

Incidental Discovery of Odontogenic Keratocyst in an Edentulous Patient: Importance of Routine Pre-Prosthetic Radiographic Evaluation

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Abstract The most common modality of treatment for completely edentulous patients are conventional removable complete dentures. The protocol for fabrication of complete denture would involve a thorough clinical examination and radiographic investigation. As a routine, unless clinical findings suggest presence of remnant tooth structures, or the patient presents himself with symptoms, the radiographic investigations are overlooked. This case report presents one such situation wherein routine radiographic evaluation of a clinically asymptomatic edentulous individual prior to prosthetic rehabilitation revealed the presence of an odontogenic keratocyst of anterior maxilla substantiating the importance of such routine pre-prosthetic radiographic investigations.

Keywords Odontogenic keratocyst · Anterior maxilla · Young patient · Prosthodontic treatment

Introduction

Conventional removable complete dentures are the most common modality of treatment for completely edentulous

patients. Thorough clinical examination is a routine procedure before prosthetic reconstruction whereas radiographic investigations are restricted only for clinically symptomatic patients. Incidental radiological finding of a cystic lesion in the present case underscores the importance of routine radiographic investigations as a part of prosthetic work up prior to rehabilitation of asymptomatic individuals.

Case Report

A 27-year-old female reported to the outpatient department for replacement of missing teeth. Patient gives history of extraction 2 months back due to generalized mobility. Patient was referred to the department of Prosthodontics for prosthetic rehabilitation. Extra-oral examination was normal. Intraorally, maxillary, and mandibular arch were edentulous (Fig. 1). An implant supported prosthesis was planned for the patient. Orthopantomogram (Fig. 2) was done as a part of routine investigation before prosthetic rehabilitation which revealed the presence of a single, well-defined, ovoid unilocular radiolucency measuring about 4×2 cm in the anterior maxilla crossing the midline in relation to periapical regions of missing anterior teeth extending from 13 to 23 region. Maxillary occlusal view (Fig. 3) was also taken which revealed a similar lesion. A provisional diagnosis of residual cyst was considered and the patient was referred to the department of Oral and Maxillofacial Surgery. Cyst enucleation (Fig. 4) was done under local anesthesia and the specimen was sent for histopathologic examination. Microscopic examination revealed a cystic lumen lined by corrugated, parakeratinised stratified squamous epithelium of 6–8 cell layer thick exhibiting palisading and characteristic tomb-stone appearance of the basal cell layer (Figs. 5 and 6). The fibrous connective tissue

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Fig. 1 Intra-oral view showing edentulous maxilla

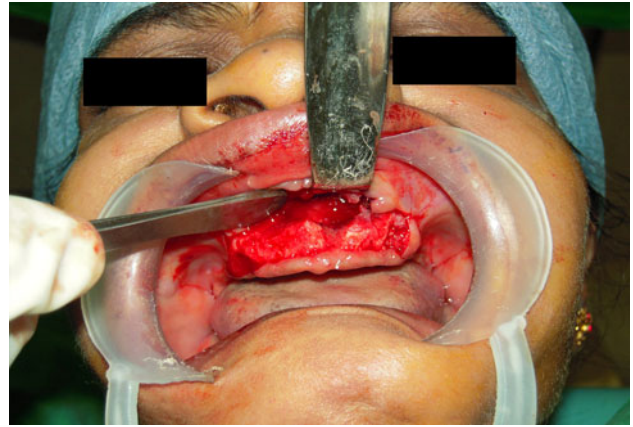


Fig. 4 Photograph showing enucleation of the cyst

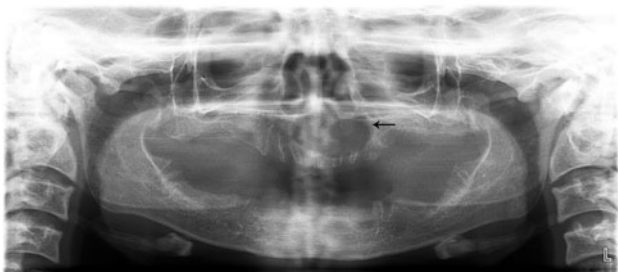


Fig. 2 Orthopantomograph showing well-defined, ovoid, radiolucent area in midline of anterior maxilla

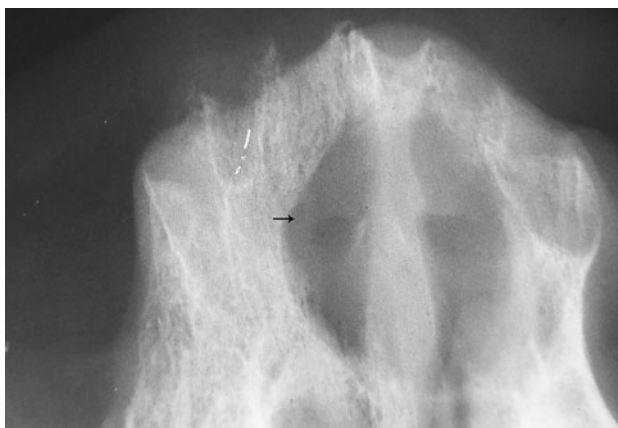


Fig. 3 Occlusal view showing the well-defined radiolucency

capsule showed a few inflammatory cells and extravasation of red blood cells. Based on these features, a diagnosis of odontogenic keratocyst was made. Following the histopathological diagnosis, the cystic cavity was reexplored under local anesthesia for chemical cauterization using Carnoy's solution to prevent recurrence of the lesion. Considering the age of the patient, clinical and radiographic investigations were done to rule out the presence of Nevoid Basal

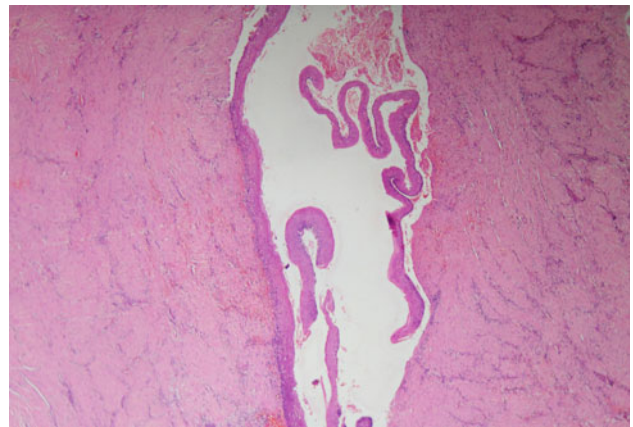


Fig. 5 Photomicrograph showing corrugated, parakeratinised cystic lining epithelium (Hematoxylin and Eosin, $\times 4$)

Carcinoma syndrome. The healing was uneventful. Patient was given an interim prosthesis (Fig. 7) and implant supported prosthesis was planned in a later date.

Discussion

Differential diagnosis of anterior maxillary radiolucencies includes a spectrum of lesions, the most common being residual cyst, nasopalatine duct cyst, globulomaxillary cyst, adenomatoid odontogenic tumour and odontogenic keratocyst (OKC), dentigerous cyst and squamous odontogenic tumour in rare instances. Although, the OKC constitutes only 3–11% of all jaw cysts, it is of great concern to clinicians because of its high recurrence rate, aggressive behaviour, and occasional association with the nevoid basal cell carcinoma syndrome. This syndrome is characterized by multiple basal cell carcinomas, cysts of the jaw, vertebral and rib anomalies, and intracranial calcification [1]. OKC was recently designated by the WHO as Keratocystic odontogenic tumour [2]. Some authors support that it should be considered as a benign

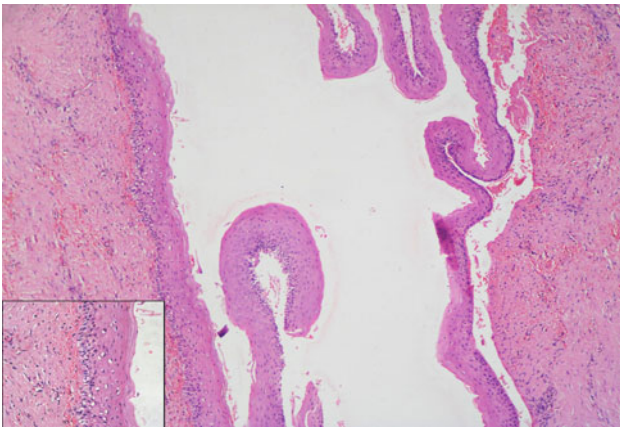


Fig. 6 Photomicrograph showing corrugated, parakeratinised cystic lining epithelium of 6–8 cell layer thickness. (Hematoxylin and Eosin, $\times 10$). Inset shows a tombstone appearance of the basal cell layer (Hematoxylin and Eosin, $\times 40$)



Fig. 7 Intra-oral photograph showing maxillary and mandibular dentures in occlusion

cystic neoplasm, due to its growth capacity and developmental characteristics related to the mutation of a suppressor tumor gene, *PTCH*, found in sporadic and in OKCs associated with Basal cell nevus syndrome [3].

Approximately 65% of OKCs occur in the mandible, with a predilection for the third molar-ramus region. Maxillary OKCs are very rare. 10% of OKCs occur in edentulous patients [4]. Very few cases are reported with OKCs crossing maxillary midline and in all reported cases the patients were of older age group [5–7]. In our case, the patient was a young edentulous female further substantiating the fact that it is important for the clinician to consider OKC in the differential diagnosis of lesions crossing maxillary midline, when they occur in a younger patient also.

Keratocysts seem to follow the way of least resistance and tend to hollow out a mandible, thereby replacing the bone

marrow, rather than giving rise to periosteal bone formation, which would result in a bony swelling [8]. Hence, failure of early and prompt diagnosis of OKCs may lead to further complications like failure of the prosthesis, lack of osseointegration in case of implant supported dentures and the most important being pathological fracture of the jaw as a result of asymptomatic hollowing out of the jaw. In the present case early detection of the cyst during a routine radiographic analysis has prevented such complications thus emphasizing the importance of such routine investigations.

Conclusion

The incidental discovery of the lesion during routine OPG taken before prosthetic treatment emphasizes the importance of such routine investigations prior to prosthetic rehabilitation of clinically asymptomatic individuals to avoid further complications. The rarity of the present case is attributed to its unusual site of involvement, the anterior maxilla, and patient being a young edentulous female in contrast to the usual presentation of OKC. This rare presentation underscores the importance for the clinician to consider OKC in the differential diagnosis of such anterior maxillary radiolucencies, when they occur in younger patients also.

References

1. Hsun-Tau C (1998) Odontogenic keratocyst: a clinical experience in Singapore. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 86:573–577
2. Sembronio S, Alberio AM, Zerman N, Costa F, Politi M (2009) Endoscopically assisted enucleation and curettage of large mandibular odontogenic keratocyst. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 107:193–196
3. Carvalho Silva GC, Carvahio Silva E, Gomez RS, Vieira TC (2006) Odontogenic keratocyst in the maxillary sinus: report of two cases. *Oral Oncol* 42:231–234
4. Chkoura A, Chbicheb S, El wady W (2009) Keratocystic odontogenic tumor: a case report and review of the literature. *Internet J Dent Sci* 6(2)
5. Woo SB, Eisenbud L, Kleiman M, Assael N (1987) Odontogenic keratocysts in the anterior maxilla: Report of two cases, one simulating nasopalatine cyst. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 64:463–465
6. Chiang Y, Chao P, Yang T, Peng B, Lee F (2006) Transnasal endoscopic marsupialisation for a large midline maxillary odontogenic keratocyst in a 6 year old child. *Int J Pediatr Otorhinolaryngol extra* 1:41–44
7. Myoung H, Hong SP, Hong SD, Lee JI, Lim CY, Choung PH, Lee JH, Choi JY, Seo BM, Kim MJ (2001) Odontogenic keratocyst: review of 256 cases for recurrence and clinicopathologic parameters. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 91:328–333
8. Stoelinga PJW (2001) Long-term follow-up on keratocysts treated according to a defined protocol. *Int J Oral Maxillofac Surg* 30:14–25