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Case Report

Odontogenic keratocyst: wolf in a sheep's coat

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ABSTRACT

Odontogenic keratocyst (OKC) is a benign yet locally aggressive odontogenic lesion with a high recurrence rate. Although more commonly found in the mandible, OKCs may rarely involve the maxilla, posing diagnostic and surgical challenges. We report the case of a 65-year-old female with a recurrent, painful swelling in the upper gingiva, radiologically presenting as a well-defined expansile lytic lesion in the maxilla. The cyst was surgically enucleated under general anesthesia, and histopathology confirmed the diagnosis of odontogenic keratocyst. The patient remained asymptomatic with no recurrence at five months follow-up. This case highlights the need for awareness of atypical presentations of OKC, the importance of histopathological confirmation, and the role of long-term follow-up to monitor for recurrence or malignant transformation.

Keywords: Odontogenic keratocysts, Maxilla,

INTRODUCTION

Odontogenic keratocysts (OKCs) are benign, locally aggressive cystic jaw lesions with genetic predisposition and have high chance of recurrence. These lesions present a unique challenge in surgery as they originate from the dental lamina or primordial odontogenic epithelium. In 2017, WHO renamed this as odontogenic cystic lesion, which happens to be a debatable change in the terminology of odontogenic lesions in recent years.[1] The most common symptoms being swelling and pain.[2] Lesions have a slight predilection for males, presenting at two peaks of life, the first during second to third decades and the second during the sixth to seventh.[3] OKCs commonly occur in the ascending ramus or angle of mandible and less commonly in the maxilla. Very rarely they have reported to arise from soft tissues like gingiva, buccal space and posterior trigone of the molar, deep facial area or the temporal muscle.[5] OKCs constitute 10-15% of all jaw lesions and 2% to 21.8% of all jaw cysts. The lesions cause resorption of bone with destruction and with displacement of neighboring teeth.[6] Clinically they may be asymptomatic and are difficult to detect in early stage or may be found incidentally on radiography.[7] They may have a ping-pong like consistency due to thinning of overlying bone or they make parchment like brittle sound when palpated. They present with swelling and pain, which is attributable due to cyst infection or pathological fracture due to the expanding lesion causing facial deformity.[7] They appear as well-defined radiolucent lesions which may be unilocular or multilocular depending upon size and stage.[1,8] CT scans are useful in identifying bone involvement and remodeling. Management depends upon presenting symptoms & can be conservative or aggressive. In view of their tendency to recur, aggressive treatment is often quite required, though morbid.[5] Aspiration of OKC often yields yellow or white, keratin-like material.[5] On microscopic examination, parakeratin-lined cyst with basal nuclear palisading is seen.[1]

CASE REPORT

A 65-year-old female presented with the complaint of painful swelling in upper gingiva since 4-5 months. Patient gave history of fall and injury to the upper jaw 10 years ago. She underwent incision and drainage at a local dental clinic multiple times, whenever the symptoms arose and was relieved temporarily. The swelling would then recur after few days.

On examination, the upper jaw appeared swollen & palpation revealed a swelling which appeared cystic, fluctuating and tender. Intraoral examination, the overlying mucosa appeared inflamed. Patient was edentulous. A CT scan was suggestive of a right sided well defined round to oval non enhancing expansile lytic lesion within maxilla showing signs of bone remodeling with thinning of its bony wall. No signs of any soft tissue infiltration.

Figure 1: Swelling over upper jaw.

Figure 2: Axial view.

Figure 3: Bony cavity.

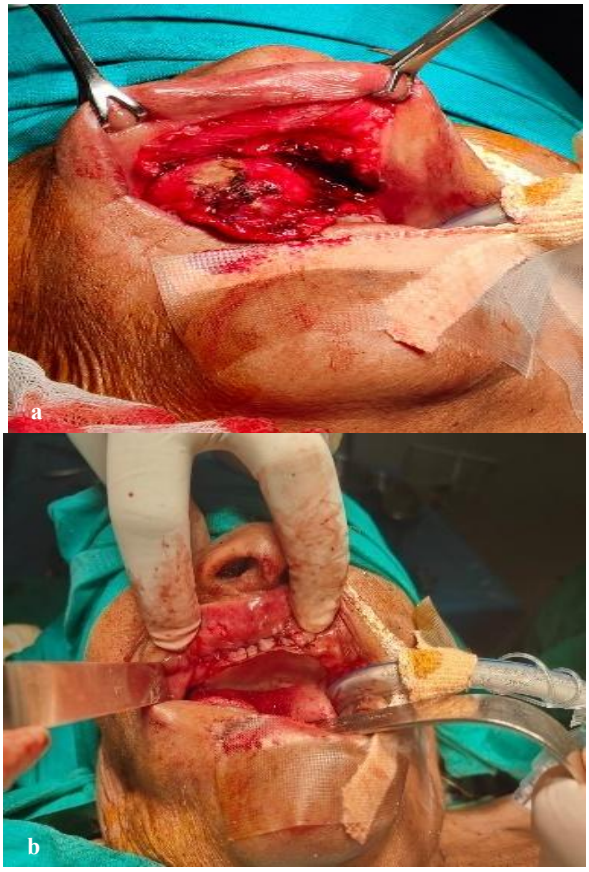

Figure 4 (a and b): Dissection and suturing.

Intervention

After proper check-up the patient was prepped and draped. The patient was taken up under general anaesthesia for excision of the mass. After 2% lignocaine infiltration a 2 cm incision was given over the swelling. The mucoperiosteal flap was elevated. and soft tissue dissection was done around the swelling. The entire cyst was enucleated. The cystic fluid and the cyst were sent

for histopathology. The surrounding thinned out bony fragments were removed to reduce the cavity. A thorough wash of betadine and hydrogen peroxide 1% was given to remove residual debris. The wound was closed in two layers with 3-0 vicryl.

The patient was asymptomatic at five months follow up with a healed cavity. Figure 5 shows tumour lined by stratified squamous epithelium with basal palisaded epithelial cells and parakeratotic layer at the top. Cystic wall was ulcerated at places with inflamed granulation tissue. The lumen showed haemorrhage and keratin flakes and the wall showed fibro-collagenous tissue with granulation tissue and areas of haemorrhage. Many congested and dilate blood vessels and chronic inflammatory infiltrates composed of lymphocytes and plasma cells were also seen. The deeper tissue shows many bony spicules along with fibromuscular tissue and minor salivary glands. There was no evidence of granuloma, atypia or malignancy.

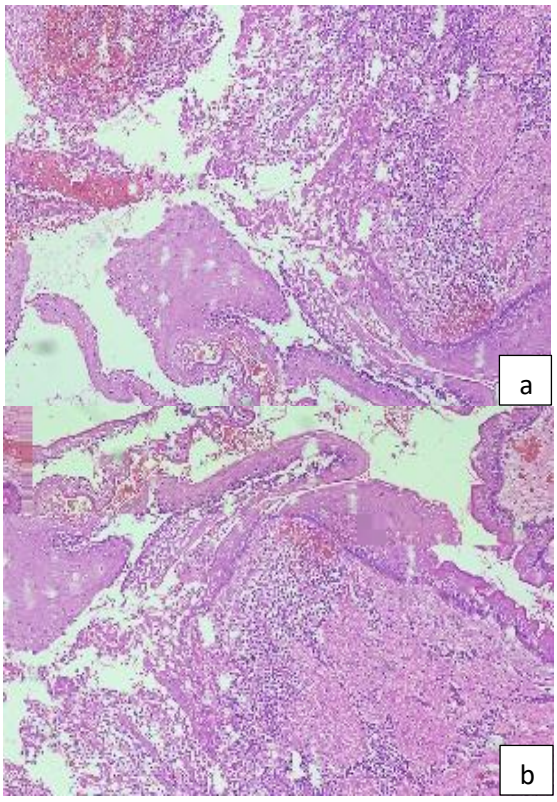

Figure 5 (a and b): Histopathology slides showed tumor lined by stratified squamous epithelium with basal palisaded epithelial cells and parakeratotic layer at the top. Cystic wall was ulcerated with inflamed granulation tissue. The lumen showed keratin flakes and the walls shows fibro-collagenous tissue.

DISCUSSION

The odontogenic keratocysts are developmental cysts that originate from remnants of the dental lamina within the jaw bones. OKCs exhibit a wide range of clinical presentations, frequently showing a paucity of symptoms in their initial phases. Swelling is the most frequently reported clinical manifestation when symptoms appear; however, pain, tooth displacement, in addition to paresthesia, may also develop. Radiographically, some OKCs appear as well-defined radiolucent lesions; these lesions may be unilocular or multilocular, depending on their size as well as their stage of development.

Clinicians employ several treatment modalities for OKCs, ranging from conservative approaches such as marsupialization along with decompression to more aggressive techniques, like enucleation with or without adjuvant therapy. Surgeons reserve resection for aggressively progressive or frequently recurring cases.[4] Adjuvant therapies, such as Carnoy's solution, cryotherapy or peripheral osteotomy can be done to reduce the risk of recurrence.[4] Several factors have been identified that contribute to the risk of recurrence, including the lesion's location, size and multilocular nature.[8]

OKCs associated with nevoid basal cell carcinoma syndrome (Gorlin syndrome) can cause higher recurrence rates. While uncommon, OKCs can rarely undergo malignant transformation, most often into squamous cell carcinoma. Therefore, careful diagnosis and long term follow up are essential. Signs that might suggest this change include rapid growth, persistent inflammation and pain.[4,5] Incomplete removal of the cyst lining and the presence of daughter cysts are also significant risk factors.[4,8] The recurrence of OKCs is a significant concern, with rates varying depending on the treatment method. Recurrence can occur several years after initial treatment, necessitating regular follow-up to ensure early detection and intervention.[1,4]

The mechanism of recurrence first proposed by Brannon et al suggested three different mechanisms which include incomplete removal, newly growing keratocysts from satellite cysts or development of a new keratocyst in the area adjacent to the site of the primary keratocysts.[9] Several studies have reported male preponderance.[1] The first incidence peaks around the third decade and the second between 50 to 70 years of age. Despite many studies in the literature examining several therapeutic approaches in managing this lesion, it is still not clear which method provides lower recurrence rates without causing significant morbidity.[4]

Topical 5-fluorouracil is known for its anti-proliferative effects on keratocystic epithelium and satellite cysts; furthermore, its use has some advantages, such as technical ease and the lack of neurotoxicity10. The risk factors associated which may cause recurrence of odontogenic keratocyst, include age, multilocularity, lesion size and radiographic characteristics. The various surgical techniques used to excise keratocysts have potential benefits that can be preservation of jaw function, reduction of recurrence, and total eradication of

the cystic lesion. Marsupialization or decompression can be done to minimize surgical.

CONCLUSION

OKCs are challenging lesions with a high recurrence potential. Treatment should be individualized, considering patient age, lesion size, location, and the presence of risk factors. Conservative treatments are preferred for smaller lesions, while more aggressive approaches may be necessary for larger or recurrent cysts. Long-term follow-up is crucial to detect any recurrence or malignant transformation & manage early. Further research, prospective studies and randomized trials are needed to gather more evidence on the effectiveness of different treatment methods and follow-up protocols.

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