

Synchronous Occurrence of Bimaxillary Odontomas with Cystic Adenomatoid Odontogenic Tumour of Mandible – A Case Report with Unusual Presentation

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ABSTRACT

Introduction: Odontogenic epithelium undergoes multiple arrays of differentiations during different stages of tooth development. Its alteration may lead to the formation of pathological entities like odontogenic hamartomas, cysts or neoplasms. Odontomas, the commonest one, represent hamartomatous growths of abortive tooth development. Adenomatoid odontogenic tumor (AOT), a benign lesion of odontogenic origin has uncertain histogenesis. It shows biphasic characters of both solid and cystic forms, stemming various opinions regarding its true nature. Synchronous bimaxillary odontomas with mandibular cystic AOT is the unique feature of this case.

Case Presentation: A 56 years old male, with swellings involving both the jaws was provisionally diagnosed by anatomical, clinical and radiological evaluations, as multiple odontomas and dentigerous cyst. Histological correlation led to the diagnosis of synchronous bimaxillary odontomas with mandibular cystic AOT.

Management and Prognosis: All the odontomas were completely excised and enucleation of the cystic lesion done. The patient is under one-year postoperative follow-up without any complications.

Conclusion: All four quadrants of the jaws are involved, three having odontomas and cystic AOT in the remaining one. This indicates severe dental dysmorphogenesis, making it an extremely rare phenomenon. Report of this novel combination is aimed as a future source of reference, as to the best of our knowledge, it's a previously undescribed entity in the literature.

Keywords: Adenomatoid odontogenic tumor, cystic, odontoma, synchronous.

INTRODUCTION

Odontogenesis involves the complex interaction between neuroectodermally derived epithelium and ectomesenchymally derived connective tissue. The spectrum of changes is subtle but results in significant differentiation during odontogenesis. Failure of differentiation at any point may lead to the formation of a hamartoma, cyst, or neoplasm.¹ Odontomas and adenomatoid odontogenic tumors (AOT) are frequently encountered hamartomatous growths of odontogenic origin with varied clinico-radiological and histopathological presentations.^{2,3}

This rare case highlights their synchronous occurrence, as they are mostly reported individually. To the best of our knowledge, it is the first reported case with bimaxillary involvement of odontomas along with mandibular cystic AOT.

CASE PRESENTATION

A 56-year-old male reported to Dr. R. Ahmed Dental College and Hospital with two swellings in the anterior region of both jaws for a duration of one year. The medical, family and past dental histories were non-contributory.

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Intraoral examination revealed a smooth swelling in the maxilla extending from 14 to 24 regions, crossing the midline. (Figure 1) Retained root of 62 was seen with absence of 11 and 21. In the mandible, a diffuse swelling extending from 31 to 36 regions was noted, obliterating the vestibule. (Figure 2) The patient reported that 31, 32, 34, and 35 had been extracted a month ago, owing to mild pain.

Preoperative orthopantomography (OPG) showed the

presence of an ovoid unilocular radiolucency extending from 32 to 36 regions, having impacted 33 in it, pushed to the lower border of the mandible. The presence of unerupted 44, 45, and 86 could be noted with an irregular radiopaque mass, obstructing their eruption paths. Within the maxilla, numerous incongruous miniature tooth-like radiopaque masses were observed extending from 14 to 24 regions, with impacted 11, 13, 21, 22, and 23. (Figure 3)

With the provisional diagnoses of multiple odontomas and a dentigerous cyst, complete surgical removal of all was done. The tumors located in the anterior region of the jaws revealed pulp like tissues with sheets of dentinoid and cementoid like areas, under the microscope. (Figure 4) The left mandibular lesion revealed the presence of a cystic lumen lined by non-keratinized squamous epithelium, with intraluminal nodules comprising rosettes, ducts and solid patterns of hyperchromatic epithelial cells. (Figure 5,6) A rare diagnosis of synchronous cystic AOT with multiple odontomas involving all quadrants of the jaws was made.

DISCUSSION

Odontomas comprise the most frequently occurring odontogenic hamartomas, accounting for 35-76% of all

odontogenic jaw lesions.² Belonging to the spectrum of 'developmental malformation- a hamartoma', its exact etiopathogenesis is still unknown.^{2,4}

The WHO has classified odontomas into two categories: the compound and complex types, which have affinities for the anterior maxilla and posterior mandible, respectively. It usually affects people in the 2nd to 3rd decade of life, with equal gender distribution.^{2,4}

Compound odontomas radiologically appear as a collection of tooth-like structures, or 'odontoids', surrounded by a thin radiolucent zone.² It can be frequently associated with impacted teeth, most commonly the canines, followed by maxillary central incisors and third molars.⁵ Complex odontomas exhibit irregular aggregations of calcified radiopaque masses.^{4,5} In the present case, findings consistent with multiple anterior compound odontomas were noted.

The histopathological analysis of a compound odontome may reveal orderly differentiation of dental tissues, contrary to a complex odontoma, which shows a haphazard conglomeration of dentinoid, cementoid, and pulp like tissues. They are usually managed by enucleation.^{2,4}

AOT was first reported by Steensland et al. in 1905 as 'epithelioma adamantium'.⁶ In 1915, Harbitz coined the



Fig. 1: Non ulcerated smooth swelling present in maxilla extending from 14 to 24 regions, crossing the midline



Fig. 2: A diffuse swelling extending from 31 to 36 regions was noted, with obliteration of regional vestibule



Fig. 3: Ovoid unilocular radiolucency extending from 32 to 36 regions having impacted 33, pushed to the lower border of mandible; maxilla showed numerous incongruous miniature tooth like radioopaque masses, extending from 14 to 24 regions

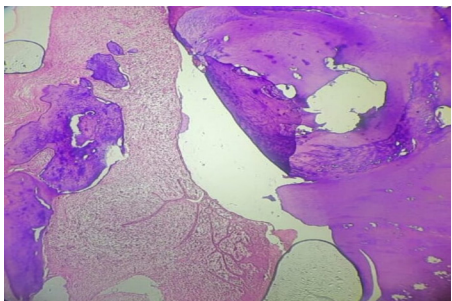


Fig. 4: Areas resembling pulp like tissues with sheets of dentinoid and cementoid materials. (H & E 10x)

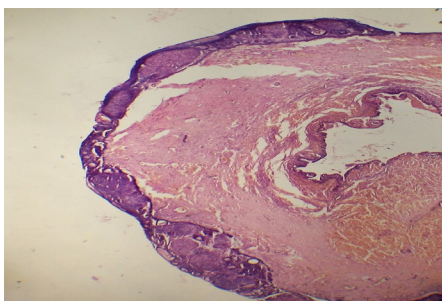


Fig. 5: Cystic lumen, with a thick fibro collagenous capsule; the cystic lining showing nodules of hyperchromatic epithelial cells projecting towards the lumen (H & E -4x)

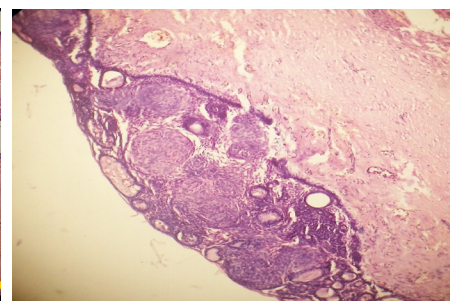


Fig. 6: Thick fibro collagenous capsule, lined by nodules of tightly packed rosettes, ducts and solid pattern of hyperchromatic epithelial cells (H & E -10x)

tumor's cystic presentation as 'cystic adamantoma'.^{6,7} The WHO incorporated the term 'Adenomatoid odontogenic tumor', as proposed by Philipson and Birn, in 1971.⁸ It has been extensively described by Marx and Stern, as a cyst rather than a tumor, with its new terminology being 'adenomatoid odontogenic cyst' (AOC). According to them, it is an odontogenic cyst arising from Hertwig's epithelial root sheath that possesses nodular projections towards the lumen. These fill it partially or completely, imparting a solid appearance.⁶ Gadewar et al. (2010) have illustrated a comparative evaluation of the demographics of 'classic AOTs vs. cystic AOTs', due to the biphasic nature of the odontogenic epithelium.^{6,8} Accordingly, the present case reinforces the cystic variant of AOT, considering its demographic and histological similarities. Further studies are invited for proper elucidation of entities like AOTs with cystification, cystic AOTs, and AOCs.

AOT, comprising 2-7% of all odontogenic tumors, generally involves the maxilla of young women and is associated with an unerupted tooth, mostly canine.³ The present subject was a 56-year-old male with impacted 33 associated AOT, which is again an infrequent demographic finding.

AOT may present itself in intraosseous (central) and extraosseous (peripheral) forms. The intraosseous variant is further subdivided into follicular and extrafollicular variants, corresponding to the presence or absence of a tooth, respectively. The radiological hallmark that differentiates AOT from dentigerous cysts is a well-defined, unilocular radiolucency surrounding an impacted tooth beyond the cemento-enamel junction, as evident in this case.^{6,7}

Histologically, the lesions may be entirely solid, partly cystic, or extensively cystic in nature. The cuboidal, columnar, or spindle shaped hyperchromatic, polarized epithelial cells may be arranged in ductal, glandular, rosette, solid, or whorl like patterns with accumulations of central eosinophilic materials, referred to as 'tumor droplets'. The tumor nodules with this architecture in the cystic lining appeared to be projecting or jutting out toward the lumen.^{6,7,8}

The reported association of dentigerous cysts with AOTs or AOTs with odontomas highlights the tumor's complex biology.⁹ Synchronous odontogenic tumors are rare findings, and a compilation of 30 instances has been done recently by Bruna

Luisa et al.¹⁰ It states that synchronous presentation of the same type of lesions is more common, which further emphasizes the rarity of the presented case as it involves two different entities.

CONCLUSION

Individually, the presence of multiple odontomas, or AOTs, has been frequently reported. The synchronous occurrence of multiple odontomas and cystic AOT together, involving all four quadrants of the jaws, is an extremely unique phenomenon. With a novel approach of histological identification and unique confirmation of the multiple lesions, surgical enucleations were carried out in all quadrants. This illuminates the extent of odontogenic dysmorphogenesis, which justifies the goal of this paper as to add this rare entity to the literature.

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