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Percutaneous embolization of aneurysmal bone cyst of the mandible: A 3-year follow-up

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ABSTRACT

Aneurysmal bone cysts (ABCs) of the mandible are rare lesions with variable characteristics, including radiographic, histological and etiopathogenic features, mainly affecting patients in the second decade of life. This report describes and discusses an interesting case of an asymptomatic 7-year-old girl diagnosed with an ABC on the left side of the jaw. The lesion was treated by percutaneous embolization with Histoacryl® injection with fluoroscopic guidance. This is the first report in the literature of the use of this fibrosing agent in the mandible. Treatment was considered successful because all teeth maintained pulp vitality, and there was no recurrence for 3 years. Therefore, percutaneous embolization with Histoacryl® injection should be considered a safe, viable, and effective procedure for the treatment of ABCs in the jaw, replacing, in some cases, surgical therapy, and thus, being an important tool in the management of pediatric patients.

1. Introduction

Aneurysmal bone cyst (ABC) is a type of osseous lesion characterized by a benign pseudocyst with fibrous connective tissue stroma and large spaces filled with blood and no endothelial lining [1].

ABCs predominantly affect patients in the second decade of life [2], as about 80% of cases occur before age 20 [3–5], mainly between 10 and 20 years of age [6], being 13 years the median age of patients reported by many studies [5,7,8]. They can occur in all skeletal bones, but they are more common in the long bones, with prevalence of only 2% in the jaws [6].

There is no consensus in the literature regarding the best therapeutic method for treating ABCs. The most commonly used treatment methods are resection, curettage, embolization and intracystic injections. The choice of treatment method varies greatly, especially in children [9]. But, despite of the method chosen, the goal is always the success of the treatment, which consists of complete reossification, with new bone formation with normal volume and mineralization characteristics [10,11], and without recurrence of the cystic lesion [10]. Thus, the aim of this study was to describe a case of a 7-year-old female patient who developed an ABC on the left side of the posterior mandible and whose treatment was performed by percutaneous embolization with Histoacryl® injection.

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2. Presentation of case

A 7-year-old female in the mixed dentition stage sought orthodontic treatment with her parents who were unhappy about the appearance of her smile.

Intraoral examination revealed a Class II molar relationship with an anterior deep bite and atresic dental arches (Fig. 1A).

An initial panoramic radiograph (Fig. 1B) showed normal condition of both dental and bone structures. Therefore, informed consent was obtained from the mother of the patient and orthodontic treatment was conducted as planned; slow expansion was performed in both arches with removable expander appliances.

The patient presented for orthodontic appointments each month and in the last month, it was possible to observe an unexpected posterior disclusion only on the right side, after 11 months of treatment (Fig. 2A). A panoramic radiograph control was requested to evaluate possible causes for such disclusion. Surprisingly, the panoramic radiograph revealed a unilocular radiolucent lesion involving the left posterior mandible (Fig. 2B). Although it did not cause any root resorption, it caused a displacement of all permanent teeth adjacent to the lesion. There was a slight inferior cortical expansion, but it was not clinically detectable.

An axial computed tomography was conducted and showed the presence of unilocular bone rarefaction with defined contours at the left posterior mandibular area, expansion of the buccal and lingual cortical bone and displacement of the basal and lingual surfaces (Fig. 3).

Anamnesis excluded important medical or trauma history. Therefore, she was referred to an endovascular surgeon. Once again, informed consent was obtained from the mother of the patient and the treatment was immediately initiated.

A biopsy was conducted, a cytobacteriologic study was performed and the ABC was diagnosed. The endovascular surgeon performed a direct puncture of the aneurysmal bone cyst and a detection procedure of the intracystic septa. The verification of the absence of venous opacification and the measurement of the lesion volume were conducted using a Visipaque contrast material injection. Then,

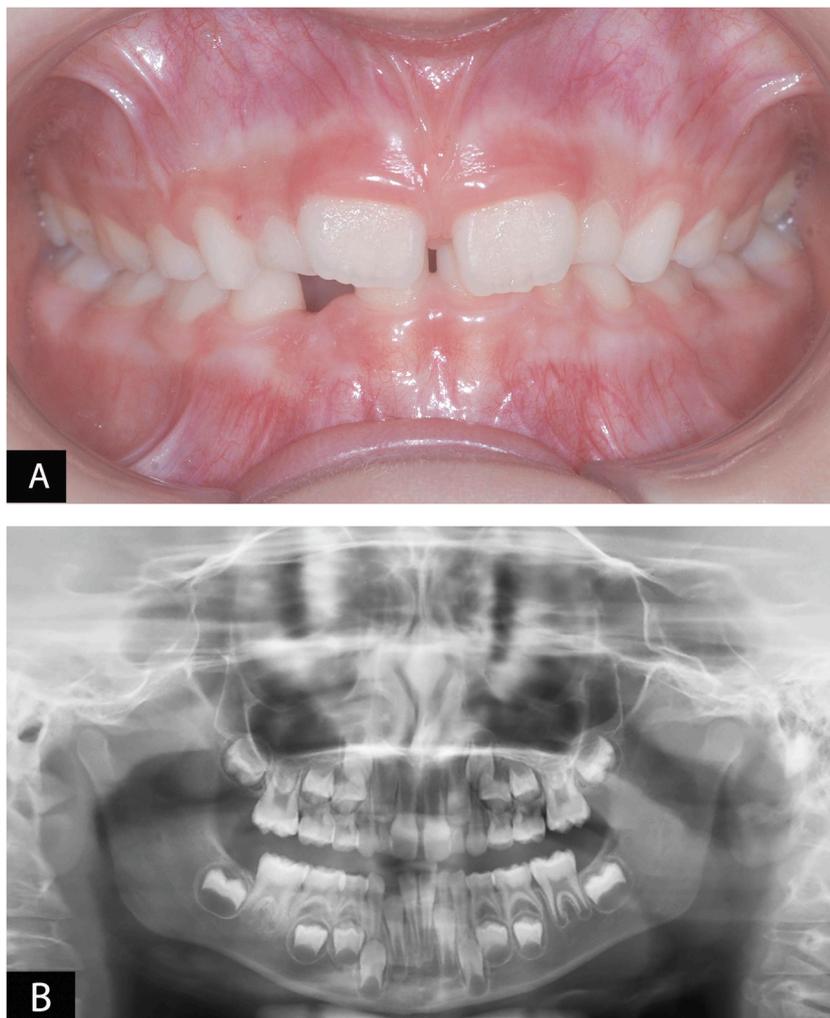


Fig. 1. (A) Pretreatment intraoral photographs and (B) pretreatment panoramic radiograph.

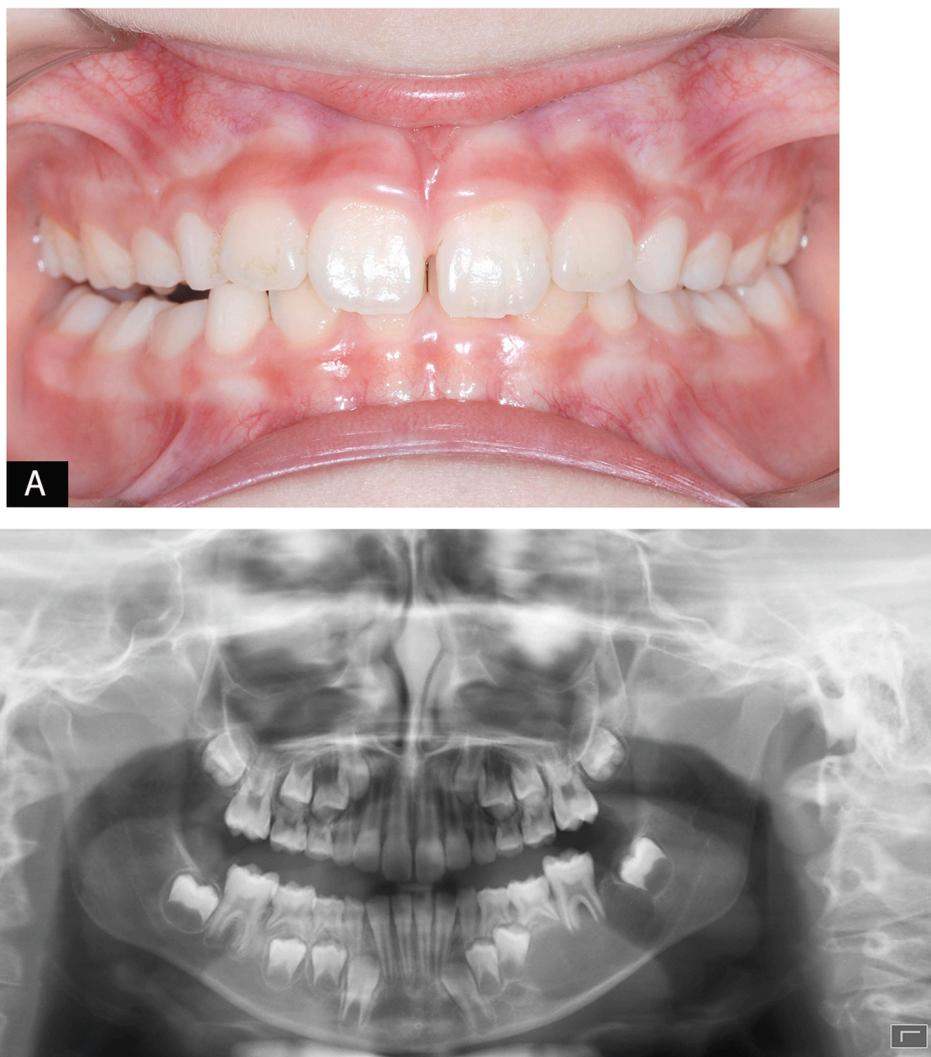


Fig. 2. (A) Posterior discusion on the right side of the mandible after 11 months of orthodontic treatment and (B) panoramic radiograph for posterior discusion control.

contrast material was aspirated, and the cystic area was washed with 0.9% saline. A combination of n-butyl-2-cyanoacrylate (Histocryl®; B. Braun, Melsungen, Germany) and lipiodol, in 1:1 ratio, was injected with fluoroscopic guidance under general anesthesia (Fig. 4).

The patient was followed for 3 years, and in the three-year posttreatment computed tomography image, there was no further sign of the cystic lesion (Fig. 5B).

3. Discussion

In some cases, the pathology of ABC may be associated with posttraumatic events, vascular malformation and genetic predisposition [9]. However, in the present case, such relationships could not be established, because the patient had no family history of such an occurrence and no history of trauma.

One of the most significant clinical characteristics in the majority of ABC patients is the increased swelling of the cyst area. Nevertheless, there was no swelling that was clinically detected, and the patient was asymptomatic. In fact, the cystic lesion was incidentally discovered in intermediary orthodontic exams that were requested by the orthodontist to evaluate the posterior discusion, which was manifested only on the right side during a short period of one month.

ABCs usually occur between 10 and 20 years of age with no sex predilection [6]. However, in this present case, the patient was 7 years old, which makes its presence an even more rare condition.

Surgical treatment, such as curettage and resection, is the most common therapy for aneurysmal bone cysts [12]. The extent of the treatment depends on the localization of the cyst, its size, its clinical characteristics and the age of the patient. However, additional

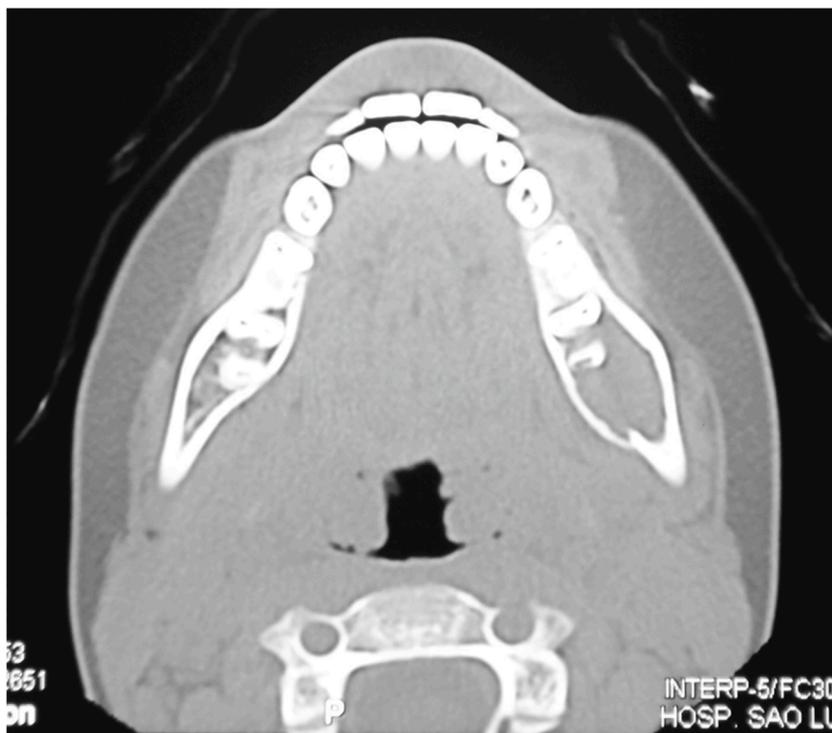


Fig. 3. Axial computed tomography image showing expansion of the buccal and lingual cortical bone on the left side of the jaw.



Fig. 4. Fibrosing agent injection with fluoroscopic guidance.

methods to improve or substitute surgical approaches have been described, including arterial embolization, radiotherapy and sclerotherapy [2,9]. With the vascular type, bleeding may be intense [6], especially when the lesion is reached. Accordingly, pre-operative embolization is commonly performed to minimize excessive bleeding during the curettage procedure.

Percutaneous embolization, or sclerotherapy, has been considered a treatment option for ABCs as reported by some authors [9,10,



Fig. 5. (A) One-year posttreatment panoramic radiograph and (B) three-year posttreatment computed tomography image.

13] primarily because, if performed by an experienced professional, it is an easy, safe, cost-effective and minimally invasive procedure compared to surgery. In addition, a success rate of over 90% with the use of sclerotherapy for treating ABCs is reported [14].

It is possible to find different fibrosing agents in the literature, including Ethibloc®, Absolute Alcohol and Histoacryl® [10]. However, the most commonly used agents are Ethibloc and Absolute Alcohol.

Histoacryl® consists of an acrylic resin (n-butyl-2-cyanoacrylate) that acts as a tissue glue, and in contact with blood, it is quickly polymerized, preventing bleeding [15]. In order to prevent n-butyl-2-cyanoacrylate solidification from occurring too fast, mixing with lipiodol is required [15]. After injection, the cystic lesion is filled with this solidifying mixture that is visualized as an opacified image (Fig. 5A) [16]. The Histoacryl® will then be reabsorbed, and the reossification process will take place in the region of the lesion (Fig. 5B) [16]. Many authors had demonstrated new bone formation occurring in different areas of the human body when sclerosis of bone lesions are performed with fibrosing agents [4,10,11,16–19].

The first use of n-butyl-2-cyanoacrylate in sclerotherapy was reported by Soehendra et al. in 1986 [20], who used this tissue adhesive agent to treat bleeding gastric varices, reporting the success of the therapy in 3 patients. Since then, many authors have begun to use this fibrosing agent in sclerotherapy.

However, since ABCs are most frequently found in the long bones, the therapeutic procedures most commonly reported also involve these skeletal regions. According to the literature, percutaneous embolization with Histoacryl® is generally used to treat ABCs in long bones [2,10]. In addition, this fibrosing agent is the only recommended to treat ABC in the skull and spine, due to the considerable inflammatory reaction caused by Ethibloc [10]. Rossi et al. reported 36 ABCs (4 in the thoracic cage, 6 in the spine, 9 in the long bones and 17 in the pelvis) treated with n-butyl-2-cyanoacrylate injection. The authors reported a success rate of 94%, and in most lesions (61%) only one embolization was required. In a total of 55 procedures performed, complications were observed only in 3 of them (5%) [4].

The literature also reports the use of Histoacryl® in other facial lesions. Alaraj et al. reported a series of 20 patients with cranial, facial, and neck tumors treated with n-butyl-2-cyanoacrylate embolization, either preoperatively or palliatively in cases of uncontrollable bleeding [21].

An interesting observation is that there are no reports of the use of n-butyl-2-cyanoacrylate in other cystic lesions of the jaws. The ABC have an uncertain etiology, and its nomenclature is still confusing because the lesion is not considered an aneurysm or a cyst due to the absence of endothelial wall [7,22]. Some authors suggest that ABCs derive from an arteriovenous malformation that may be associated with other primary bone lesions [3,4,23]. Thus, it can be speculated that the effectiveness of sclerosing agents in ABCs is related to their etiology, not working in other cystic lesions of the jaws. In such cases, surgical intervention is required.

There are no reports of the use of Histoacryl® in ABCs in the jaw, which makes this case so interesting.

In the present case, sclerotherapy was performed with Histoacryl® and lipiodol injection in a 1:1 ratio. The Visipaque contrast

material injection stage is very important, because it allows detection of venous opacification in the lesion; otherwise, the injection of the fibrosing agent is contraindicated [13]. Despite being a minimally invasive method, percutaneous embolization must be performed by a trained and experienced medical team because venous drainage is a major risk of this procedure [9].

Recurrence of ABCs usually occurs within two years, so patient follow-up should include at least 24 months [2,5]. In the present case, annual image controls for three years were conducted, and it was possible to observe the excellent regression of the ABC (Fig. 5). At the three-year follow-up, the cystic lesion had disappeared, and there was no sign of recurrence on imaging (Fig. 5B).

Percutaneous embolization has been considered a safe and viable procedure for the treatment of ABC, replacing, in some cases, surgical therapy, mainly when surgery offers a high risk to the patient or its execution is impossible. Additionally, it is a therapeutic approach that is well accepted by pediatric patients, and for some authors, it is the first-choice treatment [9,10].

In the present case, it was possible to notice the improved position of all the permanent teeth adjacent to the cystic lesion (Fig. 5B). Additionally, all teeth maintained pulp vitality.

4. Conclusion

In conclusion, percutaneous embolization with Histoacryl® injection should be considered a safe, viable, and effective procedure for the treatment of ABCs in the jaw, replacing, in some cases, surgical therapy, and thus, being an important tool in the management of pediatric patients. In addition, this case highlights the importance of dental practitioners to always be aware of clinical conditions and radiographic images of their patients because early diagnosis of particular lesions can be critical in successful treatment. The orthodontist may play an important role in the diagnosis of particular lesions because he may be the first professional sought by numerous patients.

Informed consent

This case report was conducted in a private practice. Informed consent was obtained from the mother of the patient. This case was exempted from Institutional Review Board (IRB).

The principles of the Declaration of Helsinki were followed - "All procedures performed in the present study involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards."

Conflicts of interest

The authors declare no conflict of interest.

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