the presence of many blood cavities separated by thin

fibrous septa. This finding is demonstrated with CT scans and MR imaging, which show 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. The septations of the lesion are hypervascularized and the venous injection of contrast media can help to differentiate the thin and smooth septa of ABC from other tumors, which would have nodularities^{4,10-13}.

The vertebral ABCs may present on plain radiographs a characteristic "ballooning" of the posterior elements with a thin rim, although it may be difficult to see in some cases¹⁴. Pathological fractures are common and occasionally may progress to vertebra plana¹⁵. Involvement of multiple vertebrae may occur^{4,16} and the expansion of bone into the spinal canal can compress the dural sac. The hypervascular complex, with pathological circulation and even arteriovenous shunts, can be shown in selective angiography^{11,12}. The CT scan is highly sensitive to detect calcium and demonstrates the extent of the bone destruction, showing the many cavities filled with fluid-fluid levels. On MRI the ABCs have heterogeneous signal, forming multiloculated cysts with fluid-fluid levels better seen on T2-weighted images. On T1-weighted images, the fluid-fluid levels are hyperintense, presumably due to methemoglobin deposits^{4,11,12,17}.

In our study, the three cases of ABC at C2 appeared as expansible lytic lesions of the posterior arch, showing an eggshell thin cortex barely seen on plain films. The CT scans demonstrated better the thin outer cortex of the lesion with the cavities showing hypo/isodense fluid-fluid levels. MRI performed in case 1 corroborated the presence of blood inside the lesion, showing hyperintense and isointense fluid-fluid levels on T1-weighted images.

The treatment of aneurysmal bone cyst with less recurrence index was during many years the complete surgical resection. However, the resection of cervical vertebrae ABC is risky, and can cause instability, which is even more undesirable for growing child^{3,4,8,18-20}. The treatment approach of our cases, based in the infiltration of the lesion with calcitonin and methylprednisolone, was recently described⁷. Another less aggressive surgical treatment is curettage of the lesion with bone grafting. However, in most of the cases the curettage is incomplete, increasing the risk of recurrence²⁰. Radiation therapy for ABCs was already performed, but no more accepted due to the risk of posterior malignization and development of scoliosis caused by asymmetric growing of the irradiated site¹.

The intralesional injection of calcitonin and methylprednisolone has different effects over the bone lesion

when compared to fibrosing agent. The calcitonin can induce the formation of cancellous bone and inhibit the osteoclastic activity. The corticoid can reduce the fibroblastic action and the angiogenesis⁷. This kind of treatment can take years to cure, however, previous studies showed good results, without the common recurrence and the side effects of the fibrosing agents^{4,5}.

In our cases, the doses of calcitonin and methylprednisolone were decided empirically, following the doses injected in a previous study⁷. In each session we injected under general anesthesia 200 IU of calcitonin and 120–160 mg of methylprednisolone. Although a CT-guided infiltration could be performed, we avoided this method due to the risk of patients' movements during the procedure, since all of them were younger than 15 years. However, we defined the site of puncture and injection based on the CT scans, which were also used as the imaging method of choice for the follow-up. In all cases the follow-up CT scans showed that the osteoblastic activity was persistent, with progressive ossification of the cavities in a concentric way, thickening the walls and reducing the blood content inside them. At the end of the treatment, the ABCs were almost all ossified, with shrinkage of the lesions. After being considered out of risk of fracture or bleeding of the lesion, the children had functional radiographs of the cervical spine performed with flexion and extension, assuring the stability of C2.

In conclusion, the direct injection of methylprednisolone and calcitonin inside the ABC cavities seems to be a safe procedure, with no side effects that are usually seen in cases of infiltration using fibrosing agents. The CT scan is the imaging method of choice for the follow-up of the ABCs treatment, as it shows the progressive ossification of the cavities in a concentric way. We believe that three infiltration sessions with two month intervals result in significant bone thickening, being enough to prevent pathologic fractures or bleeding. Further studies could consider a longer follow-up, as the children are still growing and it

is expected that the ossified lesions of C2 will be remodeling together with the other growing vertebrae.

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