KERATOAMELOBLASTOMA - A RARE HYBRID ODONTOGENIC TUMOR
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Abstract
The hybrid odontogenic neoplasms are rarities. The histologic features of these tumors are often identical to other individually well established odontogenic neoplasms. Numerous odontogenic cysts and tumors originate from the dental lamina. Therefore, a possibility of neoplastic change, in these ameloblastomatoid islands cannot be ruled out completely. The clinical presentation of these tumors is variable, ranging from cysts to neoplasms showing varying degrees of aggressive behavior. Most combined tumors contain features of one of the odontogenic tumors in combination with other odontogenic tumor. We present a rare hybrid lesion of an ameloblastoma with odontogenic keratocyst arising from the mandibular molar region in a 25-year-old male.

Keywords: Ameloblastoma, odontogenic keratocyst, hybrid lesion.

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Introduction
Tumors and cysts arising from the odontogenic apparatus present a considerable histopathological diversity because of the complexity and prolonged process of Odontogenesis. In 1971, The World Health Organization (WHO) published the first internationally agreed classification of odontogenic tumors and cysts, entitled Histological Typing of Odontogenic Tumours, Jaw Cysts and Allied Lesions, which included proposed criteria for designation and a standard nomenclature. During the interim period, new variants and entities have undoubtedly emerged; therefore, extensively the revised second edition provided an updated classification which is now considered the most reliable scheme.1,2 In the oral maxillofacial region, odontogenic Keratocyst & ameloblastoma are well recognized whereas hybrid odontogenic tumours giving characteristic of both have been rarely reported.

Case report
A 25 year old male patient, reported to the department of Oral Medicine and Radiology, with a chief complaint of pain and swelling on left side of face since two months. On eliciting history of present illness, the swelling was spontaneous in onset and gradually increased and attained the present size. It was associated with dull and intermittent pain. Patient had visited a private dentist one month back and underwent extraction of teeth in lower left back region but the pain and swelling on left side of face did not subside. The medical, family and habit history were non-contributory. Extra oral examination revealed, a diffuse swelling seen on the left side of the face extending medio-laterally from ala of nose to 1 cm medial to tragus of ear and superio-inferiorly extending from ala tragus line to inferior border of mandible measuring approximately 5 x 4 cm in dimension. There was no pus discharge or sinus.

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tract seen. The overlying skin was of normal color with smooth surface. On palpation, it was soft to firm in consistency and tender. It was non compressible, non reducible, non fluctuant and has smooth surface. The overlying skin was pinchable. (Figure 1) Intraoral examination revealed edentulous space in relation with 36 (Figure 2).

Based on history and clinical examination a provisional diagnosis of a Residual cyst was given. As there was minimal expansion, dentigerous cyst, odontogenic keratocyst Odontogenic myxoma and Traumatic bone cyst were considered as differential diagnosis. Patient was subjected for investigations; aspiration was done using a 23 gauge needle under local anaesthesia, in the region of edentulous space of 36. A blood tinged yellowish colored aspirate was obtained, which revealed cholesterol crystals when viewed against sunlight (Figure 3).

For further investigation, panoramic radiograph was made which showed a solitary, well defined unilocular radiolucency in periapical area of 37, 38 and extending into the left side of ramus approximately 4x3cm in dimensions. Internal structure was radiolucent with corticated borders. The distal drifting of 37 and migration of 38 in ramus of mandible region was seen with resorption of roots of 37. (Figure 4) Radiographic differential diagnosis of Odontogenic Keratocyst, Ameloblastoma, Traumatic bone cyst and Odontogenic myxoma was made.
Patient was subjected for further investigations, incisional biopsy was done and section was given for the histopathological evaluation. The histopathological examination showed a cystic lumen lined by parakeratinized stratified squamous epithelium with a fibrous capsule. The epithelium was 5-6 layers thick with surface corrugations. The basal cells were columnar with a palisaded nuclei and reversal of polarity. The junction between epithelium and connective tissue was flat suggesting a fragile attachment and at certain points there was separation of the epithelial lining. The capsule showed loosely arranged collagen fibers. These are classical features of Odontogenic Keratocyst. However, in one area of capsule, there were islands and cords of odontogenic epithelium. These cords were terminating in Peripheral Island like masses morphologically resembling dental lamina. The epithelium was proliferating within the cystic lumen shows stellate reticulum like cells. (Figure 5)

Figure 5. H & E stained section shows 5-6 layer thick parakeratinized stratified squamous epithelium with hyperchromatic nuclei. At few places epithelium is proliferating within cystic lumen shows stellate reticulum like cells.

These represent histological features of ameloblastoma. The histopathological diagnosis of Odontogenic Keratocyst showing ameloblastomatoid features was given.

The patient was advised routine blood investigations and was treated with Marsupialization. Antibiotics Cefotaxime 100 mg twice in a day was given to prevent postoperative infection. Patient was kept under observation with periodic recall of one month. During this period swelling totally reduced.

Discussion

It has been demonstrated that ameloblastoma and odontogenic keratocyst (OKC) show similar clinical characteristics. The simultaneous occurrence of ameloblastoma and OKC was first described by Siar and Ng under the name kerato-ameloblastoma (KA). It consisted of an admixture of solid and cystic components. The solid nests resembled follicular ameloblastoma with pronounced keratinization while the cystic areas bore features of OKC. The solid tumors were in direct continuity with cysts. Invasive growth into the surrounding bone was evident. KA is exceptionally rare and only 9 cases have been identified. Clinical features of KA are as follows: 1) average age was 35 years, ranging from 26 to 57 years; 2) seven tumors were located in the mandible and two in the maxilla; and 3) no gender predominance was evident. The clinical findings in the present case are consistent with those of both ameloblastoma and OKC. Norval et al. suggested that KA should be considered as a variant of acanthomatous ameloblastoma. However, the following findings argue against this possibility. The acanthomatous type of ameloblastoma was seen in significantly older patients (average age 51 years). Histologically, tumor follicles in acanthomatous type were surrounded by a peripheral layer of columnar cells which fulfill the Vickers and Gorlin (VG) criteria for ameloblastoma, and they contained central stellate reticulum like structures. On the other hand, most solid nests of KA never exhibited these pathognomonic features. The cystic component of KA is not analogous to the parenchymal cyst of a conventional ameloblastoma that shows the classic VG criteria. The two types of KA exist, ameloblastoma- like KA nearly akin to papilliferous KA and OKC- like KA.

Our case showed characteristics of OKC with ameloblastic changes in some portions in epithelial lining giving diagnosis of OKC like KA.

Conclusion

Odontogenic tumors and cysts can occur at any stage of odontogenesis. The same
embryonic derivation supports the close interrelationship of several odontogenic lesions; therefore, it is not surprising that hybrid tumors do exist. However, there is a striking histological resemblance between these Odontogenic tumor islands and dental lamina. Herein, we are presenting case of hybrid lesion of OKC with ameloblastic transformation which supports this hypothesis and is a very rare variant.

Declaration of Interest

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References