Aneurysmal Bone Cyst Maxillary Sinus: A Case Report

Supreet S Nayyar

ABSTRACT

An aneurysmal bone cyst (ABC) is a rare bone lesion. Its origin and precise nature remain unknown. It is seen as a locally destructive, rapidly expandable, and benign multicystic mass. We report a 26-year-old male patient with an ABC of the maxilla, with extensive local involvement and bony expansion that was treated surgically.

Keywords: Aneurysmal bone cyst, Maxilla, Maxillary cyst.

International Journal of Head and Neck Surgery (2020); 10.5005/jp-journals-10001-1397

INTRODUCTION

Aneurysmal bone cyst (ABC) is a benign osteolytic bone neoplasm characterized by blood-filled spaces separated by fibrous septa. ABC may occur in any bone in the body. However, only 2% of the cases are found in the head and neck. And very few cases have been reported in the literature of ABC in maxillary sinus. We present here one such similar case.

CASE DESCRIPTION

A 26-year-old male patient presented with nasal obstruction (right) and mucoid continuous nasal discharge (right) for 5 months. Anterior rhinoscopy and nasal endoscopy revealed soft globular pinkish mass in right nasal cavity in middle meatus, sensitive to touch, did not bleed on touch and middle turbinate pushed medially. Endoscope could be passed superiorly, inferiorly, and posteriorly. Nasopharynx was WNL. Computed tomography (CT) of the paranasal sinuses showed a destructive soft tissue density heterogeneous mass lesion in right maxilla with few irregular foci of calcifications noted within the lesion. Lesion was extending anteriorly till anterior wall of maxillary sinus which was thinned out and bowed anteriorly, posteriorly extending till pterygoid plates without accompanying destruction. Superiorly, there was thinning of inferomedial wall of right orbit. Superomedially, there was extension into ethmoidal air cells; and inferiorly there was thinning of floor of maxillary antrum. Lesion was reaching up to nasal septum which was pushed into left nasal cavity; laterally, there was thinning out and splaying of lateral wall of maxillary sinus. Endoscopic biopsy from nasal mass was performed in which a distinct give way into the lesion was felt while taking biopsy followed by extensive bleeding from the biopsy site. Tissue biopsy done was superficial and noncontributory. Angiography showed a high vascularity of the tumor with the main supply from unnamed branch of right sphenopalatine artery. Preoperative embolization of that branch of sphenopalatine artery was done. He underwent excision of aneurysmal bone cyst from left maxillary sinus. Weber Ferguson incision on right side with lip split was given. On raising the flap, a breach in anterior wall of maxillary sinus was noticed. The breached portion was further widened, and a part of medial wall of maxillary sinus was removed to deliver out the complete mass in toto (Fig. 1). Postoperative recovery was uneventful.

Gross examination of cut specimen showed variable blood-filled cystic spaces (Fig. 2). HPE revealed tissue bits lined partially

Department of Otorhinolaryngology, Head and Neck Surgery, Army College of Medical Sciences, Delhi, India

Corresponding Author: Supreet S Nayyar, Department of Otorhinolaryngology, Head and Neck Surgery, Army College of Medical Sciences, Delhi, India, Phone: +91 9707118035, e-mail: ssnayyar@gmail.com

How to cite this article: Nayyar SS. Aneurysmal Bone Cyst Maxillary Sinus: A Case Report. *Int J Head Neck Surg* 2020;11(4):81–83.

Source of support: Nil

Conflict of interest: None

by columnar to pseudostratified squamous epithelium (Fig. 3). Subepithelial tissue revealed variable blood-filled cystic spaces devoid of any endothelial lining and separated by fibrous septa (Fig. 4). Focal areas with calcified matrix and few bony trabeculae were seen in the intervening septa. Histomorphological features were consistent with an ABC. He was well when reported for review 2 months postoperatively.

DISCUSSION

As defined by the World Health Organization, ABC is a benign tumor-like lesion.¹ It is described as “an expanding osteolytic lesion consisting of blood-filled spaces of variable size separated

Fig. 1: Complete mass being delivered out of maxillary sinus

Fig. 2: Cut section showing blood filled spaces inside the mass

Fig. 3: Surface lining of columnar to pseudostratified squamous epithelium

Fig. 4: Subepithelial tissue revealed variable cystic spaces devoid of endothelial lining separated by fibrous septa

by connective tissue septa containing trabeculae or osteoid tissue and osteoclast giant cells." Its origin and precise nature remain unknown. It is a locally destructive, rapidly expanding, benign multicystic mass. ABCs are generally considered rare, accounting for only 1–6% of all primary bony tumors. Most studies have also found a slightly increased incidence in women. Although the ABC can appear in persons of any age, it is generally a disease of the young (but rare in the very young). About 50–70% of ABCs occur in the second decade of life, with 70–86% occurring in patients younger than 20 years. The mean patient age at onset is 13–17.7 years. Only 2% of ABCs are found in the head and neck, with 66% of these being located in the mandible.²

The true etiology of ABCs is unknown. ABCs may be caused by a reaction secondary to another bony lesion or ABCs may arise de novo, or ABCs may arise in an area of previous trauma. Different theories about vascular malformations exist; these include arteriovenous fistulas and venous blockage. The vascular lesions then cause increased pressure, expansion, erosion, and resorption of the surrounding bone. The malformation is also believed to cause local hemorrhage that initiates the formation of reactive osteolytic tissue.

In ABC of maxilla, patients present with complaints of a tender swelling mass on the face, with progressive enlargement over weeks to years. Other signs and symptoms are basically on extent

of lesion, e.g., nasal symptoms with extension nasally as in our case, eye symptoms when there is orbital extension, and recent mobility or migration of the teeth.

Preoperative diagnosis of the ABC is difficult. Radiographically, the aneurysmal bone cyst is characterized by irregular cyst-like radiolucencies intermingled with thin bony trabeculae giving a honeycomb or soap-bubble appearance. Laterally, the lesion is usually covered by a thin cortical layer.

FNAC or incisional biopsy can be done. On histology, the gross appearance of the ABC is that of a blood-soaked sponge. A thin subperiosteal shell of new bone surrounds the structure and contains cystic blood-filled cavities. The tissue within shows brownish intertwining septa. The stroma contains proliferative fibroblasts, spindle cells, areas of osteoid formation, and an uneven distribution of multinucleated giant cells. The tissue within the septations includes cavernous channels that do not contain a muscular or elastic layer in their walls. Areas of new and reactive bone formation can also be found in the ABC.

Mitotic figures are common to ABCs, but no atypical figures should be evident.

Surgery is the main treatment of ABC of the maxilla. However, due to the vascularity of the mass, preoperative angiographic examination with embolization of the feeding vessels as close to the lesion as possible, should be done. This reduces the excessive bleeding during surgery. The principle of surgery is to fully excise the tumor. Curettage with enucleation is the first choice of treatment and depends on the extent of the tumor locally.

ABC has a high recurrence rate, which varies from 26 to 56% and can be due to incomplete removal of tumor. Therefore, the patient should be kept under regular follow-up.

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