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Intraosseous Epidermoid Cyst of Jaw: A Case Report

Divya Uppala [1*], U Sivakalyan [2], UDN Sreevalli [1], Buddha Harika Varshita [1]

Mounica Tirumalareddy [3]

1. Department of Oral Pathology and Microbiology, GITAM Dental College and Hospital, India.

2. Department of Oral and Maxillofacial Surgery, GITAM Dental College and Hospital, India.

3. Department of Oral and Maxillofacial Surgery, Vishnu Dental College, India.

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**Corresponding author:*
Uppala Divya

Department of Oral Pathology and Microbiology, GITAM Dental College and Hospital, India.

Tel: +91-9966413710
Fax: +91-9966413710
Email: uppala.divya@gmail.com

ABSTRACT

Epidermoid cysts are benign, rare which can occur all throughout the body of an individual. Their occurrence in the oral cavity is rare and is difficult to distinguish from other intraoral cysts. A 27-year-old female patient presented with a swelling to the oral and maxillofacial surgery department for the removal of the cystic lesion. Based on the clinical and radiographic features the provisional diagnosis was given as odontogenic keratocyst. But the histopathological examination revealed a stratified squamous cystic epithelium with abundant keratin suggestive of an epidermoid cyst. This case report presents an uncommon finding in the oral cavity with a history of teeth extraction. Based on these findings even though epidermoid cyst is rare it should be included in the differential diagnosis of radiolucent lesions of the jaws.

Keywords: Sebaceous cyst; Cytokeratin; Keratohyalin granules; Teratoma; Dermoid cyst.

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Introduction

Epidermoid cyst occurs anywhere on the body, mostly along the embryonal lines of fusion. They constitute about 7% and 1.6% of the oral cavity. They constitute less than 0.01 % of all the oral cysts. They commonly occur on the tongue, lips, palate, floor of the mouth, jaws and interfere with deglutition, speaking and feeding. Clinically presents as a slow growing, asymptomatic masses detected mostly during the second or third decades of life [14]. Extra orally, ECs are seen in the ovaries and sacral region [15]. They have a varied origin. They can arise from the occlusion of the pilosebaceous unit, iatrogenic or surgical implantation of epithelium into the jaw mesenchyme and subsequent cyst formation. Therefore, it is also called as epidermal inclusion cyst. Epidermoid cyst is a developmental non-odontogenic benign cyst [1]. The characteristic feature is a fowl smelling discharge and inflammation is due to the deposition of laminar pattern of keratin, evidenced histopathologically. In this case report an intrabony epidermoid cyst of the ramus of the mandible.

Case Presentation

A 27-year-old female patient presented to the oral and maxillofacial surgery unit with a swelling in the left posterior mandibular region for several weeks. Intraorally the lesion measured 3 x 2cm in size extending from the retromolar region to the first molar region of the left mandible. Radiographically, there is a well-circumscribed, unilocular, radiolucent lesion with a history of extracted second molar. The lesion is not in connection with any of the teeth in the jaw (Figure-1). Based on the clinical, and radiographic features the provisional diagnosis suggests an Odontogenic cyst. On gross examination, it revealed a cystic soft tissue specimen along with numerous small bits of tissues which were pasty and sticky inconsistency with a distinct foul smell (Figure-2). On staining with routine hematoxylin and eosin stains, the histopathological features revealed orthokeratinised stratified squamous cystic epithelium resembling that of the epidermis and fibrovascular connective tissue core. The epithelial cells were arranged in 6-8 layers and the granular layer being prominent. The epithelial cells did not show any palisading or a tombstone appearance. Along with this it also revealed numerous keratin flakes in a laminar pattern. The connective tissue shows dense collagen fibers with few or very few inflammatory cells. And there are no dermal appendages (Figure-3, 4).

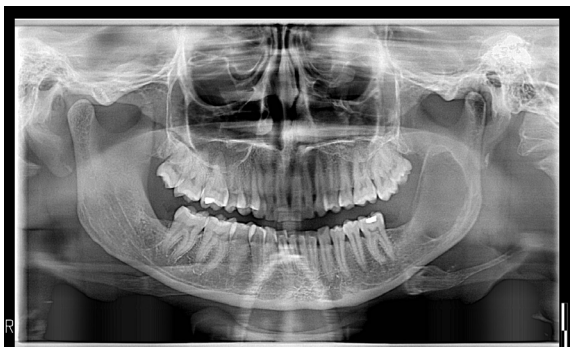

Figure 1. Radiographically, there is a well circumscribed, unilocular, radiolucent lesion with well defined borders extending from third molar region to

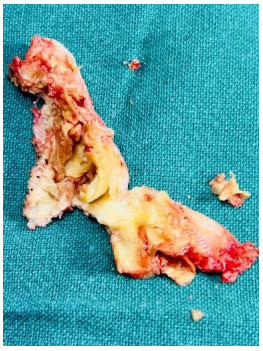

Figure 2. On gross examination the tissue specimen was cystic and soft in consistency.

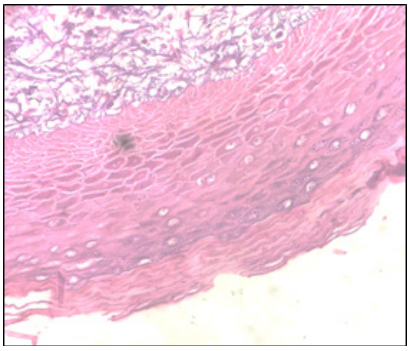

Figure 3. Orthokeratinised stratified squamous cystic epithelium resembling that of epidermis with prominent granular cell layer and fibrovascular connective tissue core. (40x H&E).

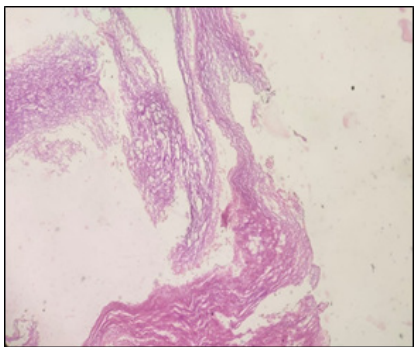

Figure 4. Fig 4: cyst lumen filled with numerous keratin in a laminar pattern. (20x H&E).

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Discussion

Epidermoid cysts are benign soft tissue neoplasms. Occurs before puberty unless associated with Gardner syndrome [8]. Some authors considered it as developmental non-odontogenic cyst [4]. These are non-aggressive [12]. It was first reported by Pinson in 1807 [4]. Represent the simplest expression of teratoma spectrum [9]. The development is of 2 types [8]: congenital and acquired. Congenital cysts result from disturbances during the early stages of embryogenesis between 3rd - 5th gestation weeks [11]. Acquired type was first recognised by Werhner and originally referred to as 'implantation cyst' by Sutton in 1895 [4]. Orcus et al [7] mentioned the theories of the formation of epidermoid cysts. The 1st theory is embryonic ectodermal entrapment which is suitable for congenital cysts that occur at the line of fusion of embryonic arches, especially 1st and 2nd brachial arches, hence their most common sites are midline and sublingual (under the tongue). Lucio et al [5] reported a rare case of congenital epidermoid cyst of the hard palate supports this theory. But this theory doesn't suit for intraosseous epidermoid cysts of jaw as there are no embryonic fusion lines in the mandibular ramus region. Another theory states that the migration of Fordyce granules from the oral mucosa through fistulous tract into bone or due to accidental surgical implantation [7].

In the present case, there is no fistula formation, but patient gives a history of extraction from which there can be possibility of surgical implantation. Jyade et al [6] reported a case of an epidermal inclusion cyst of the mandible after extraction of a third molar supports this second theory. The last theory states that metaplastic transformation of dentigerous cyst epithelium and sebaceous differentiation of cyst wall by the inductive potential of mesoderm [7]. This theory supports the association of epidermoid cyst with impacted tooth. Orcus et al [7] reported a case of bilateral intraosseous epidermoid cyst in association with impacted mandibular third molars. This theory explains the case. Congenital is the most accepted. But traumatic factor is also fairly accepted [8]. Since trauma is said to always precipitate in the formation of the implantation-type epidermoid cyst, King, termed it as 'post-traumatic cyst' [4,12]. Ramakrishna et al [4] reported a case of post-traumatic epidermoid inclusion cyst in chin region. Three etiological factors are responsible for epidermoid cyst formation: [8]

- Inflammation that is localized to the hair follicle (usual etiology).
- Non neoplastic proliferation of infundibular epithelium.
- Traumatic implantation.

Male predilection is more than females. Slow growing painless swelling. Frequently asymptomatic unless secondarily infected [1,2,3]. Epidermoid and dermoid cysts are very rare with an incidence of 7% in the head and neck, and 1.6% in the oral cavity. Most common in orbits with -47%, floor of the mouth –23%, submandibular to upper cervical region -9-24%. [11]. The most common sites in the body are the ovary and anus, intraoral sites also include the tongue, lip, palate, and cheek. Intraosseous cysts commonly occur in the skull and distal phalanges of the hand. Rarely occur in the mandibular premolar and molar regions. Extremely rare in the ramus of the mandible [8,12,13,]. The present case involves ramus. On gross examination, these consist of two main parts: capsule and contents. The capsule is usually elastic, opaque, white, smooth and glistering. The contents of epidermoid cyst consist of thick creamy, cheesy-like material, gray/tannish yellow/brown colour, sometimes have a white soapy/waxy/muddy appearance [4,8,11,13]. Due to these features, they are described as 'pearly tumour' [12]. In a case reported by Jayade et al [6] the gross specimen contained stalk-like pedicle attached to the superior aspect of the lesion extended into the mandible towards the socket of the third molar through a perforation in the inferior border, which gave the lesion the shape of a "dumb-bell". Computed tomography, Ultrasonography, and magnetic resonance imaging modalities are appropriate for diagnosis and treatment [7].

It appears as unilocular radiolucency with well-defined borders, sometimes with sclerotic borders [8]. In the case reported by Jayade et al [6] notching of lower border of mandible was evident without any cystic lesion, as the lesion is located extraosseously. In the case reported by Fuka et al [11] Magnetic resonance imaging revealed a lesion with mixed moderate and high signal intensity on fat-suppressed T2-weighted imaging in the left mandibular ramus. Gadolinium contrast-enhanced T1-weighted imaging showed no clear enhancement. The present case showed well-defined unilocular radiolucent lesion in the ascending ramus region under Orthopantomogram (OPG). Meyer et al, histologically divided these cysts into 3 types [11]:

Epidermoid cyst: no skin appendages.

Dermoid cyst: skin appendages in the connective tissue.

Teratoma-like cyst: skin appendages, connective tissue, respiratory and digestive epithelium. Microscopically, the cyst wall is lined by orthokeratinised stratified squamous epithelium with prominent granular cell layer containing kerato hyaline granules and basal cell layer with low cuboidal to flat cells. The interphase between the epithelium and connective tissue is flat with no rete ridges. The lumen is filled with flakes or laminated keratin [6,8,9,12,13]. Histologically it resembles odontogenic keratocyst (OKC) and orthokeratinising odontogenic cyst (OOC). OKC can be distinguished by the presence of basal cell palisading, reversal of polarity, keratinising epithelium with corrugations and surface parakeratin. It is very difficult to distinguish it from the OOC histologically. Radiographically can be distinguished from OOC by considering the association with the impacted or embedded tooth. But still, there is the possibility of the occurrence of the epidermoid cyst with an impacted tooth [1,2,3].

Padmapriya et al [10] conducted a study and concluded that CK 10 expressed identically in both OOCs and epidermoid cysts in surface and spinous layers. CK19 expression between OOCs & epidermoid cysts and OOCs & OKCs was statistically insignificant. OOCs may not be distinguished from epidermoid cysts both histologically and with CK 10 expression. Indicating that OOCs resemble both epidermoid cysts and OKCs. Our case is not associated with impacted tooth radiographically, thus excluding odontogenic cysts like OKC, and OOC. Histologically there are no skin appendages thus excluding the dermoid cyst. Patient gave history of extraction and during surgery, thus there was a possibility of accidental implantation of epidermoid cells. The surgeon felt foul smell from the cystic contents which are cheesy. Correlating the clinical, radiographical and histopathological features we finally derived a diagnosis of epidermoid cyst. These can be treated by conservative surgical excision [8]. The prognosis is fairly good [8]. Epidermoid and dermoid cysts have a low rate of recurrence (3%) [13,5] mainly observed in cysts located in the floor of the oral cavity [13]. Multiple intraosseous epidermoid cysts are a common manifestation of Gardner's syndrome [13]. Although malignant transformation is a rare occurrence for this type of pathological formation, several cases disclosing malignancy in the head and neck have been well-documented [13]. Heidsieck, however, reported that of 5 cases of epidermoid cysts in the mandible, one relapsed and another one changed into malignancy [12].

Conclusion

Our case is seen in the ramus of the mandible mimicking an odontogenic keratocyst. Therefore, even rare it should be included as a differential diagnosis while diagnosing radiolucent lesions in the oral cavity and further confirmation is done by immunohistochemical analysis, along with clinical history and radiographic findings. They have a good prognosis and are not aggressive. Surgical excision is the treatment of choice.

Conflict of Interest

There is no conflict of interest to declare.

References

[1] William G Shafer, Maynard K Hine and Barnet M Levy, Textbook of Oral Pathology, 9th edition, Elsevier.

[2] Brad W. Neville, Carl M. Allen, Douglas D. Damm, Angela C. Chi, Oral and Maxillofacial Pathology, 4th edition, Elsevier.

[3] Mervyn Shear, Paul Speight, Cysts of the Oral and Maxillofacial Regions, 4th edition, Wiley Black well.

[4] Ramakrishna Y, Sudhindra B, Munshi AK. Post-traumatic epidermoid inclusion cyst in the chin region. J Clin Pediatr Dent. 2009; 33(3):251-252. doi:10.17796/jcpd.33.3.812225u343773155

[5] Montebugnoli L, Tiberio C, Venturi M. A rare case of congenital epidermoid cyst of the hard palate. BMJ Case Rep. 2011;2011:bcr0720114485. Published 2011 Oct 20. doi:10.1136/bcr.07.2011.4485

[6] Jayade BV, Upadya VH, Gopalkrishnan K, Shirganvi MS. Epidermal inclusion cyst of the mandible after extraction of a third molar: case report. Br J Oral Maxillofac Surg. 2012; 50(5):e72-e74. doi:10.1016/j.bjoms.2011.09.017

[7] Toptas O, Akkas I, Tek M, Ozan F, Boran C. Intraosseous epidermoid cyst associated with impacted mandibular wisdom teeth: an uncommon entity. J Clin Diagn Res. 2014;8(7):ZD31-ZD32. doi:10.7860/JCDR/2014/9413.4630

[8] Ertem SY, Uckan S, Ozdemir H. An unusual presentation of an intraosseous epidermoid cyst of the anterior maxilla: a case report. J Med Case Rep. 2014;8:262. Published 2014 Jul 28. doi:10.1186/1752-1947-8-262

DOI: 10.18502/jcr.v11i1.16492

[9] Loxha MP, Salihu S, Kryeziu K, et al. Epidermoid Cyst of Mandible Ramus: Case Report. Med Arch. 2016;70(3):238-240. doi:10.5455/medarh.2016.70.238-240

[10] Padmapriya VM, Kavitha B, Sivapathasundram B, Nagaraj J. Comparison of cytokeratin expressions among orthokeratinized odontogenic cysts, epidermoid cysts and odontogenic keratocysts: An immunohistochemical study. J Oral Maxillofac Pathol. 2020; 24(3):472-478. doi:10.4103/jomfp.JOMFP_243_19

[11] Homma F, Karube T, Usuda S, Endo T, Asoda S, Kizu H. Epidermoid cyst arising in the mandibular ramus: A case report. Int J Case Rep Images 2021; 12:101271Z01FH2021.

[12] Blagova B, Malinova L, Ivanova V. Intraosseous Epidermoid Cyst: a Case Report. J Oral Maxillofac Res. 2022;13(4):e4. Published 2022 Dec 31. doi:10.5037/jomr.2022.13404

[13] Dzhongova E, Mihaylova V, Dimitrov I, Georgieva V. A clinical case report of an epidermoid cyst of the mandible. J of IMAB. 2023 Jan-Mar; 29(1):4770-4774. DOI: https://doi.org/10.5272/jimab.2023291.4770

[14] Puranik SR, Puranik RS, Prakash S, Bimba M. Epidermoid cyst: Report of two cases. J Oral Maxillofac Pathol. 2016; 20(3):546. doi:10.4103/0973-029X.190965

[15] Koca H, Seckin T, Sipahi A, Kazanc A. Epidermoid cyst in the floor of the mouth: report of a case. Quintessence Int. 2007 Jun; 38(6):473-7.