

A Rare Case of Maxillary Compound Odontoma with an Unusual Large Number of Denticles

Adel Sharifi Rayeni¹, Sahand Samieirad^{2,3}, Vajiheh Mianbandi⁴,
Nasrollah Saghravanian³, Elaheh Tohidi⁶

¹Post Graduate, Oral And Maxillofacial Surgery Department, Mashhad Dental School, Mashhad University of Medical Sciences, Mashhad, Iran

²Dental Research Center, Mashhad University of Medical Sciences, Mashhad, Iran

³Oral And Maxillofacial Diseases Research Center, Mashhad University of Medical Sciences, Mashhad, Iran

⁴Student of Dentistry, Student Research Committee, Mashhad Dental School, Mashhad University of Medical Sciences, Mashhad, Iran

⁵Dental Research Center, Mashhad University of Medical Sciences, Mashhad, Iran

Received 12 December 2018 and Accepted 28 January 2019

Abstract

Odontoma is the most frequent odontogenic gnathic tumor, which is considered to be a hamartomatous lesion rather than an actual odontogenic tumor. Odontoma is basically composed of enamel and dentin, while it could also have various contents of cementum and pulp tissue. These lesions may manifest in two specific forms, including the compound type with multiple, small, tooth-like structures and the complex form with amorphous calcified masses. The early detection, management, and surgical enucleation of odontomas are recommended to prevent tooth eruption disturbances and further pathological complications. The present study aimed to describe a rare case of compound odontoma in the anterior maxilla with an unusually high number of denticles in a 19-year-old Iranian female patient. 62 denticles were extracted presenting a unique case of odontoma, compared to the previously reported subjects with less number of denticles. In addition, the clinical, radiographic, and histopathological aspects, as well as the other possible systemic disorders and selected management approach, were discussed.

Keywords: Compound Odontoma, Maxilla, Denticles.

Sharifi Rayeni A, Samieirad S, Mianbandi V, Saghravanian N, Tohidi E. A Rare Case of Maxillary Compound Odontoma with an Unusual Large Number of Denticles. *J Dent Mater Tech* 2019; 8(2): 101-6.

Introduction

Odontomas are mixed odontogenic tumors defined as developmental anomalies and hamartomatous lesions caused by the growth of differentiated epithelial and mesenchymal cells (1-3). The mentioned structures may undergo functional differentiation to constitute enamel, dentin, cement and pulp tissue (1, 2, 4).

Odontomas are the most frequent odontogenic gnathic tumor, representing about 22% of all odontogenic tumors (1, 3, 5). They are classified as compound and complex tumors. Compound odontoma is a malformation in which all the dental tissues are in a well-ordered pattern pertaining to small teeth, whereas complex odontoma constitutes an irregular mass in an indistinct pattern (1, 3, 6). It is notable that odontomas are slow-growing and often non-aggressive in nature (1, 3, 7). However, the early diagnosis and surgical enucleation of these hamartomas are preferred to prevent tooth eruption disturbances (1-3, 5, 8).

To the best of our knowledge, the number of denticles found in compound odontomas has been reported to be 4-37 in the previous studies in this regard (1, 3, 9, 10). The present study aimed to describe a rare case of maxillary compound odontoma with a high number of denticles (n= 62) in a 19-year-old Iranian female patient, which has not been published in the literature previously. In addition, the clinical, radiographic, and histopathological aspects of the tumor, as well as the

possible systemic diseases and selected management approach, were discussed.

Case Presentation

A 19-year-old female was referred to our center due to slight swelling on the right side of the anterior maxilla. The patients had no history of systemic diseases or pain in this area. Intraoral examination showed numerous carious teeth due to poor oral hygiene. Moreover, a well-defined, hard, bony, non-tender swelling was observed on the labial and palatal sections with normal-colored and consistent overlying gingiva (Fig. 1-a).

The medical and family history of the patient was unremarkable, and the patient had no paresthesia and pain. No other abnormalities were detected intraorally, and there were no extraoral swelling or cervical lymphadenopathy. Laboratory tests were normal in these cases, and no history of pus or blood from the lesion was noted.

Radiological examinations indicated the presence of a dense mass from the central incisor to the first premolar on the right side of the maxilla. Additionally, an impacted mesiodens was observed in the palatal midline. The lesion had caused the distal movement of the lateral, canine, and first premolar teeth, along with a hard osseous expansion in the buccal area of the right anterior maxilla. Furthermore, the radiographic view revealed a radiopaque lesion without a radiolucent rim in the alveolar region of the right anterior maxilla which led to buccal, nasal floor, and sinus expansion. On the other hand, cone beam computed tomography (CBCT) images showed the extensiveness of the tumor. Numerous denticles were also detected in the CBCT of the lesion, which occupied a space of 3×4 centimeters from the alveolar ridge to the sinus floor (Fig. 1-b).

According to the clinical and radiographic discoveries, the patient was provisionally diagnosed with compound-composite odontoma. Specialists agreed upon extracting the mesiodens and surgically enucleating the tumor. Upon obtaining informed consent and explaining the surgical procedure and the lesion type to the patient in compliance with the ethical codes of our university, the surgical operation was performed under general anesthesia, alongside left nasal intubation. Local

anesthesia was performed using two carpules of 2% lidocaine with epinephrine 1/80000 (Persocaine-E, Darupakhsh, Tehran, Iran). A sulcular incision was made in the buccal and palatal regions. In addition, the releasing incisions were used on the buccal side in order to elevate the mucoperiosteal flap for better access to the tumor. Via intraoral access and the elimination of the lesion surrounding the bone, the lesion was divided into smaller pieces and removed.

After the discrimination of the pieces, 62 abnormal, multi-shaped denticles were detected with their fibrous capsule; the denticles were smaller than normal teeth with diverse sizes. The size of the denticles was within the range of 2-20 millimeters. In addition, fusion and dilacerations were observed (Fig. 2), and the mesiodens and infectious and unattainable roots of the right lateral maxillary and canine were extracted as well.

Under light microscope evaluation with the magnification of 40 X, the sections showed miniature teeth containing dentin with dentinal tubules, pulp, acellular cementum, and periodontal ligament-like tissues (Figs. 3-a, 3-b). During decalcification, the enamel cap on the denticles was dissected. Most denticles indicated a single root along with a root canal (Figs. 3-a, 3-b). Analyzing the clinical, radiological, and histopathological discoveries, we reached a explicit diagnosis of compound odontoma.

After the removal of the odontoma, the maxillary defect was reconstructed with an autogenous corticocancellous graft from the anterior symphysis. Additionally, an alloderm membrane (Kish Allograft Inc., Tehran, Iran) was inserted into the defect in order to cover the maxillary sinus floor before the placement of the graft particles. Finally, the buccal and palatal flaps were sutured using Vicryl 3.0 suture (Vicryl, Ethicon, Johnson and Johnson, UK) with no tension (Figs. 4-a, 4-b).

The healing process was uneventful, and the patients completed a six-month follow-up without clinical or radiographic problems (Fig. 5). Furthermore, no sign of recurrence or complications was detected in the follow-up sessions.



Figure 1. a) Pre-operative Clinical View of Patient, b) Pre-operative CBCT View of Numerous Maxillary Odontoma Tumors



Figure 2. Unusual Number of Compound Odontomas

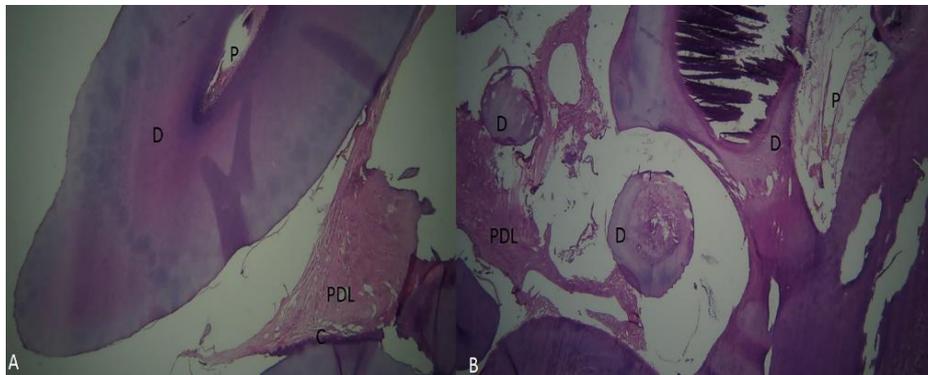


Figure 3-a, 3-b. Histopathology View (40X Magnification; D: dentin, P: pulp cavity, PDL: periodontal ligament, C: cementum)



Figure 4-a, 4-b. Surgical Reconstruction of Maxillary Defect after Odontoma Removal



Figure 5. Postoperative Panoramic View of Reconstructed Maxillary Defect

Discussion

Odontomas, asymptomatic lesions, are usually incidentally discovered during routine radiography (2). These lesions are known as the most frequent odontogenic gnathic tumors, constituting approximately 22% of all odontogenic tumors (3, 11).

Rather than actual neoplasms, odontomas are regarded as hamartomatous lesions or malformations (1-3, 7). Basically, odontomas are composed of enamel and dentin with various amounts of cementum and pulp tissue (6, 10). The term odontoma was first used by Broca in 1867 in a broad sense to refer to the tumors of the odontogenic origin (1, 6).

The World Health Organization (WHO) in 2005 categorized odontomas as two types of complex and compound (1, 2, 11). Complex odontomas are defined as tumor-like malformations (hamartoma) containing enamel, dentin, and occasionally cementum. On the contrary, compound odontomas are defined as the tumor-like malformations (hamartoma) with a various number of tooth-like structures known as denticles (1, 2, 6, 11). These odontogenic tumors can develop anywhere in the dental arches. The majority of the odontomas located in the anterior region of the maxilla are of the compound type, whereas most odontomas located in the posterior areas, especially the mandible, are recognized to be complex odontomas (4, 10). In fact, compound odontomas appear to be more frequent than complex odontomas (1, 4, 10); therefore, most of these cases are observed in the maxilla (5).

The precise etiology of odontomas remains unknown (1, 2, 12, 13). Nevertheless, it has been suggested that traumas and infections at the site of the lesion may be closely correlated with the incidence of these tumors (1, 4, 8, 10, 11). Furthermore, family history and genetic mutation have been proposed as the possible predisposing factors (7).

According to the literature, compound odontomas have no gender predilection and may be diagnosed at any age, especially during the first two decades of life (1, 2, 4, 10, 13). In general, these lesions are asymptomatic and slow-growing and seldom exceed the size of a tooth (1-3,5). Occasionally, they may grow to large sizes, causing cortical bone expansion (1-5, 7, 12, 14), which was observed in our patient. Since the clinical symptoms are not frequent, most lesions are discovered on routine radiographs. However, an affected patient may seek dental treatment when a permanent tooth or multiple teeth fail to erupt (1, 3, 7, 10, 13).

Odontomas are often observed in permanent dentition and are rarely associated with the primary teeth (1, 3), which is consistent with the patient examined in the present study. The radiographic aspects of odontoma are the characteristics for diagnosis (2). Odontomas could be sub-classified based on their radiographic appearance alone. Complex odontomas are indicated as an irregular mass of calcified material in which a thin radiolucent area with a smooth periphery surrounds, while the compound type shows calcified structures pertaining to teeth at the center of a well-defined

radiolucent lesion (1, 3, 4). The radiographic differential diagnoses of complex and compound odontomas include cementoblastoma, ameloblastic fibro-odontoma, periapical cemento-osseous dysplasia, and florid cemento-osseous dysplasia (1, 5).

Histologically, enamel matrix, dentin, pulp tissue and cementum are often detected in odontomas. Histopathological examination is compulsory for an accurate diagnosis of odontomas. The histological examination often displays the enamel matrix, dentin, pulp tissue, and cementum, which may correlate normally or abnormally (1, 3, 4).

Compound odontomas are composed of tooth-like structures pertaining to the pulp tissue in the central portion surrounded by dentin shells and partially covered by enamel components. Complex odontomas are conglomerated masses without the normal organization of dentin, enamel, enamel matrix, cementum, and areas of pulp tissue. Odontoma is surrounded by the connective tissue capsule that is similar to the follicle that covers a normal tooth (1-4). The conservative surgical removal of compound odontomas has remained the treatment of choice, which is associated with a slight risk of recurrence (1-3, 5, 8, 14). Therefore, the authors of this paper followed this protocol in the treatment of the patient.

Odontomas often grab the attention by the complications they cause such as over-retention, impaction, and the delayed eruption of the primary and permanent teeth. Although odontoma interfere with eruption, usually, no resorption of the adjacent tooth roots is detected (1, 3). Cases with the devitalization of the adjacent teeth, dentigerous cysts, and pindborg tumors with compound odontomas have also been reported in the literature (1, 3, 12, 13, 15). Therefore, the early detection and management of odontomas are highly recommended in order to prevent tooth eruption disturbances and further pathological complications (1-3,15).

Since odontomas are commonly associated with impacted or supernumerary teeth, systemic syndromes and hereditary diseases (e.g., cleidocranial dysostosis or Gardner's syndrome) should be considered (1, 11). However, neither systemic nor hereditary symptoms were found in our patient.

To the best of our knowledge, the number of the denticles found in compound odontomas have been reported to be 4-37 in the previous studies in this regard (1, 3, 9, 10). For instance, in a literature review by Amado et al (9) conducted on 38 cases suffering compound odontoma, the number of denticles was reported to be within the range of 4-28. In addition, Sharma et al (3). Have reported a case of compound odontoma with 37 denticles in the anterior maxilla of a 17-year-old male patient.

Interestingly, we presented a rare case of maxillary compound odontoma with a higher number of denticles (n=62) in a 19-year-old female patient. In fact, the extracted denticles in the present case exhibited concrescence, fusion, and dilacerations. 62 denticles were extracted presenting a unique case of odontoma, compared to the previously reported subjects with less number of denticles.

Conclusion

The odontomas diagnosis and management are essential to facilitating the elimination of the causes of eruption disturbances and avoiding the subsequent pathological complications. For the early detection of odontomas, panoramic radiography and proper clinical examinations are strongly recommended. The associated complications could be prevented via surgical excision followed by histopathological analysis. Moreover, the meticulous follow-up of the case clinically and radiographically plays a key role in assessing the eruption of the unerupted/impacted teeth, especially in the cases with extensive odontomas. It is notable that odontomas have favorable prognosis with rare cases of relapse. Therefore, odontomas timely diagnosis and surgical enucleation followed by curettage are suggested to avoid complications such as over-retention, impaction, and delayed eruption of the permanent teeth.

Conflicts of interest

None declared.

Acknowledgments

Hereby, we extend our gratitude to the Research Counselor of Mashhad University of Medical Sciences in Mashhad, Iran for assisting us in this research project.

References

1. Pippi R. Odontomas and supernumerary teeth: is there a common origin? *Int J Med Sci.* 2014;11(12): 1282-1297.
2. Bordini J, Contar CM, Sarot JR, Fernandes Â, Machado MÂ. Multiple compound odontomas in the jaw: case report and analysis of the literature. *J Oral Maxillofac Surg.* 2008;66(12): 2617-2620.
3. Sharma U, Sharma R, Gulati A, Yadav R, Gauba K. Compound composite odontoma with unusual number of denticles—A rare entity. *Saudi Dent J.* 2010;22(3): 145-159.

4. de Oliveira BH, Campos V, Marçal S. Compound odontoma-diagnosis and treatment: three case reports. *Pediatr dent.* 2001;23(2): 151-157.
5. Erdogan Ö, Keceli O, Öztunc H, Evlice B, Ayberk Altug H, Günhan Ö. Compound odontoma involving the four quadrants of the jaws: A case report and review of the literature. *Quintessence int.* 2014;45(4): 341-344.
6. Junquera L, de Vicente JC, Roig P, Olay S, Rodríguez-Recio O. Odontoma intraóseo erupcionado: Una infrecuente patología Intraosseus odontoma erupted into the oral cavity: An unusual pathology. *pathology.* 2005;10(1): 248-251.
7. Khanum N, Shivalingu MM, Lingaraju N, Basappa S. Compound-complex odontoma: A case report of a rare variant. *J Indian Academy Oral Medici Radiol.* 2014;26(4): 463.466.
8. Nelson BL, Thompson LD. Compound odontoma. *Head neck pathol.* 2010;4(4): 290-291.
9. Amado SC, Gargallo JA, Berini LA, Gay CE. Review of 61 cases of odontoma. Presentation of an erupted complex odontoma. *Medicina oral y Patologia y Medicina Bucal.* 2003;8(5): 366-373.
10. Ikram R, Rehman AA. Compound Odontomas In Saudi Child—A Case Report. *Int j health sci.* 2013;7(2): 242-246.
11. Kumazawa Y, Kawai T, Shirase T, Yagishita H, Yoshida K, Yosue T. Multiple calcifying lesions in the maxilla and mandible of a 4-year-old girl: report of a rare case and literature review. *Oral Radiol.* 2014;30(3):259-264.
12. Ayberk H. Compound odontoma involving the four quadrants of the jaws: A case report and review of the literature. *Quintessence Int.* 2014(4): 341-344.
13. Sun L, Sun Z, Ma X. Multiple complex odontoma of the maxilla and the mandible. *Oral surg, oral medic, oral pathol and oral radiol.* 2015; 120 (1): e11-e16.
14. Gujjar PK, Sahni P, Pereira T, Zingade J. Multiple compound odontomas in the Jaws: A rare case report. *J Clin Diagn Res.* 2015;9(12): ZD05-ZD06.
15. Han PP, Nagatsuka H, Siar CH, Tsujigiwa H, Gunduz M, Tamamura R, et al. A pigmented calcifying cystic odontogenic tumor associated with compound odontoma: a case report and review of literature. *Head face med.* 2007;3(1): 35-36.

Corresponding Author

Sahand Samieirad

Dental Research Center, Mashhad University of Medical Sciences,
Mashhad, Iran

Address: Oral and maxillofacial surgery department,

Mashhad dental school, Ferdowsi university campus, Azadi square, Mashhad, Iran

Mobile: 09128137859, Fax: +98513-8829500

Email: samieerads@mums.ac.ir