Hybrid odontogenic tumor of calcifying odontogenic cyst, ameloblastic fibroma, and complex odontoma: A rare case presentation

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ABSTRACT

Mixed odontogenic Tumor comprise two or more structurally different components.They comprise of more than two contrasting lesions are extremely rare and often pose a significant diagnostic challenge. Gorlin cyst is a rare developmental odontogenic cyst with locally pugnacious nature and have been reported to occur in association with other odontogenic lesions like Miscellaneous ameloblastic tumor, odontoma, AOT, ameloblastoma, Epithelio-mesenchymal ameloblastoma and myxomateous odontogenic tumor. Herein, we present a case of Mixed odontogenic tumour of Gorlin cyst, complex odontoma, and miscellaneous ameloblastic tumour occurring in the left mandibular region of a young female patient.

Keywords

Hybrid odontogenic tumor, Calcifying odontogenic cyst, Ameloblastic fibroma, Complex odontoma

INTRODUCTION

Mixed odontogenic lesion (MOL) is the term pertinent to certain lesions that combine the tissue characterstics of more than two kinds of known cysts that have primary site which is mostly similar [1]. The clinicoradiographic presentation of these lesions may range from non-invading cysts or hamartomatous lesions to benign or malignant neoplasms [2]. These lesions often present a dilemma in terms of diagnosis and treatment due to the fact that their histogenesis, although common, remains controversial and clinical features poorly understood. A hybrid odontogenic lesion consisting of more than two disparate lesions is extremely rare.

It is the most commonly reported hybrid odontogenic lesion is calcifying odontogenic cyst (COC), a rare entity accounting for less than 2% of all odontogenic lesions, often combined with odontoma [1,3]. COCs predominantly occur in individuals within the age group of 20-40 years in the incisor and canine region [4]. These lesions

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demonstrate a locally assertive clinical behaviour and unveil a variousness of clinical symptoms along with histological characteristics ranging from a cystic contusion to a solidified tumor [5].The morphological development of denti in the neighbouring connective tissue can be induced by the epithelial lining of COC and its intermingling with odontoma is rather common [4].Gorlin cyst is being delineated to occur alongwith other mixed tumours such as ameloblastoma, ameloblastic fibrodontoma, adenomatoid odontogenic tumor, ameloblastic fibroma, odontoameloblastoma, and calcifying epithelial odontogenic tumor [6].

Herein, we present a case of hybrid odontogenic tumour of COC, complex odontoma, and ameloblastic fibroma in the left mandibular region of a young female patient. To the best of our knowledge, very few cases of more than two disparate odontogenic lesions occurring together have been reported and only one other case of the aforementioned combination has been previously documented.

Case Report

A female patient aged 8 years had visited to the outpatient division of Kalinga Institute of Dental Sciences with the complaint of a painless swelling in the left lower jaw region from past 3-4 weeks. The patient did not report any neurosensory disturbances in the lower lip or chin area.There is nothing attributing as far as past medical, personal and family history of patient is concerned . General physical examination of the patient was unremarkable.

Extra-oral examination did exhibit features of facial asymmetry. It was predominantly due to the presence of a solitary, diffuse swelling on the left side of the face extending from the parasymphysis region to angle of the mandible (Figure 1). The swelling was roughly oval like in contour, nearly 2 cm × 2 cm in dimension . It had smooth surface and poorly defined margins. No visible pulsations or secondary changes were evident. The swelling was non-tender, hard in consistency, non-reducible, and non-compressible. Intra oral examination revealed a diffuse swelling extending from permanent mandibular left lateral incisor to primary mandibular left second molar region with slight vestibular obliteration. The uniformity of excrescence was firm alongwith non-palpable tenderness showing compression. None of the mixed dentition teeth in the affected area exhibited mobility.

Three-dimensional cone beam computed tomography imaging was performed to accurately determine the extent of the lesion. Imaging revealed a single locus radiolucency with borders clearly defined and radiopaque flakes. That radiolucent area measured 23.2 mm mesiodistally and 20.8 mm buccolingually. Expansion of both buccal and lingual cortical plates was evident. In addition, perforation of lingual and buccal cortical plates was seen in relation to 74, 75, and unerupted 34, 35, 36 (Figure 3).

On the basis of clinical and radiographic features,its provisional diagnosis was mention COC. Hard tissue odontoma and adenomatoid odontogenic tumor were considered as differential diagnoses. Aspiration biopsy yielded clear straw-coloured fluid. An excisional biopsy was performed in the left lower vestibular region in relation to 73, 74, and 75 (Figure 2), and the lesion was removed in toto with adequate margin. The biopsy specimen was placed in 10% formalin and sent for histopathological evaluation. The specimen was found to have an elastic consistency during grossing. The overall swelling of the cystic cavity was 0.5 cm thick. The sample was routinely processed, and sections were stained using hematoxylin and eosin stains. In addition, flecks of calcified mass along with some portion of the excised specimen was subjected to decalcification and then observed under the microscope.

Microscopic examination revealed a connective tissue capsule composed of parallelly arranged collagen fibres lined by 8-10 cell layers thick epithelial lining demonstrating basal cell palisading and numerous ghost cells. The basal cells were ameloblast-like tall columnar cells exhibiting reversal of polarity, while the other cells resembled stellate reticulum. Several ghost cells were found to be conglomerated and showed evidence of calcification in the epithelial lining. The connective tissue capsule also showed scattered ghost cell conglomeration with evidence of calcification. Few multinucleated giant cells were seen adjoining the ghost cells. Few epithelial islands were also evident. Diffuse infiltration of chronic inflammatory cells, chiefly lymphocytes, along with numerous endothelium-lined blood capillaries and extravasated red blood cells was also discernible. Decalcified tissue section showed the features of an odontoma, i.e., a large mass of tooth tissue. The structure showed dentinal tissue covering the inner layer of enamel, followed by cementum and pulpal tissue deposited in an abnormal arrangement

(Figure 4).

The final diagnosis of mixed tumour of Gorlin cyst, Mixed Ameloblastic tumour and complex odontoma was made based on histopathological features. The patient was followed up for 90 days and there was no evidence of recurring signs and symptoms.

DISCUSSION

Mixed Odontogenic tumours showing the presence of two or more odontogenic lesions affecting the same primary site, are negligible. These lesions have been reported to have a female predilection and predominantly occur in young adults, with an average age of 24.5 years [1, 7]. However, our patient was only 8 years old, which is an uncommon age for the presentation of this lesion.

Several possible mechanisms have been suggested to explain the pathogenesis of HOLs. These include coalition of two or more distinct lesions, transformation and induction of one or more lesions simultaneously[8]. Despite the various suggested pathogeneses, no consensus has been reached upon. In the present case, ameloblastic fibroma-like areas were seen in the connective tissue wall of COC in proximity to the epithelium in majority of the areas. Thus, it may be considered that AF was induced by the epithelium of COC [4]. In addition, it can also be contemplated that the odontoma resulted from budding of extra odontogenic epithelial cells derived from the dental lamina.

Pontes et al. performed a systematic review and meta-analysis on Mixed odontogenic tumour to analyse their epidemiological/clinical features and biological behaviour. They reported that the mean age of occurrence was 24.9 years and that the lesion showed a female predilection. COC associated with odontoma was the most common HOL. The lesions commonly presented as asymptomatic swellings and most commonly affected mandibular premolars and molars. Radiographic presentation was that of unilocular radiolucent lesions with well-defined borders. Recurrences rarely occurred and were associated with central giant cell granuloma. Less aggressive surgical approaches seemed to the treatment of choice [1]. Clinical presentation and radiographic findings of our patient were similar to those summarized in this study except the age of patient.

Raza et al. reported a case series of HOLs and concluded that most HOLs affected young females in the second decade of life, commonly showed cemento-ossifying fibroma and ossifying fibroma as hybrid components, and that a conservative approach to management was adequate [9]. Our case findings were not in concordance with those of this case series.

Several cases of hybrid COC and ameloblastic fibroma have been reported previously [4]. To the best of our knowledge, only one other case report of hybrid odontogenic tumor of calcifying odontogenic cyst, ameloblastic fibroma, and complex odontoma has been reported [10]. This lesion occurred in a young male patient in the right maxillary region and invaded into the right maxillary sinus. The clinical presentation and lesional behaviour of this patient differed from ours.

Conservative management of the lesion is usually the treatment of choice for HOLs [1]. The case in discussion,its lesion was excised more in an invasive manner because ofthe presence of mixed ameloblastic tumour, and the patient has been followed for 90 days without any signs of recurrence.

CONCLUSION

HOLs are rare lesions showing combined histopathological characteristics of two or more odontogenic cysts/ tumors. Herein, a mixed tumor of Gorlin cyst, ameloblastic fibroma, alongwith complex odontoma was detected in an 8 year old female patient, which is an uncommon age for the presentation of this lesion. Excision of the lesion completely with regular follow-ups.

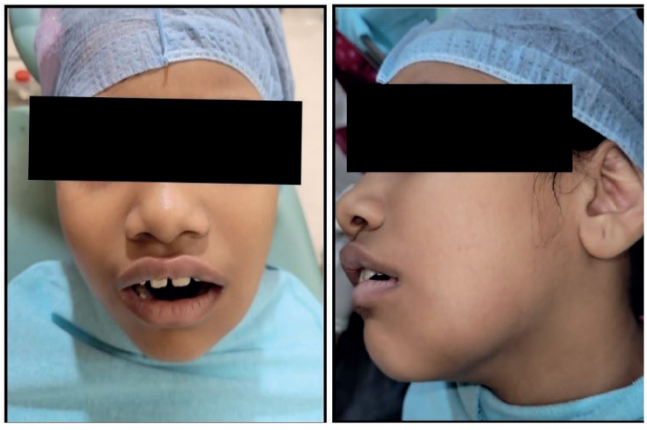

Figure 1: Extraoral photograph of the patient revealing the presence of a solitary, diffuse swelling on the left side of the face extending from the parasymphysis region to angle of the mandible.

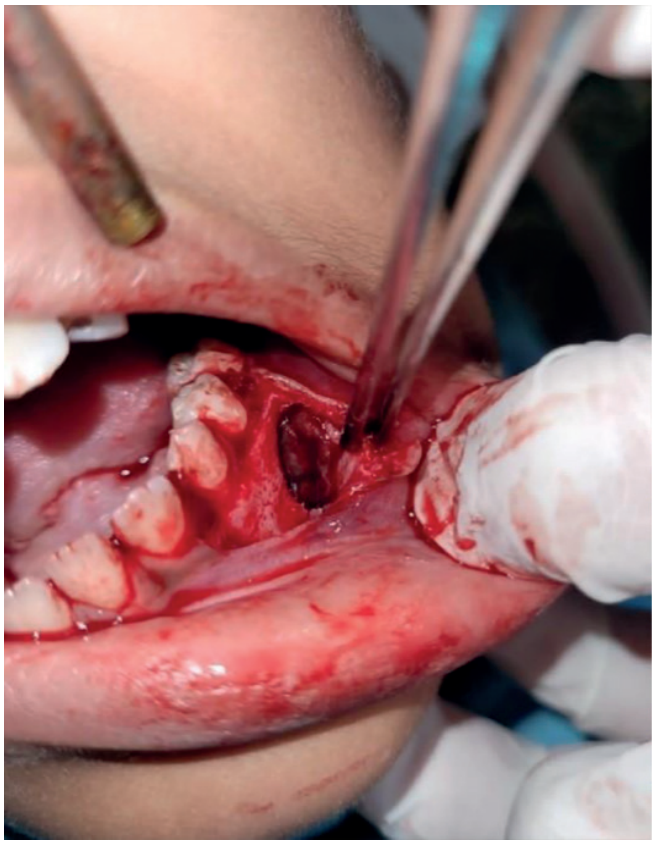

Figure 2: Intraoral photograph showing the surgical site. Lesion was removed in toto with adequate margin from the left lower vestibular region in relation to 73, 74, and 75.

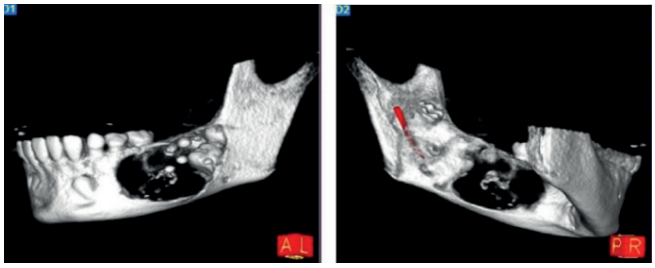

Figure 3: Three-dimensional cone beam computed tomography images revealing unilocular radiolucent lesion with well-defined borders and radiopaque flakes. Perforation of buccal and lingual cortical plates was evident in relation to 74, 75, and unerupted 34, 35, and 36.

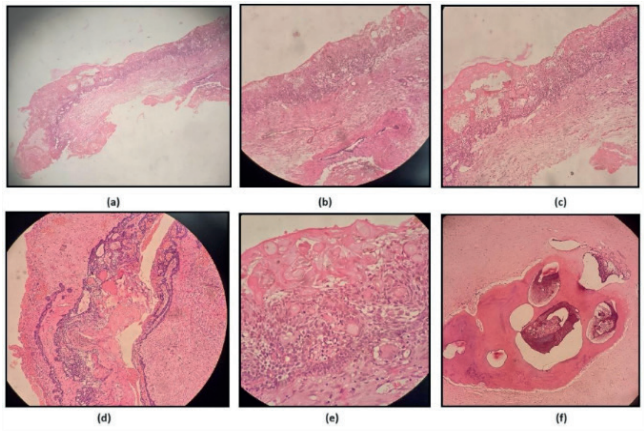

Figure 4: Histopathological findings of the lesion. (a) Low-power photomicrograph (4X) revealing areas representing calcifying odontogenic cyst, ameloblastic fibroma, and odontoma. (b-d) Photomicrographs showing COC lined by ameloblastomatous epithelium with well-defined cuboidal basal ells and suprabasal stellate reticulum-like cells (10X). (e) Several conglomerated ghost cells are seen with evidence of calcification in the epithelial lining along with a few multinucleated giant cells (40X). (f) Photomicrograph revealing complex odontoma with dentin, enamel matrix, and intervening stroma (10X).

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