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Odontogenic Keratocyst with Vascular Component - A Case Report

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Abstract: Odontogenic keratocyst(OKC) is a developmental cyst of odontogenic origin. The unique features of the cyst's histopathology make it distinct from other cysts of the jaws. Generally, it presents with no symptoms with the unilocular or multilocular radiographic feature. A 33-year-old male patient with a painless swelling in the lower right posterior region of the jaws was reported to the outpatient department of our college. There was slight facial asymmetry and swelling on the buccal side. The right submandibular lymph nodes were palpable, and OPG showed a well-defined radiolucency. The FNAC showed numerous keratin flecks with RBC, and the inflammatory background was reported as OKC. The gross specimen was blackish brown, with histopathology features quite atypical since numerous endothelial-lined blood vessels of various sizes filled with RBCs were in the connective tissue capsule. Along with the classic elements of OKC, such as stratified squamous para-keratinised epithelial lining with grooves, there was also a rete ridge formation in the epithelial lining, and it was reported as an Odontogenic Keratocyst. The presence of blood vessels can activate the epithelial cells, thus giving rise to rete ridge formations. There are also possibilities for the presence of other vascular malformations. Regardless of recurrences in OKCs, factors that indicate the virulence of the cyst should be considered in the treatment. The multipotentiality of these odontogenic remnants and their reactive response in the surrounding tissues can be varied. Here we discuss a unique case report of OKC in a 33-year-old male patient with many vascular channels in the ramus-molar region of the mandible with an unusual histopathological presentation for OKC.

Keywords: Odontogenic keratocyst, Vascular channels, Aggressive factors, Recurrences

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1. INTRODUCTION

Odontogenic Keratocyst(OKC) has several features that explain the aggressive nature of the lesion. Also, recurrences are frequent over many years and more common in the basal cell nevus syndrome-associated OKCs. Therefore, the cyst may show features of aggressive behaviour. Various histopathological features predict the recurrences to an extent. They include the development of new cysts from the basal layer that shows reverse polarity, satellite cysts, epithelial rests and inflammation.[1,2] The epithelial lining getting separated from the connective tissue capsule is another possible factor along with the increased mitotic activity in the basal and suprabasal cell layers and subepithelial hyalinization for the recurrences. OKC, so far, still needs to give complete satisfaction after the treatment. The characteristic finding of OKC includes a keratin-filled lumen and classic histologic features of a thin layer of stratified squamous para keratinised epithelium, flat interphase between the epithelial lining and connective tissue.[3] Also, a part of the basal epithelium or remnants are frequently found where the OKC is attached to the mucosa. Generally, more common among males in the second to the fourth decade, with a mean of 37 years in the posterior mandible with a specification to the angle and the ascending ramus. They constitute around 11.6% of the total occurrence of jaw cysts. The cyst grows along the length of the bone more than buccolingually, making facial asymmetry challenging to diagnose. OKCs differ tremendously in their behaviour, predicting potentiality related to their origin and various factors influencing their proliferation. However, OKC remains a questionable cyst with no definite conclusion. Some studies try to implicate the clinicopathological features to relate to the recurrences.[4,5] OKC is restored to its previous terminology in the 4th edition of the odontogenic cysts and tumours classification by WHO from odontogenic keratocystic tumours due to the lack of evidence of its true neoplastic nature. Nevertheless, because of the incidence of recurrences, information on the OKC histopathology presentation is essential to understand this peculiar cyst better and treat it accordingly. A large number of blood vessels may increase the proliferative capacity of the epithelial cells.[6,7] Here, we present an unusual case of OKC with blood vessels varying in size.

2. CASE PRESENTATION

A 33-year-old male patient reported to our college with a chief complaint of painless swelling in the lower right posterior region of the jaws. He did not have any relevant medical or family history. The node was small initially but grew to its current size according to the patient's record. He had no other similar swellings or trauma. Besides alcohol consumption, the patient did not have any other harmful oral habits like chewing or smoking tobacco. Extraoral examination revealed slight facial asymmetry on the exemplary aspect of the face. There was a swelling on the buccal aspect on the intraoral examination (Figure 1A). No visible colour changes were noted compared to the adjacent skin on the surface of the swelling. The right submandibular lymph nodes were palpable 4x3.5cm in size and mobile with tenderness. Orthopantomography(OPG) revealed a well-defined radiolucency bordered by a radiopaque sclerotic border. (Figure 1B). The FNAC showed numerous keratin flecks on PAP and H&E staining (Figure 2A&2B), with RBC and inflammatory background, reported as OKC. The gross specimen was unusual for the OKC (Figure 2C). The histopathology was atypical since numerous endothelial-lined blood vessels of various sizes filled with RBCs were in the connective tissue capsule. Along with the classic features of OKC, such as stratified squamous para keratinised epithelial lining with corrugations, there was also a rete ridge formation in the epithelial lining and was reported as an Odontogenic Keratocyst(Figure 3A,3B&3C).

Fig 1A: Clinical picture showing intraoral swelling extending from the distal of the right second molar on the proper posterior aspect of the mandible.

Fig1B: OPG showing a well-defined large radiolucency from the distal aspect of the right second molar to the mesial surface of the left lateral incisor of the mandible bordered by a radiopaque sclerotic border.

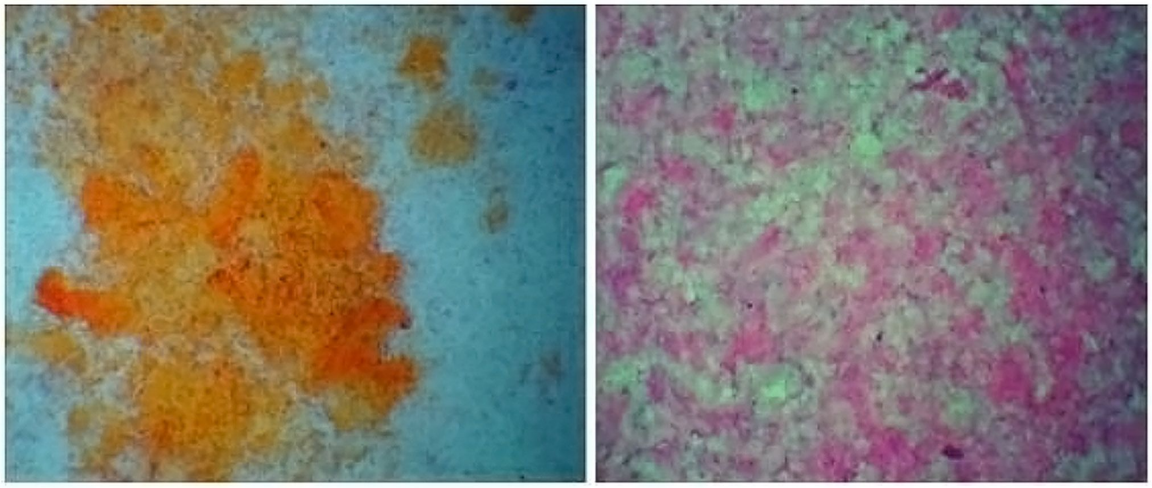

Fig 2A&2B: H&E and PAP smear taken from the aspirated cystic fluid shows flecks of keratin with RBC and inflammatory background (Mag10x)

Fig 2C: Gross specimen firm in consistency, cystic with measuring around 2.5x3.5x1mm and blackish brown.

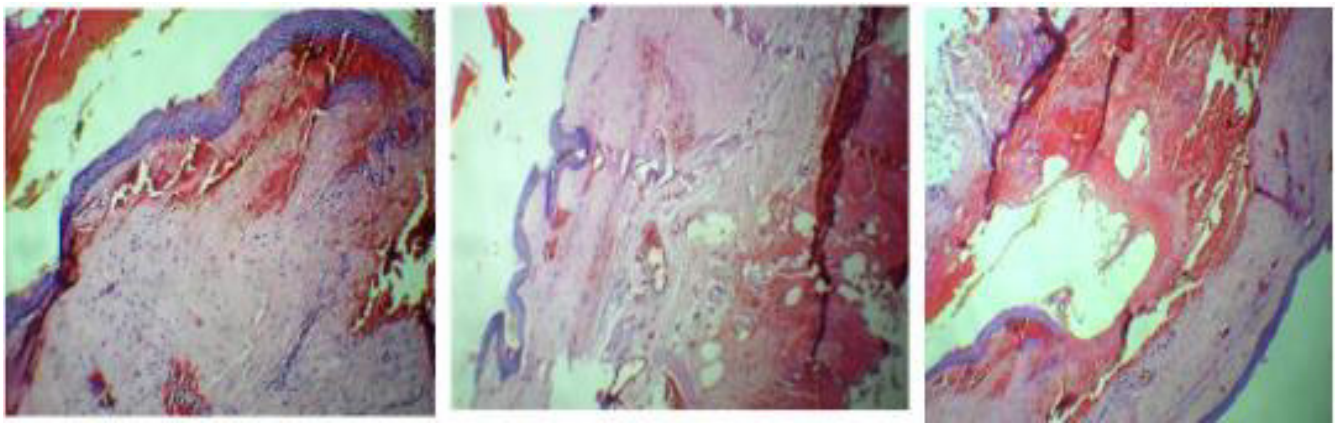

Fig 3A&3B: (Mag4x) & 3C(Mag10x) Photomicrographs of H&E stained sections show para keratinised epithelial lining with many endothelial-lined vascular channels in the connective tissue capsule.

3. DISCUSSION

Neovascularisation in tumours generally indicates the aggressive factor in them. Many literature studies show vascular endothelial growth factor(VEGF) is essential in the pathogenesis and proliferation of OKC. In our case lesion was extensive, starting from the distal of the right second molar to the mesial surface of the left lateral incisor of the mandible. This significant involvement in the anteroposterior direction occupies most of the mandible's body without much noticeable swelling.[8,9] The cystic lumen of OKC contains fluid filled with a cheesy substance of keratinaceous debris. The corrugated para keratinized stratified epithelium lining, thin and uniform thickness, with basal layer, typically consists of a palisaded layer of hyperchromatic cuboidal or columnar epithelial cells. Small daughter cysts, cords, or islands of the odontogenic epithelium may be present within the fibrous wall. Epithelial-mesenchymal interactions are a part of odontogenic cysts and tumour origin since they arise from the odontogenic tissue residuals. The budding and proliferation of the basal epithelium into the connective tissue was mentioned first by Ahlfors et al.[10,11] Angiogenesis is the evidence of connective tissue changes resulting from the accumulation of epithelial lining, indicating stroma upregulation of transforming growth factor-β. The activity of the numerous micro-vessels shows the aggressive local behaviour of OKC. Some studies suggest a relationship between the thrombosis of the blood vessels and capillaries and the proliferation of epithelial cells.[6,12] Presence of blood vessels can be theorised for many other reasons besides angiogenesis. It could have been present as a separate entity or due to any trauma. In our case, the patient did not undergo any extraction or had an incidence of trauma. Huge vascularity can form due to various other reasons, a secondary change, or already existing lesions like hemangioma or hamartoma. It is observed that one of the aetiology is the presence of abnormal proliferation of blood vessels during odontogenesis.[13-15]

4. CONCLUSION

Odontogenic keratocyst controversies have been enduring for many years because of their biological nature. The inductive capacity of cell remnants in the odontogenic apparatus has the potential to form odontogenic cysts and tumours with variations. The formation of blood vessels is part of epithelial mesenchyme interaction though the presence of large blood vessels is unclear; combining two lesions can be possible. Nevertheless, the presentation of OKC does signify the nature of the cyst; thus can lessen the main challenge of recurrences after removal.

5. AUTHORS CONTRIBUTION STATEMENT

R.A. and S.S.V. conceived the case study and were in charge of the overall direction, analysis and planning. R.A. and S.S.V. carried out the implementation. R.A. and S.S.V. took the lead in writing the manuscript. K.P. provided critical feedback and helped final shaping of the manuscript.

6. CONFLICT OF INTEREST

Conflict of interest declared none.

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