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CASE REPORT

Aneurysmal bone cyst of the zygomatic bone

L.K. Surej Kumar & P. Varun Menon

Department of Oral and Maxillofacial Surgery, Kerala Institute of Medical Sciences (KIMS), Trivandrum, Kerala, India

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Correspondence to
LK Surej Kumar
Department of Oral and Maxillofacial Surgery
Kerala Institute of medical sciences (KIMS)
P.B.No.1, Anayara P.O
Trivandrum 695029
Kerala
India
Tel.: +919447032516
Fax: +91-472-258 7874
Email: surejkumarlk@gmail.com

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Abstract

Aneurysmal bone cyst (ABC) is a rare haemorrhagic, non-neoplastic lesion, most commonly seen involving the long bone and spine. It is characterised by its expansile nature, high vascularity and multi-cystic appearance. It is a rare lesion in the maxillofacial region with its occurrence more in the mandible than in the maxilla. So far only four cases of ABC in the zygoma have ever been reported in the English literature. Here we report a rare case of ABC of zygomatic bone with a short review.

Introduction

Aneurysmal bone cyst (ABC) is a benign, non-neoplastic lesion of the bone, characterised by replacement with fibro-osseous tissue containing blood-filled sinusoidal or cavernous spaces. It is commonly found in the long bones and spine, and its incidence in the maxillofacial region is rare and the mandible is often affected more than the maxilla. So far it has been found that only four cases of ABC of zygoma have been reported till date[1]. Though the aetiopathogenesis still remains a mystery, various authors have proposed different theories for the same. The characteristic signs of ABC include sudden growth and bi-cortical expansion with a cystic appearance. The rapid destruction of the bone and cystic changes often mislead the clinicians in terms of malignancy.

Case report

A 63-year-old male came to oral and maxillofacial department with a complaint of a swelling on right cheek region since 3 months (Fig. 1) and had visited various centres for the same. He had been under intravenous antibiotics for the same from different centres and also had undergone extraction of the maxillary teeth for the same. No history of trauma was elicited.

Clinical examination revealed a large swelling extending from infraorbital margins to the anterior wall of maxilla and laterally to the zygomatic arch. Skin over the swelling was smooth without punctum and non-adhered. Fine-needle aspiration cytology (FNAC) was done extraorally and dark coloured blood was the aspirate. Computed tomography revealed bicortical expansion of a zygoma with the probability of an expansile osteolytic lesion involving right zygomatic bone with a cystic space (Fig. 2). Correlating CT findings, clinical findings and aspirate, a provisional diagnosis of ABC was made. An interesting observation was that, one week post-operatively a sinus formation was seen at the FNAC site. Surgical excision of the lesion was planned under General anesthesia (GA).

An extraoral approach was decided to facilitate the excision of the orocutaneous fistula along with the lesion. Through a standard Webber–Fergusson incision, extraoral sinus was marked along with the

Weber–Fergusson incision, and flap was raised. Intraoperative cortical plate over the sinus tract was seen thinned out (Fig. 3), the outer cortical plate was removed and the inner cortical plate was smoothened with a vulcanite bur till a smooth bony surface was obtained (Fig. 4). Local advancement flap from cheek was taken and closure was done primarily (Fig. 5). Post-operative phase was uneventful (Fig. 6).

Discussion

In spite of a descriptive history for more than 70 years since the first ABC was described, the clinical nature, behaviour, aetiology and treatment still remains a surgical dilemma. An ABC is an expansile, often multilocular, osteolytic lesion, with blood-filled spaces separated by fibrous septa containing giant cells and reactive bone[2]. Jaffe and Lichtenstein[3] first described it as a distinct pathological entity and gave its definition. The term aneurysmal is in fact related to an expansion or distension and cyst usually represent the blood-filled cavity[4,5].

Figure 1 Pre-operative clinical picture.

The ABC is usually seen occurring in the initial two decades of life and accounts for 1–2% of all primary bone tumours[6]. It is commonly seen in long bones and vertebrae and rarely seen in jaws, and the mandible is affected twice as frequently as the maxilla[7–9]. Another study has also shown the mandible to be more involved than the maxilla in a ratio of 55% to 45%. Both males and females were equally affected without sex predilection[8,9]. The rarest of the site for occurrence is the zygomatic bone with only four cases so far been reported till date[1].

The aetiology of the ABC is still uncertain and various theories have been proposed. According to Lichtenstein[3], a change in local haemodynamics perhaps following thrombosis of a sizeable vein or an anomalous arterio-venous communication may have resulted in the formation of ABC. As there was no evidence of muscular tissue in the vascular channel, Lichenstein theory was discarded. Some authors[10] suggested complex connective tissue replacement of a canalised haematoma of bone marrow. They proposed that if the circulatory connections are maintained by haematoma with damaged vessels, then an ABC develops and on the other hand, if this connection is blocked then a giant-cell granuloma is formed. They also suggested it as a false aneurysm as blood was seen circulating through it[10]. No further advancement in the aetiology has taken place since then.

Clinically, ABC presents as an asymptomatic lesion that may grow rapidly in time, resulting in

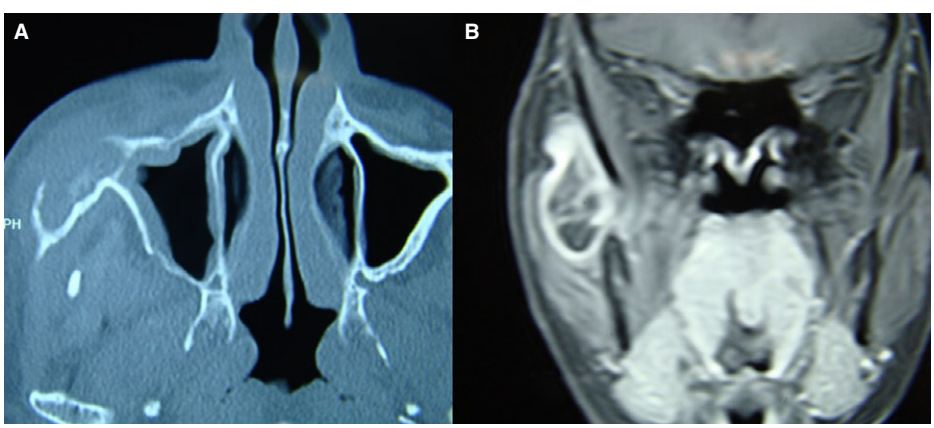

Figure 2 Axial and coronal CT images.

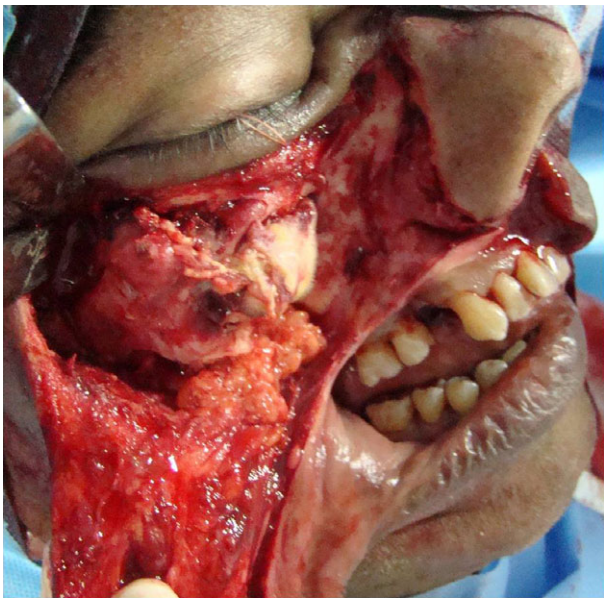

Figure 3 Standard incision placed and flap raised.

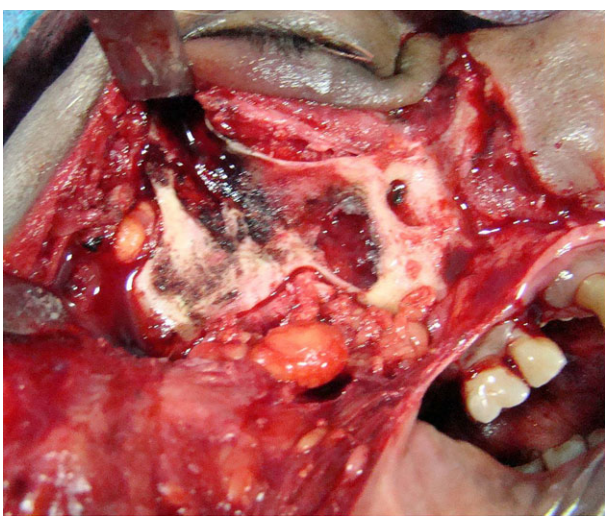

Figure 4 Intraoperative picture after smoothening the bony surfaces.

Figure 5 Closure of surgical site.

Figure 6 Follow-up post-operative picture.

expansion and destruction of the surrounding bone structure; subsequent invasion of the cortical bone and compression on adjacent nerve, soft tissue or joint inevitably results in pain[1,7,8].

Definite diagnostic criteria of an ABC have yet to be established in spite of more than half a century since its first presentation. Radiographic findings are suggestive, but not definitive, in diagnosing the lesion. The imaging studies, even CT, does not provide a clear diagnosis and ABC may be added to other list of differential diagnosis like venous malformation of the bone, ameloblastoma, central giant-cell granuloma, myxoma, intraosseous haemangioma, chondroblastoma and osteoblastoma[10]. Normally, they present as cortical expansion with radiolucent or mixed variant giving a cystic appearance. CT is usually helpful in knowing the extent of the lesion rather than knowing the diagnosis[11].

Aspiration is considered as one of the diagnostic techniques, but our experience though with single case aspiration can result in formation of orocutaneous fistula, so aspiration should be preferentially avoided or postponed unless immediate surgery is planned.

Various treatment modalities available for ABCs include: simple curettage, complete excision, radiation therapy, embolisation or a combination of these methods[12]. Sometimes these lesions especially in lower limbs are very aggressive[12] and are treated by amputation or excision. No such aggressive lesions have been reported in head and neck region, so curettage of the lesion has been advocated. We too advocated curettage with exception that in our case the overlying was excised as the aspiration had resulted in orocutaneous fistula formation.

A relatively high recurrence rate of 10–20% is seen in case of ABCs; however, recurrence is usually rare when the tumour is completely removed[12]. There is not much of known recurrence rate reported for cases in head and neck region and occurrence of ABC in zygoma even rarer as only four cases has been reported till date[1], and regular follow-up has to be maintained to evaluate the recurrence.

Conclusion

Although science is so advanced, no protocol has been established for proper diagnosis and treatment of ABC. Its aetiopathogenesis as well as its diagnostic and treatment dilemma continues. Perhaps research still going on in these various aspects will probably bring in new dimensions in the treatment of ABC in future.

Conflict of Interest

The authors confirm that there are no conflicts of interest.

Ethical Approval

None required.

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