Case report

RECURRENCE OF ANEURYSMAL BONE CYST OF THE MANDIBLE: A CASE REPORT

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ABSTRACT

Aneurysmal bone cysts are benign osteolytic lesions with rapid growth, thin walls containing numerous blood-filled cavities. They occur mainly in the pediatric population but are rarely diagnosed in the facial skeleton. Although benign, the condition can be locally aggressive and cause significant weakening of the affected bone structure. This case report presents a 14-year-old boy with Burkitt's tumor (Non-Hodgkin's lymphoma), previously diagnosed with an aneurysmal bone cyst a year ago. He was admitted for treatment at the Clinic of Maxillo-Facial Surgery of the University Hospital "St. George" Plovdiv due to pain and swelling in the left molar area of the mandibula, dating from 2 days. A recurrence of an aneurysmal bone cyst measuring 55/25 mm, which does not require resection of the mandible, was found. After a decision for surgical removal of the formation, under general anesthesia, and preparation of a three-cornered mucoperiosteal flap, the mandibular bone, which is lysed and thinned by the formation, was trepanned. The roots of the affected teeth were resected, and the formation was extirpated. A gauze drain was placed in the formed cavity, and the wound was sutured. Drug therapy included Cefotaxime 2x1.0 - 10 days, starting 3 days before surgery, and Paracetamol 2x1 - 3 days, 2 days before and 1 day after surgery. Prophylactic examinations were scheduled on day 3, 7, 10, 15 after the operation, as well as 3, 6, and 12 months post-op, due to the recurrent nature of the formation.

Keywords: nonodontogenic, aneurysmal bone cyst, mandible, maxillofacial surgery,

BACKGROUND

An aneurysmal bone cyst(ABC)is a benign osteolytic lesion with rapid growth, thin walls containing numerous blood-filled cavities [1]. It was first described in 1893 as an ossifying hematoma by Van Arsedale [2, 3]. ABC is considered a result of a vascular malformation within the bone, though the ultimate cause of the malformation is still under debate [4]. It occurs mainly in the pediatric population [5], most commonly affecting individuals in their second decade of life and can occur in any bone in the body[6]

but is rarely diagnosed in the facial skeleton [7]. ABC accounts for 1.5% of neodontogenic tumors of the jaw bones [8]. The average age of occurrence is 13 years, and 80% of patients are less than 20 years old with no gender predilection [9].

Histologically the ABC is considered a pseudocyst due to the absence of an epithelial wall. Recurrence is very typical, which confirms the aggressive behavior of this type of lesion [9].

Although nonodontogenic cysts of the jaws are relatively uncommon, it is imperative the clinician understand their clinical behavior and management because failure to do so can result in significantly increased patient morbidity [10]. Although benign, the condition can be locally aggressive and cause significant weakening of the affected bone structure. Strong pressure on the surrounding tissues in some cases can lead to deformities, neurological symptoms (depending on the location) and pathological fractures [1,3]. The etiology and pathophysiology are not yet clear [11], and currently, the standard of care and most widely used treatment is intralesional curettage [5].

CASE DESCRIPTION

The patient is a 14-year-old boy with a concomitant Burkitt's tumor (non-Hodgkin's lymphoma). He was admitted for the second time for treatment at the Clinic of Maxillo-Facial Surgery of the University Hospital "St. George" Plovdiv. At the first admission (12 months ago), he was diagnosed with an aneurysmal bone cyst measuring 24/37 mm. in the area of teeth 34, 35, 36, and the cyst was removed surgically by performing a cystectomy. The current admission was due to pain and swelling again in the left molar area of the mandibular bone, dating from 2 days.

The general status of the patient was without deviations. On extraoral examination, there was no facial asymmetry. Intraoral examination revealed normal poisoning of the mouth and a swollen vestibular plate of the lower jaw on the left side in the area of the molars, and on palpation, pain was provoked. A panoramic radiograph (orthopantomography) (Figure 1) revealed a cystic formation in the area of the lower jaw on the left.

Fig. 1. Orthopantomography, showing cystic formation in the area of the lower jaw on the left.

Due to the suspicion of a malignant process, after consultation with a pediatrician, a puncture was performed intraoperatively, and 35 ml. of blood was withdrawn (Figure 2). Pre-surgically, teeth 31, 32, 33, 34, 35, 37 were prepared endodontically, except for 36, which did not allow treatment due to canal obliteration. A CT scan of the head was performed to specify the possible subsequent intervention. An expansile multicystic lesion, representing a recurrence of an aneurysmal bone cyst measuring 55/25 mm, which does not require resection of the mandible, was found.

Fig. 2. Withdrawal of blood from the cystic formation.

After a decision for surgical removal of the formation, under general anesthesia, and preparation of a three-cornered mucoperiosteal flap in the area from the first incisor (31) to the last molar tooth (38) (Figure 3), the mandibular bone, which was lysed and thinned by the formation, was trepanned. The roots of the affected teeth were resected, the formation was extirpated and then sent for histological examination, with the morphological results corresponding to the clinical diagnosis (Figure 4). A gauze drain was placed in the formed cavity, and sutures (Figure 5) were placed, which were removed on day 10 post-op.

Fig. 3. Preparation of three-cornered mucoperiosteal flap.

Fig. 4. The extirpated cystic formation sent for histological examination.

Fig. 5. Check-up 3 days after the operation.

Drug therapy included Cefotaxime 2x1.0 - 10 days, starting 3 days before surgery, and Paracetamol 2x1 – 3 days, 2 days before and 1 day after surgery. Prophylactic examinations were scheduled on days 3, 7, 10, 15 after the operation, as well as 3, 6, and 12 months post-op, due to the recurrent nature of the formation. The patient made an uneventful recovery.

DISCUSSION

It is well documented that cysts within the pediatric population tend to be developmental and odontogenic in nature. Nonodontogenic cysts within the jaws, such as ABC, are not a common finding in either the adult or pediatric patient population [10]. Bernier and Bhaskar described the first case of ABC in the jaws in 1958 [12]. ABC accounts for 1.5% of the nonodontogenic, nonepithelial cysts of the mandible [8]. It is found more frequently in the mandible than the maxilla (3:1) with preponderance for the body, ramus and angle of the mandible [11] and rarely affects the condyles [13, 14].

ABCs have been classified as primary and secondary, where primary may be divided into congenital or acquired types [3]. Histopathologically, the vascular variant is more common and consists of about 95% of total cases of ABC, while the solid type is rare, occurring in only 5–8% of cases [7, 13].

ABC is a lesion with a unique clinical, radiological and diagnostic finding [14]. Although pediatric jaw lesions are uncommon, symptoms such as swelling and

pain could indicate potential pathologic findings and require panoramic examination [15]. Patients usually undergo different modalities of imaging, namely X-ray, CT and MRI, which show an expansile multicystic lesion of the respective bone site [16].

Management of pediatric jaw lesions should consider the biologic behavior of the lesion, maxillofacial development, and growth [15]. Curettage, bone grafting, or resection are the common types of surgical treatment. Currently, the standard of care and most widely employed treatment is intralesional curettage. However, tumor recurrence with curettage alone is common and has driven some to propose a multitude of adjuvants with varying efficacy and risk profiles [5]. For some clinicians, the treatment of choice is surgical resection, considering the high recurrence rate [7].

In a review of the literature, curettage or resection was employed equally, and recurrence rates were 60% after curettage, 20% after resection. All recurrences occurred within 12 months, correlating with the presented case report [17].

Some authors consider drug treatment and state that denosumab provided a meaningful clinical and radiological improvement in ABC; however, studies with more cases are required for routine use of denosumab in ABC [18].

CONCLUSION

Maxillofacial ABCs are rare lesions with variable clinical and histological presentations. Treatment of choice is most commonly curettage, or in some cases resection, depending mostly on the location and size of the formation, as well as on the biologic behavior of the lesion and maxillofacial development in young individuals which it predominantly affects. Due to the high recurrence rate of ABC, a close post-operative follow-up for at least 12 months is recommended.

Abbreviations:

ABC -Aneurysmal bone cyst

CT – Computer tomography

MRI - Magnetic resonance imaging

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