Accepted Manuscript

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PII: S1529-9430(16)00432-0
DOI: http://dx.doi.org/doi: 10.1016/j.spinee.2016.02.042
Reference: SPINEE 56930

To appear in: *The Spine Journal*

Received date: 6-9-2015
Revised date: 28-1-2016
Accepted date: 23-2-2016

Please cite this article as: Liberto Brage, Héctor Roldán, Julio Plata-Bello, Martel Diego, Víctor García-Marín, Transoral vertebroplasty for a c2 aneurysmal bone cyst, *The Spine Journal* (2016), http://dx.doi.org/doi: 10.1016/j.spinee.2016.02.042.

Title

Transoral vertebroplasty for a C2 Aneurysmal Bone Cyst

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ABSTRACT

BACKGROUND: Aneurysmal bone cysts at the cervical spine represent a real challenge both diagnostically and therapeutically, especially in young patients.

PURPOSE: To present an unusual case of a C2 aneurysmal bone cyst expanding the body in its entirety in a young adult successfully treated with a transoral vertebroplasty.

STUDY DESIGN: Case Report.

METHODS: We report the case of a 17 year-old woman with a history of cervical pain and occipital headache after a car accident. Routine X-rays disclosed a C2 lesion. Her neurologic examination was normal. Computed tomography showed a lytic lesion occupying almost the entire body of the C2 vertebra. The cortical bone was intact but notably thinned. MRI imaging revealed a cystic image with blood inside. Transoral vertebroplasty was selected among other surgical options for the following reasons: 1) to improve the clinical symptoms, and 2) to prevent future vertebral collapse with devastating neurological consequences. Under general anesthesia and continuous neurophysiological monitoring, we conducted a fluoroscopic-guided transoral vertebroplasty through a Jamshidi needle. A cytology sample from the cystic lesion was taken through the needle.

RESULTS: The blood smear showed no tumoral cellularity. There were no complications during surgery or postoperative infections. After 4 years of follow-up, the patient is pain-free and leads a normal life.

CONCLUSIONS: Transoral vertebroplasty seems to be a direct, safe, and effective technique to stabilize cystic lesions that endanger the stability of C2 and to improve symptoms. Aneurysmal bone cysts should be included in the differential diagnosis of lytic lesions at the vertebral body of C2.

Keywords: Axis, Aneurysmal Bone Cyst, Stability, Transoral, Vertebroplasty, Antibiotics.

INTRODUCTION

Aneurysmal bone cysts (ABC), representing 1.4% of all primary bone tumors and 15% of all primary spine tumors [1], are a rare benign condition, sometimes self-limiting, and generally located in the posterior arch of lumbar vertebrae, followed by the thoracic, cervical, and sacral segments [2]. Despite their benign nature, they can be locally aggressive, and can result in pathological fractures and neurological complications [3]. They can appear as primary bone lesions (in 70 % of the cases) or secondary (in about 30 %), when ABC-like areas are encountered inside other bone conditions (giant cell tumors, chondroblastoma, telangiectatic osteosarcoma, osteoblastoma) [4]. Although not pathognomonic, ABC are usually diagnosed based on their typical radiological appearance, consisting of multilocular cysts with fluid-fluid levels on MRI and bony septations on computed tomography [5].

The treatment of ABC is controversial. Multiple treatment modalities have been tried with variable improvement and recurrence rates. The options include curettage with or without bone grafting, complete excision, arterial embolization, intralesional drug injections (steroid and calcitonin) and radiation [6]. Intralesional curettage is the technique most often used in the spine [7,8]. Nevertheless, complete excision is the optimal approach to achieve local control of the tumor as it prevents recurrence, but this treatment exposes the patient to high surgical morbidity [9,10]. Radiotherapy has been indicated in unresectable lesions, as it has numerous and severe complications [11]. Arterial embolization has been used preoperatively to decrease surgical bleeding and also as a stand-alone approach for inoperable cases [12].

We report an unusual case of an ABC at C2 affecting the entire vertebral body successfully treated with transoral cementation in a young adult, in addition to discussing the rationale for the selected treatment.

METHODS

Our case is a 17 year-old female who reported a long history of cervical pain and occipital headaches. She had previously sought out medical attention, but no clear diagnosis had been achieved. She was subsequently involved in a car accident and sustained a whiplash injury. On routine cervical X-rays, a radiolucent lesion in the vertebral body of C2 was found (Fig. 1A). For this reason she was referred to our hospital. On examination, the only physical finding was neck pain without any neurological deficits. A computed tomography scan (CT-scan) showed a lytic expansile lesion occupying almost the entire body of C2 with intact but notably thinned anterior and posterior cortical boundaries. Multiple partitions could be seen inside the cavity (Fig. 1B,C). Magnetic resonance imaging (MRI) revealed a cystic lesion containing blood (Fig. 1C).

Given the symptoms, patient age and structural compromise of the integrity of C2 by the lytic lesion, we decided to perform a transoral vertebroplasty with three main objectives: 1) to improve the clinical symptoms, 2) to prevent future vertebral collapse with devastating neurological consequences, and 3) to obtain a pathological sample of the content of the lesion.

The surgery was performed under general anesthesia and continuous neurophysiological monitoring. The patient underwent systemic and local antibiotic prophylaxis in the oral cavity. The patient was placed supine with the head hyperextended and a Dingman mouth gag was used. The inner part of the mouth was draped. The entry point was located in the throat with the aid of biplanar fluoroscopy (Fig. 2A). A Jamshidi needle was used to reach the lesion allowing the evacuation of the hematic content, which was sent to the Pathology Department for postoperative diagnosis (Fig. 2B). We then performed the fluoroscopy-guided vertebroplasty by introducing approximately 1 cc of PMMA (Confidence Spinal Cement System™, DePuy Synthes, Inc., Warsaw, IN) (Fig. 2C), which easily filled in all the cavities. Once the final result was verified, the Jamshidi needle was withdrawn and the pharynx was inspected. A small bleeding point was controlled by applying low pressure with gauze. The procedure lasted

an hour and a half, and the patient awoke from anesthesia without complications. Oral rinses with chlorhexidine and intravenous cefazolin were prescribed for 72 hours after surgery, and no local or systemic infection signs occurred. A plain cervical radiography was taken confirming the integrity of both cortical boundaries and the patient was discharged from hospital after three days of admission.

The blood smear was normal without any tumoral cells. This confirmed our diagnosis of ABC.

After four years of follow-up, the patient is pain-free and leads a normal life. Plain X-Rays, CT-scan, and MRI show no signs of recurrence or progression of the C2 lesion (Fig. 3A,B,C).

DISCUSSION

To our knowledge this is the second case reported of a transoral cementation of an ABC at C2 [16]. This article describes the case of a 58 year-old male who had a C2 fracture secondary to a lytic lesion, treated with vertebroplasty through a transoral route. Our case is the first one in a young adult.

The main pathological feature of ABC is the presence of many blood cavities separated by thin fibrous septa. This fact explains the radiological image of a great cavity with many hollows in the interior filled with blood [9]. Our case showed, on both MRI and CT-scan, the typical radiological appearance of an ABC. MRI demonstrated a hyperintense signal at T2WI consistent with blood content (Fig. 1C), while the CT-scan showed the extent of the bone destruction with integrity of the cortical boundaries and several cavities filled with fluid (Fig. 1B,C). Blood was aspirated from the vertebral body of C2 through the Jamshidi needle. Pathological analysis was normal without any tumoral cells, which was therefore consistent with ABC [20].

Instability is fairly clear in literature when there is a fracture at C2 or a

C1-C2 or C2-C3 subluxation. Moreover, acute cord compression is reported in the literature in the absence of complete vertebral collapse [21]. We decided to indicate an invasive treatment in this case because the patient was young, active, symptomatic, and the very thin outer walls of the C2 body posed a significant risk of fracture with potentially catastrophic consequences. Stand-alone embolization and infiltration of fibrosing agents have been advocated for treating ABC [6,12]. However, these procedures were discarded from the beginning as none of them stabilizes C2 and the presence of very thin walls might not contain fluids from extravasation into the spinal canal. What to do from this point was a matter of thoughtful debate.

Open surgery of cervical ABC implies a combined approach. The anterior approach is used to accomplish the curettage, electrocautery and bone grafting, whereas the posterior approach is necessary to stabilize the segment [20]. Usually, at C2, the posterior stabilization requires an occipitocervical fusion [20]. The disadvantages include being a double procedure with a fairly complicated postoperative course and a high risk of intraoperative bleeding [22,23]. In fact, many authors advise to carry out a selective arterial embolization to prevent hemorrhage [12,20]. We also considered this patient as a high-risk case for curettage, as the boundaries were very thin and easy to pierce, with eventual neurological consequences.

In our case, as ABC show huge cavities filled with blood, vertebroplasty seemed to be an option. Sometimes bone septa of ABC are not complete, and the cavities are separated only by thin fibrous walls that can be torn by pressurized cement, so one injection can be enough to fill in all the cavities. Nevertheless, even though vertebroplasty at C2 seems to represent an important tool to control the pain and to prevent vertebral collapse and spinal cord compression, only a small number of cases have been published [14,15,17,18,19,24,25]. The advantages over open procedures include a quicker and easier method without an external incision in a young patient [26]. Percutaneous vertebroplasties at the thoracic and lumbar vertebral bodies are usually performed through a transpedicular route. At the subaxial cervical spine, an anterolateral oblique

approach is typically used [15]. To the best of our knowledge, the transpedicular approach to reach the body of C2 is not a safe option due to the proximity of the vertebral arteries and the small size of the pedicles [19]. The most straightforward access is provided by the transoral route, which has the advantage of the short distance to the C2 vertebral body, thereby avoiding important vascular structures [15,16]. The most significant drawback is crossing a naturally contaminated area. Infection rates for open transoral procedures vary widely from 0% to 50%, depending on preoperative site preparation, prophylactic antibiotic use, duration of the procedure, and the extent of tissue disruption [13]. In our patient we did not have any infectious complications. We provided systemic and local antibiotic prophylaxis, performing a rapid procedure through a minimum puncture in the oropharynx and were very careful not to touch the rest of the oral cavity with the Jamshidi needle (we draped the inner part of the mouth to prevent this). Cefazolin was continued for 72 hours after surgery until discharge. Daily pharyngeal inspection was conducted until discharge, and no sings of infection were noted. Some authors have described mixing PMMA with local antibiotics [27]. We did not do this as we were not certain about the effect of using a different substance on PMMA final consistency, and we were concerned about PMMA leakage at such a critical level as C2.

To accomplish the percutaneous vertebroplasty in our patient we selected polymethyl-methacrylate (PMMA) as opposed to biological cement for several reasons. Firstly, the use of biological cement in complex cases in not well known, with only a handful of papers describing the long-term effects of these materials, especially in case of leakage [28]. Secondly, even with integrity of cortical boundaries, we have more confidence in high-viscosity cement, as we do know how it changes over time until it becomes dough so it can be safely applied under radiological guidance [29]. Thirdly, the exothermic polymerization process that occurs with PMMA, reaching temperatures superior to the critical level for protein denaturation, can provide a tumoricidal effect in a lesion with high rates of recurrence [30].

As stand-alone cementation is not an established treatment for ABC, the ability to achieve local control of the lesion cannot be predicted beforehand, so patients must be followed up carefully. Nevertheless, cases of resolution of ABC after minor intralesional procedures have been published, which probably reinforces the nontumoral nature of these lesions [2].

CONCLUSION

Transoral vertebroplasty may constitute a safe and effective treatment to easily stabilize cystic lesions that compromise the stability of C2. ABCs affecting the vertebral body need to be considered when making the differential diagnosis of cystic lesions at C2 in young adults.

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FIGURE LEGENDS

Fig. 1. Preoperative images: (A) Plain radiography showing a lytic lesion at C2 body. (B), (C) Sagittal and axial CT-scan demonstrating an expansile osteolytic lesion. Note the integrity of the thin cortical boundaries and multiple partitions in its interior. (D) Preoperative T2WI MRI showing blood content inside the lesion.

Fig. 2. Intraoperative images: (A) Image of the operating room showing patient's position under biplanar fluoroscopy. (B) Blood coming out of the lesion through the Jamshidi needle. No pathological cells were found postoperatively. (C) Fluoroscopic image depicting the Jamshidi needle inside the lesion and the cement filling the cavities.

Fig. 3. Postoperative images: (A), (B) Plain radiographs showing the final result of the cementation. (C) CT-scan demonstrating that the lesion is completely full of cement. No extravasation of cement occurred.