Dental Research Journal

Case Report

Rare case of bilateral complex odontoma associated with mandibular bicuspids

Rangeeth Bollam Nammalwar[1], Joyson Moses[1], Sathya Jeeva[2]

[1]Departments of Pediatric Dentistry and [2]Oral Pathology, Thai Moogambigai Dental College and Hospital, Chennai, Tamil Nadu, India

Received: July 2016
Accepted: September 2017

Address for correspondence:
Dr. Rangeeth Bollam Nammalwar,
Just for KIDDS Dental Clinic, 3, Seetha Nagar Main Road, Nungambakkam, Chennai - 600 034, Tamil Nadu, India.
E-mail: dr_rangeeth@yahoo.com

ABSTRACT

Among oral pathologic lesions, odontomas are the most common odontogenic tumor of the jaws and generally asymptomatic. They are mostly diagnosed as a result of routine radiographic investigation. In this report, we describe a very rare occurrence of bilateral complex odontoma in the region of the mandibular second premolar. Conservative surgical procedure was planned, and removal of the odontomas was carried out through the socket after extraction of the deciduous second molars, and the diagnosis was confirmed with a histopathological study.

Key Words: Odontoma, odontogenic tumors, radiopaque, Tooth, Unerupted

INTRODUCTION

Odontogenic tumors derive from the tooth-producing tissues or their remnants that remain entrapped either within the jawbones or into the adjacent soft tissues. From a biological point of view, some of these lesions represent hamartomas with varying degrees of differentiation, whereas the rest is benign or malignant neoplasm with variable aggressiveness and potential to metastasis.[1] The classification approved at the Editorial and Consensus Conference held in Lyon, France (WHO/IRAC) in July 2003 has placed odontoma under tumors of containing odontogenic epithelium with odontogenic ectomesenchyme with or without dental hard tissue formation.[2] A histopathological study on a South Indian population showed that odontogenic tumors constituted 4.13% of 11,843 registered biopsies, mandible was affected in 74.02% cases, and odontoma was 7.77% of all cases.[3]

Odontomas are generally asymptomatic and constitute casual findings in the course of routine radiological studies, particularly in the second and third decades of life.[4,5] Complex odontomas present as an irregular mass and compound odontomas present similar to that of normal teeth, with both being distinct radiological entities.[6] Case reports have shown that odontomas can be associated with other lesions such as dentigerous cyst[7] and can rarely erupt into the oral cavity and can give rise to moderately serious conditions, particularly in the presence of over infection of the lesion. Despite their benign nature, eruption into oral cavity can give rise to pain, inflammation, and infection. A search for case reports on bilateral complex odontoma revealed one report in a Swiss (CD-1) mouse,[8] bilateral odontoma involving both the mandible and maxilla in an adult,[9,10] and bilateral complex odontoma in a 4 years old as a result of neonatal mandibular

Access this article online

Website: www.drj.ir
www.drjjournal.net
www.ncbi.nlm.nih.gov/pmc/journals/1480

How to cite this article: Nammalwar RB, Moses J, Jeeva S. Rare case of bilateral complex odontoma associated with mandibular bicuspids. Dent Res J 2018;15:220-3.

distraction therapy.[11] In this report, we present a rare case of child patient with bilateral complex odontoma in the region of the mandibular second premolar.

CASE REPORT

A 12-year-old male child patient born out of nonconsanguineous marriage was brought to the Department of Pedodontics and Preventive Dentistry, with the chief complaint of unaesthetic appearance due to irregular placement of teeth by the parents. The clinical examination revealed retained mandibular deciduous second molars. A panoramic radiograph revealed two radiopaque irregular structures in the mandible bone similar to crowns of teeth in relation to distal roots of the left and right second deciduous molars and in distal aspect relation to the crown of the second premolars slightly [Figure 1]. Intraoral periapical radiographs showed the calcifications in relation to the distal root of the second deciduous molars with a slight lingual orientation and resorption of the distal roots. A provisional diagnosis of complex odontoma was considered and management included extraction of the deciduous second molars to allow the permanent premolars to erupt, followed by exposure of the complex odontoma through the socket itself [Figure 2] while carefully preserving the permanent second premolar. The excisions were planned 2 days apart starting with the right side, and both procedures were done under local anesthesia with pathology being removed conservatively through the extraction socket itself following which the surgical sites were sutured with 3-0 Vicryl suture material and the patient was advised review after 1 week. Both the right and left odontomas were elevated through the sockets and the tissue covering them were curetted and sent for histopathological examination [Figure 3]. Histopathology of the calcified structures under ×40 magnification revealed poorly formed dentin matrix [Figure 4a] and cementoid areas with lacunae containing cementocytes [Figure 4b], consistent with complex odontoma. The excised surrounding soft tissue with the odontoma showed a poorly differentiated benign lesion composed of irregularly arranged fibrocollagenous tissue, with the cellular fibrous matrix that was containing fibroblasts and portions of the odontogenic epithelium [Figure 4c]. Although a 3-month follow-up was present, the patient has been referred for long-term follow-up in his native place.

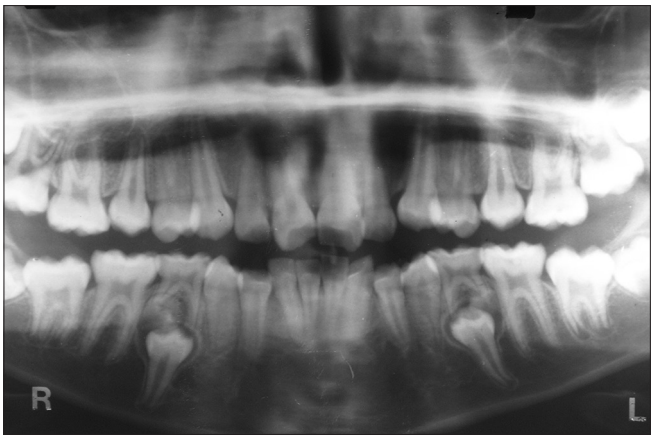

Figure 1: Panoramic radiograph revealing bilateral calcified irregular mass in the region of the second premolars.

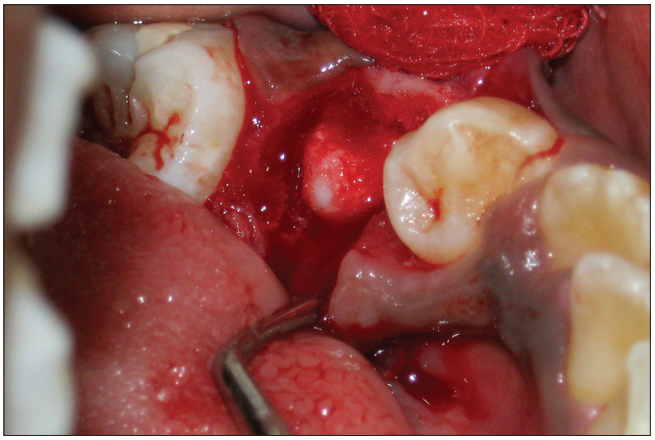

Figure 2: Odontoma in the left side of mandible being excised through the socket itself.

DISCUSSION

Our patient reported with the chief complaint of unaesthetic appearance of teeth; a routine radiographic evaluation exposed the presence of this lesion. Since the patient was not associated with any syndromes or systemic illness such as cleidocranial dysostosis, Garner's syndrome, Herman Syndrome, the provisional diagnosis of odontoma was made and an excision was carried out.. Case reports have shown that male patients are more likely to be diagnosed with odontomas than female patients and mostly seen in the 11–20-year age group with Caucasians predominating and the maxilla being more frequently affected with compound odontomas.[12] Although the etiology is unknown, several theories described such as local trauma, infection, family history, and genetic mutation. Genetically, they may be inherited from a mutant gene or interference, possibly postnatal with the genetic control of tooth development. Several

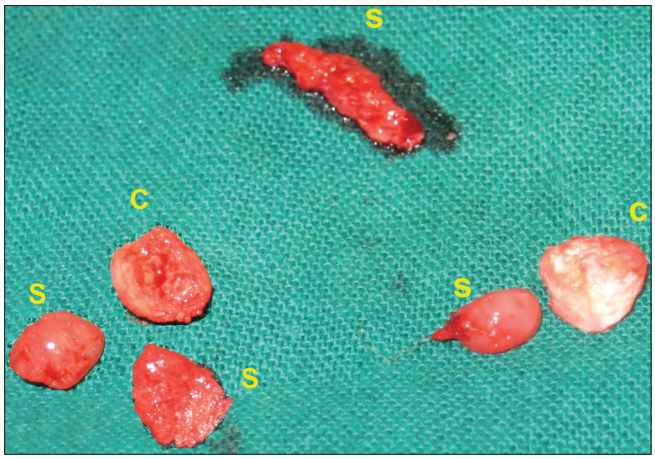

Figure 3: Odontomas (C) excised from both sides of the mandible with soft tissue (S) covering the odontoma.

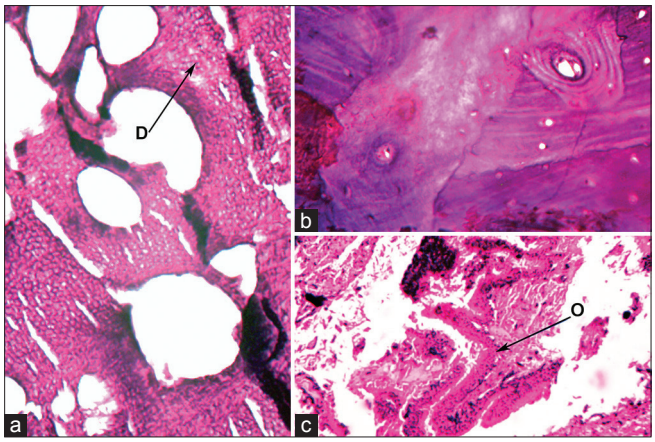

Figure 4: Photomicrograph of decalcified hematoxylin- and eosin-stained section reveals (×40). (a) Poorly formed dental tubules (D); (b) cementoid areas with lacunae containing cementocytes seen; (c) fibrocollagenous tissue, with the cellular fibrous matrix containing fibroblasts (o).

factors may cause anomalous tissue development in odontomas such as unsuccessful or an altered ectomesenchyme interaction in the earliest phase of dental germ development and/or alterations in the subsequent phases of the development of these tissues.[13]

Histological features show complex odontomas as primary or immature dentin as the predominant component, though enamel is also found, exhibiting two possible types of distribution. In a highly calcified area close to the central core or in a hypocalcified zone with immature enamel, immature cement was also observed with the external connective tissue capsule surrounding the lesion.[14] A clinicopathological study revealed enamel matrix in 90%, dentin in 100%, cementum in 88%, pulp tissue in 96%, fibrous capsule in 93%, ghost cells in 83%, reduced enamel epithelium in 86%, and nests of odontogenic epithelium in 58% of odontomas.[15] Similar to this study, we had also encountered the fibrous tissues and odontogenic epithelium in our histopathological evaluation of the excised specimen.

A review of studies that have shown bilateral occurrence has shown that it was initially proven to occur in CD-1 mice induced under laboratory conditions.[8] Most cases reported are adult patients such as a case report of a 41-year-old female patient with bicortical expansion and displacement of teeth. The extent of the lesion warranted a computed tomography scan which depicted a hyperdense mass hear the floor of the maxillary sinus and mandible.[9] Another case of a 20-year-old female patient reported with coronoid hypoplasia and complex odontoma in the mandible along with missing teeth.[10] A 4-year-old female patient diagnosed with Pierre Robin syndrome was diagnosed with bilateral complex odontomas in the posterior aspect of the mandible. The patient had a history of mandibular distraction which highlights the fact that externally placed distractors carry a potential for injury to internal structures of the mandible; therefore; the theory of local trauma inducing odontoma formation may be substantiated with this evidence.[11]

CONCLUSION

The bilateral occurrence of complex odontoma is a very rare condition, and it was diagnosed as a matter of routine radiographic diagnostic process. Detection and removal of odontomas, mostly a very simple process, is always advised as there is a high risk of complications. Despite their benign nature, these lesions are well recognized by their growth pattern and should be completely removed to avoid secondary complications, with an excellent prognosis following surgery. This case report also emphasizes the need for routine radiological investigation for the early management of conditions that can disrupt the harmony of the dentition or oral cavity as a whole.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients

understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship
Nil.

Conflicts of interest
The authors of this manuscript declare that they have no conflicts of interest, real or perceived, financial or nonfinancial in this article.

REFERENCES

1. Mosqueda-Taylor A. New findings and controversies in odontogenic tumors. Med Oral Patol Oral Cir Bucal 2008;13:E555-8.
2. Imran A, Jayanthi P, Tanveer S, Gobu SC. Classification of odontogenic cysts and tumors - Antecedents. J Oral Maxillofac Pathol 2016;20:269-71.
3. Gupta B, Ponniah I. The pattern of odontogenic tumors in a government teaching hospital in the Southern Indian state of Tamil Nadu. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2010;110:e32-9.
4. Philipsen HP, Reichart PA, Praetorius F. Mixed odontogenic tumours and odontomas. Considerations on interrelationship. Review of the literature and presentation of 134 new cases of odontomas. Oral Oncol 1997;33:86-99.
5. Amado Cuesta S, Gargallo Albiol J, Berini Aytés L, Gay Escoda C. Review of 61 cases of odontoma. Presentation of an erupted complex odontoma. Med Oral 2003;8:366-73.
6. Neville BW, Damm DD, Allen CM, Oral and Maxillofacial Pathology. 4th ed. Philadelphia, PA: WB Saunders; 2016. p. 53674-5.
7. Sales MA, Cavalcanti MG. Complex odontoma associated with dentigerous cyst in maxillary sinus: Case report and computed tomography features. Dentomaxillofac Radiol 2009;38:48-52.
8. Dayan D, Waner T, Harmelin A, Nyska A. Bilateral complex odontoma in a Swiss (CD-1) male mouse. Lab Anim 1994;28:90-2.
9. Srivastava A, Annaji A, Nyamati SB, Singh G, Shivakumar G, Sahana S, *et al*. Complex odontoma in both the jaws: A rare case report. J Orofac Res 2012;2:56-60.
10. Dar MA, Alaparthi R, Yalamanchili S, Santosh AB. Bilateral coronoid hypoplasia and complex odontoma: A rare concurrence of developmental pathology and odontogenic tumour of the mandible. BMJ Case Rep 2015;2015. pii: bcr2015212022.
11. Hammoudeh JA, Kleiber GM, Nazarian-Mobin SS, Urata MM. Bilateral complex odontomas: A rare complication of external mandibular distraction in the neonate. J Craniofac Surg 2009;20:973-6.
12. Owens BM, Schuman NJ, Mincer HH, Turner JE, Oliver FM. Dental odontomas: A retrospective study of 104 cases. J Clin Pediatr Dent 1997;21:261-4.
13. Kaugars GE, Miller ME, Abbey LM. Odontomas. Oral Surg Oral Med Oral Pathol 1989;67:172-6.
14. Miki Y, Oda Y, Iwaya N, Hirota M, Yamada N, Aisaki K, *et al*. Clinicopathological studies of odontoma in 47 patients. J Oral Sci 1999;41:173-6.
15. Chang JY, Wang JT, Wang YP, Liu BY, Sun A, Chiang CP. Odontoma: A clinicopathologic study of 81 cases. J Formos Med Assoc 2003;102:876-82.