CASE REPORT

An erupted compound odontoma

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SUMMARY

Odontomas are familiar entities but their eruption into the oral cavity is an extraordinary occurrence, which may be associated with pain, infection, malocclusion, etc. Not many cases of erupted odontomas have been reported in the literature. This paper puts forth a case of erupting odontoma in an attempt to add to the list of reported cases of this unique pathology.

BACKGROUND

Odontomas are now more widely accepted to be hamartomas rather than neoplasms and are composed of dental epithelial as well as mesenchymal elements, that is, enamel, dentin, cementum and pulp tissue.¹ The term 'odontoma' was coined by Paul Broca in 1867. He defined odontoma as a tumour of complete dental tissue.² WHO has classified odontomas into complex and compound odontomas. The malformation where the dental tissues are seen in a less orderly pattern are considered as complex odontomas whereas those where dental tissues are seen in a more orderly arrangement are considered as compound odontomas.³

Odontomas have also been classified as central, peripheral and erupted based on their location either inside the bone or in the soft tissues covering the tooth-bearing areas, with a tendency to exfoliate⁴ or the ones which, in exceptional instances, break through the mucosa and appear in the oral cavity,⁵ respectively. This article puts forth an unusual case of erupted compound odontoma in a 14-year-old female patient.

CASE PRESENTATION

A 14-year-old female patient reported to the dental clinic with an undesirable appearance of the upper front teeth since 4–5 months. Extraoral examination was unremarkable. Clinical intraoral examination revealed normal complement of teeth in maxillary and mandibular arches. A yellowish-white lobulated mass with the colour and texture similar to that of enamel was noted in the first quadrant. The unaesthetic appearance of this tooth-coloured hard tissue mass made the patient apprehensive and forced the patient to visit the dental clinic. Apart from the presence of this pathological structure the patient was free from any symptoms. The mass was present on the labial aspect in between the maxillary right central and lateral incisors (11, 12), towards the cervical portion of their roots (figure 1). The surrounding gingival tissue was mildly inflamed and a band of gingival tissue was running between two lobules of the lesion. The lesion was about 1.2×1 cm in dimension and hard in consistency and painless on palpation.

Figure 1 Photograph showing enamel coloured mass in between 11, 12.

INVESTIGATIONS

The patient was advised a set of radiographs (including intraoral periapical and occlusal radiographs) to assess the precise location and extent of the lesion, as well as its relations to the surrounding anatomical structures (figures 2 and 3). Radiographic evaluation revealed multiple small teeth-like radio-opaque structures or denticles in maxillary anterior region in relation to the cervical portion of roots of maxillary right central and lateral incisors. Based on the clinical and radiographic assessment, a provisional diagnosis of erupted compound odontomas was given.

DIFFERENTIAL DIAGNOSIS

A compound odontoma hardly presents any difficulties in differential diagnosis due to its typical radiographic presentation, that is, tooth-like radio-opacities within a well-defined lesion. It has been observed that some cases which are radiographically thought to be the intermediate stage of a compound odontoma; on histopathological examination may exhibit few areas of ameloblastic proliferation and thus have to be categorised as ameloblastic odontomas.^{6,7}

Figure 2 Occlusal radiograph showing radio-opaque denticles in maxillary anterior region.

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Figure 3 Intraoral periapical radiograph showing denticles in between 11, 12.

Clinical and radiographic features, along with gross examination, of a compound odontoma are quite distinct and hence it is not difficult to reach a final diagnosis. This is unlike the complex odontomas which have a more elaborate differential diagnosis.

TREATMENT

The lesion was removed under local anaesthesia through an intraoral approach, taking care to remove all denticles ([figure 4](#)). The denticles were then sent for histopathological examination which confirmed the diagnosis of compound odontoma ([figure 5](#)).

OUTCOME AND FOLLOW-UP

The postoperative healing was uneventful ([figure 6A](#)) and post-operative radiograph revealed complete removal of all denticles and normal healing ([figure 6B](#)).

Figure 4 Gross specimen showing denticles.

Figure 5 Photomicrograph showing normal relationship of tissues in teeth.

DISCUSSION

The term 'odontoma' literally means tumour of odontogenic origin, but it is used for a specific entity having epithelial and mesenchymal origin, with typical clinical and histological features.³ Though earlier odontomas were considered as hard tissue tumours of odontogenic origin,⁸ it is now more widely accepted that these represent hamartomatous malformations rather than any neoplasms.³

Despite the fact that coining of the term 'odontoma' is credited to Paul Broca² the description of 'erupted odontoma' is credited to Rumel *et al.*⁹ Erupted odontomas are rare entities and add to the types of odontomas which may be intraosseous or extraosseous, thus extending the list of types of odontomas. The exact cause of this pathological entity is not clear but it has been suggested that genetic basis, infections, inflammatory factors, trauma or even hyperactivity of odontoblasts may play a role in the development of odontomas.^{9–10} Apart from these factors it is the persistence of a part of dental lamina which has been thought to play a vital role in the development of odontomas.¹¹ Ide *et al.*¹² have supported this theory by hypothesising that the gingival rests of Serre retain their capacity of epithelial-mesenchymal interactions which can lead to formation of odontomas. Further Pindborg¹³ had suggested that if left untreated odontomas can erupt into the oral cavity, this occurrence which has been reported by various authors and also in the current paper reinforces the theory of continued ability of epithelial-mesenchymal interaction in the cell rests.

Figure 6 (A) Postoperative photograph showing uneventful healing. (B) Postoperative intraoral peri apical.

The total incidence of this pathology has been reported varying from 22% to 67% of all odontogenic neoplasms, and compound odontomas have been reported to be more common than the complex ones.^{2 14} Studies stating its incidence as a hamartoma are not found. The relative incidence of erupting odontomas is very low and the current case represents this exceptional situation where the odontoma has erupted into the oral cavity. As per the analysis performed by Cuesta *et al*¹⁴ odontomas have a predilection for maxillary incisor and canine region. In the current case also the affected site was the maxillary anterior region.

According to a review by Cuesta *et al* among the erupted category of odontomas most are of the compound type as was also seen in the case under discussion. According to these authors 52.2% of the reported cases have been seen in women as was also the current case. But few studies have reported the incidence to be twice as high in women as compared to men while contradictory reports with a male predilection are also there.¹⁴

The age of occurrence of odontoma, as per study by Phillipsen *et al*,¹⁵ can vary from 2 to 74 years, with the complex and compound type showing a peak in the second decade of life. Even the case reported in this paper was seen in the second decade thus the age being consistent with the reported peak age of occurrence.

Most of the reported cases of odontomas are asymptomatic and the patients generally report with missing tooth. Few cases may present with swelling and occasionally infection and lymphadenopathy may be noted.¹⁶ Although often asymptomatic erupting odontomas may be associated with pain and swelling, recurrent infections or halitosis.^{10 17 18} Further an erupting odontoma can even lead to malocclusion.¹⁸ In the current case as well the patient was asymptomatic.

On radiographic evaluation odontomas usually reveal to be radio-opaque masses with radiolucent halo but the radiological appearance can vary with the developmental stage of the odontoma. Three stages have been recognised based on amount of calcification. Stage 1 shows radiolucency due to lack of calcification (soft odontoma); partial calcification is seen in the intermediate stage, while the third stage shows complete calcification with radio-opacity and thin radiolucent periphery. The radio-opacities appear as miniature teeth in compound odontomas and as irregular radio-opaque masses in complex type of odontomas.¹⁹ In the present case miniature teeth-like structures were completely radio-opaque thus confirming the third radiographic stage of a compound odontoma.

Histologically odontomas consist of epithelial and mesenchymal tissues and their cells with apparently normal morphology. These tissues show a disordered arrangement in complex odontomas, whereas compound odontomas with miniature teeth-like structures show a well organised tooth-like relationship of tissues. This indicates a higher degree of differentiation of cells in compound odontomas as compared with the complex odontomas.^{19 20} In the case under consideration, well organised dental tissues were evident during histopathological examination (figure 5) thus representing a compound odontoma.

The treatment of choice for odontomas is generally conservative enucleation and the possibility of recurrence is very low.²¹ Further it has also been noticed that enucleation is easy in cases of odontoma and the adjacent teeth even if displaced are seldom harmed by the procedure because usually there is a septum of bone in between the teeth and the odontoma. Orthodontic treatment may be required to correct the malocclusion if it occurs.¹ According to Blinder *et al*²² cases of larger odontomas have certain special considerations. In relation to compound odontomas, like the present case, removal of all small tooth-like structures should be ensured and this should be postoperatively checked using radiographs. Although only few denticles were seen in the current case on surgical intervention, lesions containing upto 100 denticles have also been reported.¹⁹

Learning points

- ▶ Unlike intraosseous odontomas, erupted odontomas are rare lesions.
- ▶ Despite their benign nature, eruption of an odontoma into the oral cavity can give rise to pain, inflammation, infection, etc.
- ▶ Encompass lesions with intriguing aetiology, which were once considered to be benign neoplasms but now are categorised as hamartomas.

Competing interests None.

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REFERENCES

- Batra P, Duggal R, Kharbanda OP, *et al.* Orthodontic treatment of impacted anterior teeth due to odontomas: a report of two cases. *J Clin Pediatr Dent* 2004;28:289–94.
- Rajendran R, Sivapathasundharam B. *Shafer's textbook of oral pathology*. 7th edn New Delhi: Elsevier, a division of Reed Elsevier India Pvt. Ltd, 2012:259–316.
- Junquera L, de Vicente JC, Roig P, *et al.* Intraosseous odontoma erupted into the oral cavity: an unusual pathology. *Med Oral Patol Oral Cir Bucal* 2005;10:248–51.
- Serra-Serra G, Berini-Aytés L, Gay-Escoda C. Erupted odontomas: a report of three cases and review of the literature. *Med Oral Patol Oral Cir Bucal* 2009;14: E299–303.
- Tozoglu S, Yildirim U, Buyukkurt MC. An erupted complex odontoma. *N Y State Dent J* 2010;76:52–3.
- Wood NK, Goaz PW. *Differential diagnosis of oral lesions*. 3rd edn. St Louis, MO: CV Mosby Company, 1985:515–35.
- White SC, Pharoah MJ. *Oral radiology: principles and interpretation*. 5th edn. St Louis, MO, USA: Mosby, an imprint of Elsevier, 2004:410–57.
- Rumel A, de Freitas A, Birman EG, *et al.* Erupted complex odontoma. Report of a case. *Dentomaxillofac Radiol* 1980;9:5–9.
- Levy BA. Effects of experimental trauma on developing first molar teeth in rats. *J Dent Res* 1968;47:323–7.
- Gomel M, Seçkin T. An erupted odontoma: case report. *J Oral Maxillofac Surg* 1989;47:999–1000.
- Vengal M, Arora H, Ghosh S, *et al.* Large erupting complex odontoma: a case report *J Can Dent Assoc* 2007;73:169–73.
- 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* 2005;34:552–7.
- Pindborg JJ. *Pathology of dental hard tissues*. 1st edn. Philadelphia, London & Toronto: WB Saunders Company, 1970:393–401.
- Amado Cuesta S, Gargallo Albiol J, Berini Aytés L, *et al.* Review of 61 cases of odontoma. Presentation of an erupted complex odontoma. *Med Oral* 2003;8:366–73.
- 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.
- Hidalgo-Sánchez O, Leco-Berrolcal MI, Martínez-González JM. Meta-analysis of the epidemiology and clinical manifestations of odontomas. *Med Oral Patol Oral Cir Bucal* 2008;13:E730–4.
- Nik-Hussein NN, Majid ZA. Erupted compound odontoma. *Ann Dent* 1993;52:9–11.
- Ferrer Ramírez MJ, Silvestre Donat FJ, Estelles Ferriol E, *et al.* Recurrent infection of a complex odontoma following eruption in the mouth. *Med Oral* 2001;6:269–75.
- Reichart PA, Philipsen HP. *Odontogenic tumours and allied lesions*. 1st edn. Quintessence Publishing Co., 2004:271–92.
- Piattelli A, Trisi P. Morphodifferentiation and histodifferentiation of the dental hard tissues in compound odontoma: a study of undemineralized material. *J Oral Pathol Med* 1992;21:340–2.
- López-Areal L, Silvestre Donat F, Gil Lozano J. Compound odontoma erupting in the mouth: 4-year follow-up of a clinical case. *J Oral Pathol Med* 1992;21:285–8.
- Blinder D, Peleg M, Taicher S. Surgical considerations in cases of large mandibular odontomas located in the mandibular angle. *Int J Oral Maxillofac Surg* 1993;22:163–5.

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