

Peripheral compound odontoma: A rare case report

SADJ OCTOBER 2024, Vol. 79 No.9 P493-496

V Peranovic,¹ BK Bunn,² P Gwengu³

ABSTRACT

Background

Odontomas are currently classified as benign mixed epithelial/mesenchymal odontogenic neoplasms which are located within the tooth-bearing regions of the jaws. Despite being considered neoplasms, the clinical characteristics of these lesions are much the same as hamartomatous lesions. Peripheral odontomas are rare entities that are located entirely within the adjacent soft tissue of the mucosa and/or gingiva. Apart from location, peripheral odontomas are morphologically, radiologically and pathologically indistinguishable from their intraosseous counterparts.

Case report

An 18-year-old female patient presented with an “irritation” on the mucosa of her hard palate posterior to the maxillary incisor teeth. She reported a small, tooth-like growth in the area which had been the same size for several years. Furthermore, she was asymptomatic with no other dental concerns and, in addition, she did not have any aesthetic or functional problems. Clinical examination revealed multiple tooth-like structures which had erupted as a mass posterior to tooth 11 and tooth 12. Radiographic examination confirmed the presence of a single, oval, well-circumscribed lesion partially encapsulated and superimposed over the mid aspects of the roots and partially over the crowns of the maxillary incisor teeth. The features noted are consistent with those of a peripheral compound odontoma.

Conclusion

Peripheral odontomas are exceedingly rare lesions which bear a resemblance and origin to their central intraosseous counterparts. This case demonstrates a peripheral compound odontoma which had not disrupted or affected the adjacent dentition.

Keywords

Odontoma, benign mixed epithelial/mesenchymal odontogenic neoplasm, compound odontoma, peripheral compound odontoma, extra-osseous compound odontoma, complex odontoma, hamartoma.

INTRODUCTION

During embryonic tooth development, the interaction which occurs between the ectodermal and mesenchymal (“ectomesenchyme”) tissue results in the well-timed, accurate and intricate development of both the deciduous and secondary teeth in a process of reciprocal induction. Interference, epigenetic or genetic alterations of normal tooth development may give rise to the development of a wide array of both benign and malignant odontogenic neoplasms. The majority of these are located within the bone in an intraosseous location with relatively few identified in soft tissues alone.¹⁻³ Although, at present, the World Health Organization (WHO) still classifies odontomas as odontogenic neoplasms, their frequency of occurrence and nature are more akin to those of a hamartoma.⁴ A hamartoma may be defined as a disorganised mass of tissue which is normal in structure and growth located within tissue at a site which it would ordinarily be located within.⁵

Peripheral variants of intraosseous odontogenic neoplasms are identical in their structure and morphology except for their location outside of bone. They are generally located within the gingiva and mucosa.⁴ At present, the WHO considers odontomas to represent “tumour-like” malformations composed of dental hard and soft tissues.^{1,6} Odontomas are classified into two types – complex and compound. Both forms usually present clinically as small, well circumscribed, mixed radiopaque/radiolucent lesions within the maxillary and mandibular bone. Complex odontomas comprise an amorphous, haphazard mixture of dental hard and soft tissue while a compound odontoma comprises numerous tooth-like structures which contain dental hard and soft tissue in close proportion to what is seen in normal tooth structure. It is rare for odontomas to erupt exclusively within the surrounding soft tissue although these have been documented.^{2,4,5,7-10} These are termed peripheral odontomas. Peripheral odontomas may be either compound or complex just as their intraosseous counterparts are.

The objective of this paper is to document the presence of a peripheral compound odontoma which had erupted into the oral mucosa without affecting the adjacent dentition.

CASE REPORT

An 18-year-old female patient presented with a main complaint of an irritation on her palatal mucosa adjacent to the posterior aspects of tooth 11 and tooth 12. She reported that there was no associated pain, sensitivity or discomfort in this region. She did not complain of poor aesthetics or of any functional difficulties. There was no medical, social or

Authors' information

1. V Peranovic, BDS, DipDent (Oral Pathology), MDS (Maxillofacial and Oral Radiology), Department of Maxillofacial and Oral Radiology, School of Oral Health Sciences, Sefako Makgatho Health Sciences University, Pretoria, South Africa ORCID: 0009-0007-5657-3341
2. BK Bunn, BDS, FCPATH (SA) Oral, MDent (Oral Pathology), Department of Operative Dentistry, School of Oral Health Sciences, Sefako Makgatho Health Sciences University, Pretoria, South Africa ORCID: 0000-0001-5699-4997
3. P Gwengu, DipOH, BDS, MpH, MDent (Comm Dent), Department of Operative Dentistry, School of Oral Health Sciences, Sefako Makgatho Health Sciences University, Pretoria, South Africa ORCID: 0000-0002-1429-2396

Corresponding author

Name: BK Bunn
Email: belindabunn@gmail.com
Tel: 082 708 5868

Author's contribution

1. V Peranovic – clinician who retrieved the case information, contribution to conceptualisation, writing and editing
2. BK Bunn – conceptualisation, writing, editing and submission
3. P Gwengu – writing and editing

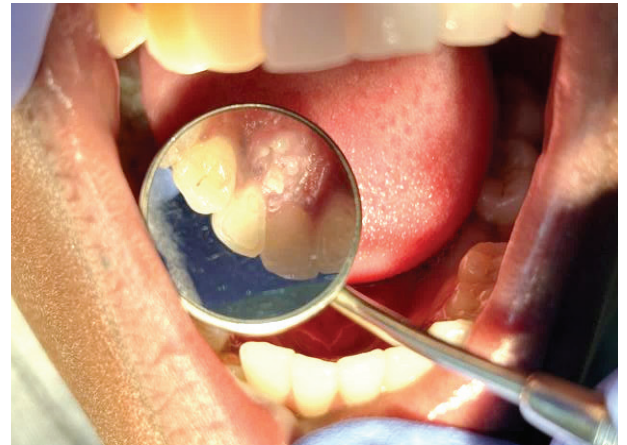


Figure 1. The clinical photograph above shows the intra-oral appearance of the patient's teeth in occlusion. From this view there is no evidence of pathology and no extra-oral abnormalities. (Left)
Figure 2. The clinical photograph above shows intra-oral presentation of the palatal aspect of teeth 11, 21, 22 and 23 with a mouth mirror. Reflected in the mirror is an erupted compound odontoma comprising numerous denticles. (Above)

family history of significance. There was no preceding history of trauma or evidence of infection. Intra-oral examination showed all permanent teeth to be present and well aligned in both dental arches. There were no missing teeth, no retained deciduous teeth, no supernumerary teeth and no evidence of diastema formation. The patient had no dental complaints. A class one occlusion was noted in which a mild crossbite was identified in the region of the right canine and premolar teeth (Figure 1).

Examination of the palatal mucosa showed a slight swelling located on the posterior aspect of teeth 11 and 12. The lesion appeared to comprise multiple protruding tooth-like structures resembling small crowns of teeth. On palpation, the swelling and calcified tooth-like structures were hard, non-tender and immobile. The surrounding palatal mucosa was normal in appearance, being non-erythematous with no evidence of inflammation or of ulceration (Figure 2).

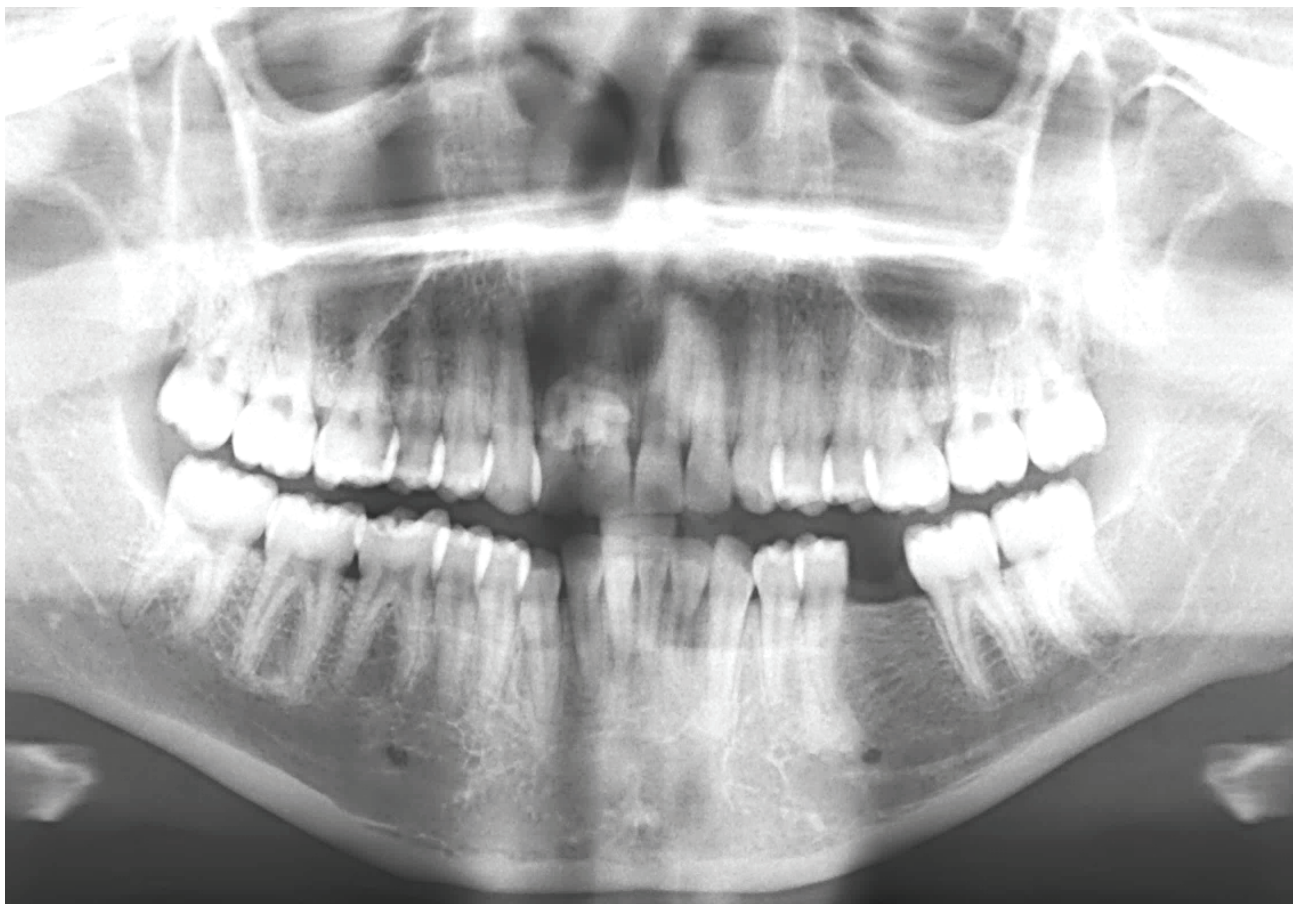


Figure 3. Panoramic radiograph showing the superimposition of an erupted compound odontoma on the palatal aspects of the roots and crowns of tooth 11 and tooth 12.

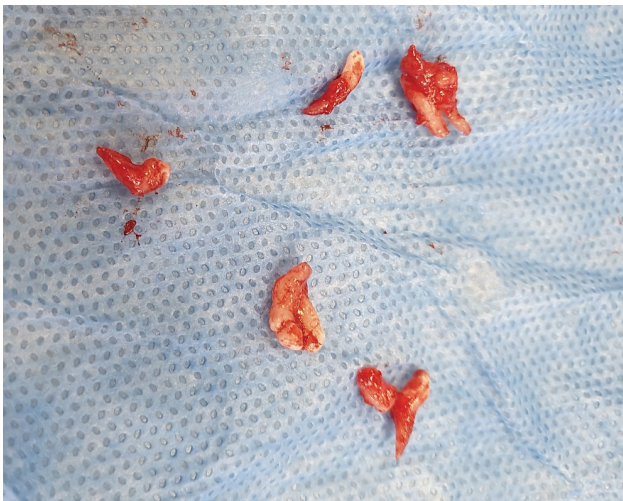


Figure 4. The clinical photograph depicts the multiple, somewhat misshapen, tooth-like structures which were obtained at surgical removal of the lesion. The denticles were attached to each other by minimal connective tissue and were separated easily.

Panoramic radiograph showed a single oval, well-defined lesion measuring 15mm in diameter, located in the anterior maxilla where it is superimposed over the roots of tooth 11 and 12 slightly obscuring them. The lesion had a predominantly radiopaque appearance but was distinctly composed of individual small tooth-like structures (denticles) surrounded on one aspect by a fibrous capsule. The surrounding anatomical structures were in no way affected by the lesion. There was no clinical evidence of tooth displacement, nor of root resorption. The adjacent teeth are all vital (Figure 3).

Under local anaesthesia, the odontoma was enucleated, resulting in excision of multiple tiny toothlets (denticles) with minimal attached soft tissue. These tooth-like structures were submitted for histology. The pathology report confirmed the presence of a compound odontoma (Figure 4).

DISCUSSION

Odontomas are classified as benign mixed epithelial-mesenchymal neoplasms by the World Health Organization.¹¹ Odontomas are typically located within the bone where they are identified as small round radiopaque/radiolucent anomalies either representing compound or complex varieties. Compound odontomas are encapsulated lesions comprising numerous miniature teeth/denticles. These tiny denticles are clustered together and are fully representative of pulpal tissue, dentine, cementum and enamel in the correct proportions but on a much smaller scale. Complex odontomas, conversely, are also well demarcated radiopaque lesions which comprise an amorphous mass of dental hard material including pulpal tissue which are arranged in a haphazard manner.⁵ The consequences of odontomas include possible impaction of adjacent teeth or malpositioning of surrounding teeth. Furthermore, there have been reports documenting secondary infection within odontomas. It is extremely rare for an odontoma to spontaneously erupt without causing any complications to the surrounding dentition. An odontoma which erupts completely into the soft tissue with no remaining association to the underlying bone is termed an “erupted odontoma”.⁴ An odontoma which has partially erupted into the adjacent soft tissue which may or may not include eruption into the oral cavity but demonstrates some form of attachment to the underlying bone even if it is in the form

of a ligament or part of a fibrous band or connective tissue capsule, the lesion is referred to as a peripheral odontoma. Peripheral odontomas are thus the soft tissue counterparts of their intra-osseous forms. Compound odontomas tend to occur within the anterior regions of the maxilla while complex odontomas are more frequently encountered in the mid to posterior regions of the mandible.^{5,10}

Intra-osseous odontomas are the most prevalent odontogenic neoplasm with a documented frequency of 45.8%. True peripheral odontomas are a rare phenomenon. Most peripheral odontomas have been documented to occur in the maxilla and are generally identified as asymptomatic, slow-growing masses in children.⁴ The denticles/tooth-like structures identified in compound odontomas may show variation in morphology but will all be composed of pulpal material surrounded by dentine, cementum and immature enamel.

It has been postulated that the WNT/ -catenin pathway is activated in embryonic SOX2 positive dental stem cells which then play a role in the development and formation of odontomas.

The exact aetiopathogenesis of peripheral odontomas is not well known or understood. It has been proposed that small remnants or cell rests of the dental lamina (such as the cell rests of Serres) may retain the ability to undergo reciprocal induction with the surrounding mesenchyme resulting in the formation of organised (compound) or disorganised (complex) masses termed odontomas. A point of debate related to these lesions has been encountered in the literature. Odontomas could be classified as intra-osseous, peripheral or erupted. By definition, an intra-osseous odontoma is, as its name implies, located entirely within bone. The peripheral odontoma, likewise, is located entirely within soft tissue. The term “erupted odontoma” appears to be a misnomer as, in most cases, there remains superficial attachment to the underlying bone and, as such, should be regarded as intra-osseous odontomas, even if a small part of the odontoma partially erupts into the oral cavity.^{4,12} Furthermore, the debate around the potential eruptive force and complete morphogenesis of a peripheral odontoma has ensued. However, since a true peripheral odontoma lacks a periodontal ligament or attachment to bone, there is virtually no potential for it to fully erupt with little evidence to suggest continued tooth development or maturation.^{12,13} There are several plausible theories as to the origin of the peripheral odontoma.

As previously mentioned, it is suspected that random remnants of the dental lamina may represent the origin of the peripheral odontoma; however, there is no agreement as to what stimulates the activity of these rests to initiate proliferation.¹⁴⁻¹⁷

Alternative theories suggest origin from the basal surfaces of the oral epithelium or because of activated ectopic neural crest cells.⁹

The clinical and morphological appearance of a peripheral compound odontoma is unique enough to identify two possible differential diagnoses. In this scenario, one must definitively exclude the possibility of a supernumerary tooth. The recommended treatment of choice for peripheral odontomas is simple surgical excision for which no recurrences have been reported to date.

CONCLUSION

Peripheral compound/complex odontomas are rare odontogenic lesions which may perplex the uninitiated OHCW. The slow growth, lack of symptoms in association with the lesion and the lack of origin from the underlying bone, together with curative surgical removal as well as increased awareness of such lesions, may increase the detection and reporting rate of such cases to more accurately represent the prevalence of these lesions within the communities we serve.

REFERENCES

1. Wright JM, Vered M. Update from the 4th Edition of the World Health Organization Classification of Head and Neck Tumours: Odontogenic and Maxillofacial Bone Tumours. *Head Neck Pathol.* Mar 2017;11(1):68-77
2. Manor Y, Mardinger O, Katz J, Taicher S, Hirschberg A. Peripheral odontogenic tumours - differential diagnosis in gingival lesions. *Int J Oral Maxillofac Surg.* Apr 2004;33(3):268-273
3. Soluk Tekkesin M, Pehlivan S, Olgac V, Aksakalli N, Alatlı C. Clinical and histopathological investigation of odontomas: review of the literature and presentation of 160 cases. *J Oral Maxillofac Surg.* Jun 2012;70(6):1358-1361
4. Soluk-Tekkesin M, Balkan B, Akatay DK, Atalay B. A Rare Case of Peripheral Compound Odontoma and Review of the Literature. *Head Neck Pathol.* Sep 2022;16(3):913-917
5. Agrawal B, Gharote H, Nair P, Shrivastav S. Infected complex odontoma: an unusual presentation. *BMJ Case Rep.* Aug 24 2012;2012
6. Soluk-Tekkesin M, Wright JM. The World Health Organization Classification of Odontogenic Lesions: A Summary of the Changes of the 2017 (4th) Edition. *Türk Patoloji Derg.* 2018;34(1)
7. Ledesma-Montes C, Perez-Bache A, Garcés-Ortiz M. Gingival compound odontoma. *Int J Oral Maxillofac Surg.* Aug 1996;25(4):296-297
8. da Silva Rocha OKM, da Silva Barros CC, da Silva LAB, de Souza Junior EF, de Morais HHA, da Costa Miguel MC. Peripheral compound odontoma: A rare case report and literature review. *J Cutan Pathol.* Aug 2020;47(8):720-724
9. Ide F, Obara K, Mishima K, et al. Peripheral odontogenic tumor: a clinicopathologic study of 30 cases. General features and hamartomatous lesions. *J Oral Pathol Med.* Oct 2005;34(9):552-557
10. Machado C de V, Knop LA, da Rocha MC, Telles PD. Impacted permanent incisors associated with compound odontoma. *BMJ Case Rep.* Jan 12 2015;2015
11. 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.* Mar 1997;33(2):86-99
12. Ide F, Ito Y, Nishimura M, Kusama K, Kikuchi K. Peripheral developing odontoma: Is it so rare? *J Cutan Pathol.* Jan 2021;48(1):200-201
13. Giunta JL, Kaplan MA. Peripheral, soft tissue odontomas. Two case reports. *Oral Surg Oral Med Oral Pathol.* Mar 1990;69(3):406-411
14. Silva AR, Carlos-Bregni R, Vargas PA, de Almeida OP, Lopes MA. Peripheral developing odontoma in newborn. Report of two cases and literature review. *Med Oral Patol Oral Cir Bucal.* Nov 1 2009;14(11):e612-615
15. de Oliveira BH, Campos V, Marçal S. Compound odontoma - diagnosis and treatment: three case reports. *Pediatr Dent.* Mar-Apr 2001;23(2):151-157
16. Custodio M, Araujo JP, Gallo CB, Trieveller M. Gingival complex odontoma: a rare case report with a review of the literature. *Autops Case Rep.* Jan-Mar 2018;8(1):e2018009
17. Ide F, Mishima K, Saito I, Kusama K. Rare peripheral odontogenic tumors: report of 5 cases and comprehensive review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* Oct 2008;106(4):e22-28

Online CPD in 6 Easy Steps



The Continuing Professional Development (CPD) section provides for twenty general questions and five ethics questions. The section provides members with a valuable source of CPD points whilst also achieving the objective of CPD, to assure continuing education. The importance of continuing professional development should not be underestimated, it is a career-long obligation for practicing professionals.

