Maxillary Odontogenic Keratocyst Presenting as Palatal Sinus

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Abstract Odontogenic keratocyst are known for their recurrence and local bone destruction. They have bimodal age presentation. OKC is common in lower jaw than upper jaw. They are usually asymptomatic and diagnosed during radiological investigation. We are reporting this case because of its age presentation (70 years), site of presentation (maxillary canine region) and mode of presentation (chronic palatal pus discharging sinus).

Keywords Odontogenic keratocyst · Parakeratotic keratocyst · Palatal sinus · Enucleation

Introduction

Odontogenic keratocyst are commonly present in lower jaw. Maxillary region is the rare site of its occurrence. It is common in younger age group though late presentation also reported. They are known for recurrence in spite of complete removal because of presence of residual active epithelium. Radiograph shows a unilocular radiolucency with sclerotic border. Use of Carnoy's solution reduces chances of recurrence.

Case Report

A 70 years male presented with swelling over the upper right side of lip, over the gums and over the palate on the same side since 1 year and pus discharge from the right side of palate since 3 months. There was no history of any

trauma, tooth extraction, recurrent cold, nasal obstruction and epistaxis. There was no history of diabetes, hypertension and TB. On oral cavity examination, there was a pus discharging sinus on the hard palate area on right side (Fig. 1). On palpation there was no underlying bony palate, it was completely destroyed by the disease and pus can express out on pressing the swelling. It was tender on touch. A soft to firm compressive swelling present over the upper gum region from premolar region to the midline. It can be felt up to the floor of the nasal cavity. CT scan showed a well defined cystic mass occupying anterior wall of maxilla from premolar region to the incisor region of opposite side and destruction of palate of the same side (Fig. 2). Mass was pushing inferior turbinate medially. We approached the cyst by sub labial incision (Fig. 1). On aspiration there was straw color fluid came out. Cyst was excised completely with intact wall. Cyst wall was tightly adherent to the underlying palatal mucoperiosteum. Histopathological examination showed the features of odontogenic keratocyst (presence of squamous cells lining) with infiltration of plasma cells, lymphocytes (showing features of chronic inflammation) (Fig. 3). Patient is doing well in follow ups.

Discussion

Odontogenic keratocyst (OKC) of the jaw are developmental cysts arising from cell rests of the dental lamina, the oral epithelial lining of the developing tooth follicle [1]. Some authors consider it a benign cystic neoplasm rather than a developmental cyst [2]. It is common in second and third decade of life and in males (M:F, 1.3:1). Some investigators have reported a bimodal age distribution with an additional peak in the fifth and sixth [3] decades. They

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Fig. 1 **a** Preoperative photograph showing palatal swelling with sinus, **b** Intraoperative photograph showing cyst wall

Fig. 2 CT scan showing well defined mass in the anterior wall of maxilla (**a, b**)

Fig. 3 Histopathology showing epithelium lining with chronic inflammatory cells

represent approximately 4–10% of all odontogenic cysts and have an incidence of 3.5–5 per million populations per year [4]. Body of mandible especially molar region and vertical ramus are the common sites of its occurrence [5]. They are usually single and multiples are associated with Gorlin–Goltz syndrome (nevroid basal cell syndrome) [6]. It is same as other cysts in these regions in presentation but

only recurrence rate is high as compare to other types. Recurrence rate is in range 13–60%, which can be compare with Ameloblastoma [7]. Clinically they are asymptomatic but can present as toothache, swelling and bone destruction. Histologically they are classified as Parakeratotic and Orthokeratotic, depends upon type of keratin production [8]. Orthokeratotic produces keratin similar to normal skin

and does not contain nuclei. It is considered less aggressive as compared to Parakeratotic. Some authors prefer its independent identity. Parakeratotic type is common (80%) and an aggressive cyst as in present case. Its keratin is more disorganized and contains nuclei (Para keratin). OKC has active epithelium. The increased cell activity is evident by the presence of elevated level of oxidative enzymes and acid phosphatase, which indicate high metabolic and lysosomal activities. These cysts are filled with keratinous debris (with the consistency of toothpaste) and this can become infected. Epithelial lining has high mitotic activity and rarely may become dysplastic and develop into squamous cell carcinoma [9]. The following radiographic features are characteristic of OKCs: (1) a distinctly corticated, often scalloped, border; (2) expansion, especially toward the lingual (medial) side, and growth along the length of the bone; (3) displacement of developing teeth and/or separation or resorption of the roots of erupted teeth and extrusion of erupted teeth; (4) a radiolucent lumen, and occasionally a cloudy or milky appearance of the lumen on the panoramic radiograph; (5) the occasional presence of multiple or bilateral cysts, suggestive of basal cell nevus syndrome [10]. The epithelial lining is not as inactive as that of other cysts and there is tendency for these cyst to expand through marrow spaces and hence their multilocular radiological appearance. CT scan shows low attenuation because of low protein content (less than 4 g). Contrast study is required to differentiate hemorrhagic cyst and other vascular malformations. On MR images, an OKC typically has low to intermediate signal intensity on T1-weighted images and high signal intensity on T2-weighted images. This pattern may be caused by fluid containing a low concentration of protein, which results in intermediate signal intensity on T1-weighted images [8]. OKC requires complete removal. Enucleation with or without continuity defect are the treatment of choice. Presence of residual epithelium or an epithelial remnant after enucleation of the cyst and presence of satellite cysts in the cyst's wall are the contributory factors for recurrence. Nowadays, this is not the case as careful enucleation in conjunction with the application of Carnoy's solution reduces recurrence to less

than 10% [11]. During surgery, it is advised to avoid leaving cyst remnants; the bone cavity should be deeroofed to its maximum diameter to ensure adequate access. Post-operative follow-up with regular radiographic examination is important with OKCs because of the potential for recurrence. OKCs usually recur within 5 years after surgery, but they can recur more than 15 years later [12].

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