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CALCIFYING ODONTOGENIC CYST WITH COMPLEX ODONTOMA- A RARE CASE REPORT

Maxillofacial Surgery

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ABSTRACT

Calcifying odontogenic cyst (COC), also referred to as Gorlin cyst, is a rare benign cystic lesion of odontogenic origin, accounting for 1–2% of all odontogenic cysts. The Gorlin cyst is an atypical and distinctive lesion that exhibits characteristics of both a solid neoplasm and a cyst. It predominantly affects individuals in the second to fourth decades of life and shows a predilection for the anterior regions of the jaws, particularly the maxilla. Clinically, it manifests as a slowly enlarging, painless swelling and may be discovered incidentally on radiographic examinations. The diagnosis hinges on the comprehensive analysis of clinical, radiological, and histological features.We present an unusual case of COC accompanied by odontomas in a 14-year-old male who presented to our Outpatient Department with an asymptomatic left midfacial swelling. The clinical and radiographic findings were suggestive of an adenomatoid odontogenic tumour and a dentigerous cyst. Following careful consideration, the decision was made to enucleate the lesion, and the histopathological examination corroborated the diagnosis of a Gorlin cyst.

KEYWORDS

Calcifying odontogenic cyst, Gorlin cyst, Odontomas, Enucleation

INTRODUCTION

COC, initially delineated by Gorlin et al. in 1962, represents a rare developmental odontogenic lesion, constituting approximately 1–2% of all odontogenic cysts. The WHO classifies it as a benign cystic neoplasm of odontogenic origin, characterized histologically by a cystic epithelial lining featuring ameloblastoma-like basal cells, the presence of ghost cells, and varying degrees of calcification. Although traditionally regarded as a cyst, this lesion may exhibit neoplastic behaviour, particularly in its solid variant known as CCOT.

COC manifests most prevalently during the second to fourth decades of life, exhibiting no pronounced gender bias. There is, however, a slight propensity for occurrence in the anterior regions of the jaws, particularly within the maxilla. These lesions may present as asymptomatic, gradually enlarging swelling, frequently associated with impacted or unerupted teeth. Radiographically, COC is characteristically depicted as a well-defined unilocular or multilocular radiolucency, with or without radiopaque foci, contingent upon the extent of calcification..

This article highlights the rare case of COC associated with complex odontomas in a 14-year-old male child with emphasis on its clinical features, diagnosis and surgical management.

A 14year male reported to department of oral and maxillofacial surgery with a chief complaint of a painless swelling in the anterior region of the maxilla, which had been gradually increasing in size over the past three months. The patient did not report any history of trauma or infection in the region.

On extraoral examination, there was mild facial asymmetry due to a diffuse swelling over the upper lip and nasolabial region. The overlying skin was normal in colour and texture, and no regional lymphadenopathy was noted.

Intraoral examination revealed a well-defined, firm, non-tender swelling extending from the left lateral incisor to the left premolar region (Fig.1). The overlying mucosa appeared normal. There was slight buccal cortical expansion, and the affected teeth were non-mobile.

For further investigation OPG and CBCT were advised. On radiographic examination, it showed well defined mixed radiolucent-radiopaque lesion, roughly ovoid in shape extending Antero posteriorly from left lateral incisor to left 1st molar associated with multiple toothlike structures and impacted 23,25 (Fig. 2). Deflection of lateral incisor root observed.

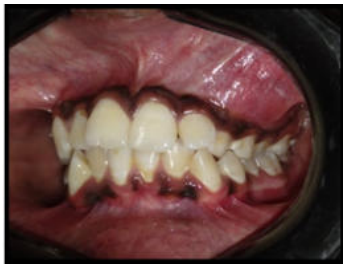

Figure 1 INTRA ORAL CLINICAL PHOTOGRAPH

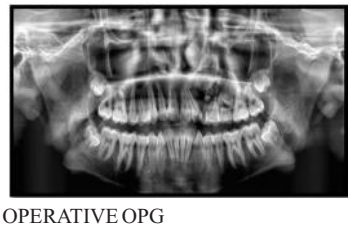

Figure 2 PRE OPERATIVE OPG

On CBCT axial view (Fig. 3), there is buccal cortical expansion. Radiopaque toothlike structures present in the lesion

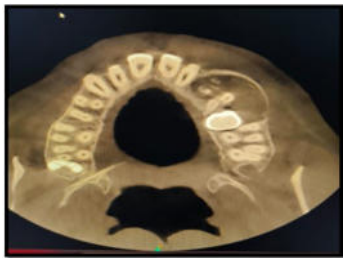

Figure 3 CBCT AXIAL VIEW

Based on these findings, we chose to access the enucleation of lesion in

our case. The patient was prepared under aseptic conditions and local anaesthesia was administered (2% lidocaine with 1:80,000 epinephrine). A crevicular incision was made from the left maxillary lateral incisor to the left maxillary premolar with vertical releasing incisions to provide adequate access. A full-thickness mucoperiosteal flap was elevated to expose the underlying bony lesion. Upon exposure, a well-demarcated, expansile lesion was identified in the anterior maxilla.

Figure 4 EXPOSURE OF CYSTIC LESION

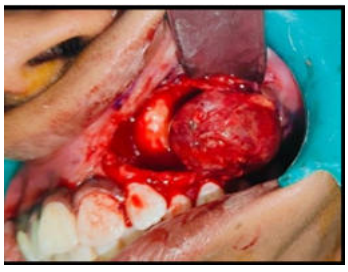

Figure 5 ENUCLEATION INTOTO

Buccal bone overlying the lesion was removed carefully using a round bur under copious irrigation to expose the cystic lesion (Fig. 4) and associated calcified structures.

Enucleation of the cystic lesion was performed in toto (Fig. 5) taking care to preserve the integrity of the surrounding anatomical structures. Multiple hard tissue masses were noted within the cystic cavity, which were removed and preserved for histopathological examination. The involved unerupted tooth maxillary canine and retained deciduous molar were extracted.[1]

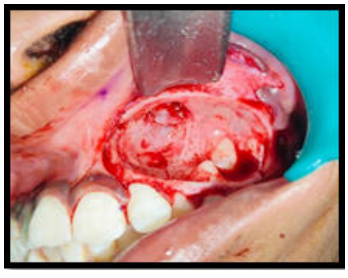

Figure 6 POST OPERATIVE PHOTOGRAPH AFTER ENUCLEATION

The surgical site was thoroughly irrigated with sterile saline, and haemostasis was achieved (Fig.6). The flap was repositioned and sutured with 3-0 vicryl using interrupted sutures. The patient was prescribed antibiotics, analgesics, and antiseptic mouthwash, and was instructed to maintain oral hygiene. Postoperative healing was uneventful. The patient was followed up at regular intervals to assess healing and monitor for any recurrence. The sample was sent for histopathologic examination. (Fig.7,8)

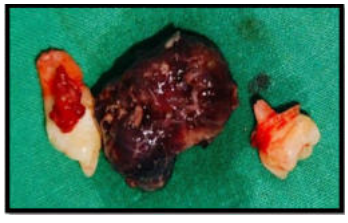

Figure 7 EXCISED LESION

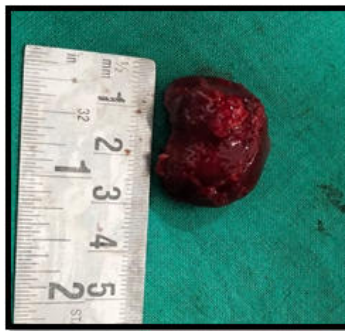

Figure 8 GROSS SURGICAL SPECIMEN

On histopathological examination it shows cystic cavity lined by cystic epithelium of cuboidal to columnar basal cells with reversal of polarity of nucleus and superficial stellate cells resembling stellate reticulum with prominent aggregates of amorphous eosinophilic ghost cells across, prominent foci of dentinoid with lamellar calcifications. The decalcified section shows disorganized calcified mass resembling dentinoid, cementoid, focal area of tubular dentin, enamel rods with intervening connective tissue and aggregates of ghost cells. It is suggested of COC with complex odontomas.

DISCUSSION

COC is an exceptional lesion that exhibits characteristics of both a solid neoplasm and a cyst, thus it was delineated as a distinct entity by Gorlin. The majority of these lesions are non-neoplastic. Alternative terminologies employed include dentinogenic ghost cell tumour, CCOT, and atypical adamantinoma. COC manifests a diverse array of clinical, pathological, and histological attributes. It bears clinical and radiological similarities to other lesions such as CEOT, ameloblastic fibro-odontoma, AOT, dentigerous cyst, and other odontogenic cysts. Radiographically, in the early stages, the lesion appears entirely radiolucent; as it matures, calcifications develop, resulting in a mixed radiopaque and radiolucent lesion. The presence of calcifications is a particularly distinctive finding in COC. Some researchers have noted that a common and significant radiological result of COC was root resorption of neighbouring teeth's roots. [2]

The incidence of enveloped teeth in COC is approximately 32%. According to Praetorius et al., the intraosseous variant of the cystic lesion tends to erode the bone rather than induce expansion, with 75-77% of the lesions leading to root resorption due to their involvement with the apices of the teeth. Treatment typically involves surgical enucleation and curettage. The prognosis for patients with COC is favourable, with only a limited number of recurrences reported. When a COC coexists with other odontogenic tumours such as ameloblastoma, the treatment regimen and prognosis remain consistent with those of the associated tumours.

Enucleation is the preferred intervention for central cystic lesions; however, occasional recurrences have been documented in a minority of cases. En bloc resection, accompanied by meticulous and extended follow-up, is the recommended management protocol for neoplastic COC. The present case was surgically enucleated, and a six-month follow-up revealed no recurrence. The patient continues to be monitored.

CONCLUSION

COC is an atypical developmental odontogenic cyst that clinically and radiologically mimics other more prevalent jaw pathologies. The lesion has consistently been a subject of ambiguity regarding its dual nature, leading to a multitude of nomenclatures and classifications. Due to its nonspecific clinical and radiographic characteristics, histopathological assessment remains pivotal for accurate diagnosis. Prolonged follow-up is recommended to mitigate the risk of recurrence.

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