CASE REPORT

A complex odontoma of the anterior maxilla associated with an erupting canine

Ajay Pillai,¹ Swapnil Moghe,¹ Mithilesh K Gupta,¹ Abhishek Pathak²

¹Department of Oral & Maxillofacial Surgery, People's Dental Academy, Bhopal, Madhya Pradesh, India

²People's Dental Academy, Bhopal, Madhya Pradesh, India

Correspondence to

Dr Swapnil Moghe,
drmogheswapnil@gmail.com

SUMMARY

Many developmental anomalies of the jaw are accidentally diagnosed during radiographic investigations for other symptoms. Complex odontomas are odontogenic tumours, relatively rare in the anterior maxilla and generally asymptomatic. Frequently the odontoma may interfere with the eruption of teeth. This paper describes a case of complex odontoma in an 8-year-old girl, which resulted in the palatal displacement of the lateral incisor and an erupting permanent canine. A calcified mass was seen on the radiograph and was provisionally diagnosed as an odontoma which was surgically enucleated. Routine follow-up was carried out for more than 1 year and no recurrence was seen.

BACKGROUND

Odontomas are benign tumours of odontogenic origin consisting of enamel, dentine, cementum and pulpal tissue. These tumours are characterised by slow growth and constitute 22% of all odontogenic tumours. There are two types of odontomas: compound and complex.¹ Compound odontomas appear as numerous miniature or rudimentary teeth. Complex odontomas appear as amorphous conglomerates of hard tissue.¹ The compound type is approximately twice as common as the complex type. Some are associated with spontaneous eruption of teeth.²⁻³ Odontomas are known as mixed odontogenic tumours because they consist of tissues of epithelial and mesenchymal origin. These tissues differentiate fully, resulting in the deposition of enamel by ameloblasts and dentine by odontoblasts. This tumour presents more commonly in children and adolescents, and more frequently in the maxilla than the mandible. The compound type tends to occur in the anterior part of the jaws, and the complex type tends to present in the posterior part. Histopathologically, normal-appearing enamel, dentine, cementum and pulp may be seen.¹ Odontomas are considered to be a type of odontogenic tumour that may cause the impaction of both primary and permanent teeth.⁴⁻⁵ Most are found when a patient is evaluated radiographically for tooth eruption disturbances.⁶ Although most maxillary odontomas are small, and easily diagnosed and removed by the practitioner, removal of a larger odontoma may be challenging. Adherence to certain surgical principles is essential to prevent problems and allow for the atraumatic and successful removal of large maxillary odontomas. This article presents a case involving a large, complex odontoma present over the anterior maxillary region, which had displaced a primary lateral

incisor and a primary canine palatally, along with management and successful treatment outcome.

CASE PRESENTATION

An 8-year-old girl presented with a painless enlargement of the left anterior maxilla that had gradually grown bigger over 1 year (figure 1). She also reported the changing position of the upper left primary lateral incisor and primary canine. The young patient was well nourished and had no other developmental anomaly detected on clinical examination.

On local examination, a hard bony swelling measuring approximately 1 cm × 1.5 cm, painless and non-tender was located on the left side of the maxillary anterior region with a displaced left primary lateral incisor and primary canine. The patient was in good general health, and the clinical and laboratory examinations were within normal limits. Subsequently, surgical enucleation of the lesion was planned in accordance with its size.

INVESTIGATIONS

Orthopantomography showed a multiple dense radiopaque mass with a surrounding radiolucent rim, occupying the anterior portion of the left maxilla associated with an erupting canine (figure 2).

Excisional biopsy confirmed the diagnosis of a complex odontoma. The histopathological picture revealed irregularly arranged eosinophilic areas with dentinal tubules resembling dentine-like tissue. Basophilic areas were also present in the form of irregular small masses resembling cementum.

CrossMark

To cite: Pillai A, Moghe S, Gupta MK, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2013-200684

Figure 1 Preoperative clinical picture.

Figure 2 Preoperative panoramic radiograph.

DIFFERENTIAL DIAGNOSIS

The clinical and radiographical features suggested the diagnosis of odontoma, although other differential diagnoses include calcifying odontogenic cyst, calcifying odontogenic tumour, fibro-osseous lesion, ameloblastic fibro-odontoma and osteoblastoma. Ossifying fibroma may closely mimic an odontoma but can be differentiated from it by the fact that it is well circumscribed and usually separates easily from its bony bed.

TREATMENT

The operation was performed with the patient under sedation and local anaesthesia. A full thickness mucoperiosteal flap was raised buccally from the left lateral incisor extending up to the mesial aspect of the first left molar (figure 3). A thin shell like bone covering the odontoma was removed exposing the lesion (figure 4). Using a series of chisels and curettes, the lesion was carefully and completely removed from the maxilla, preserving the erupting canine and premolars (figure 5). Haemostasis was achieved, closure of mucoperiosteal flap was carried out and the specimen was sent for histopathological examination. The patient's postoperative course was uneventful.

OUTCOME AND FOLLOW-UP

The histopathological examination confirmed the diagnosis of a complex odontoma (figure 6). No clinical or radiographic evidence was found of recurrence of the mass or any other related complications after 6 months of follow-up (figure 7). The patient continues to be on regular follow-up.

DISCUSSION

Odontomas are the most common odontogenic tumours and are classified as benign, mixed and calcified odontogenic

Figure 4 Thin shell-like bone removed after exposing the lesion.

tumours. Most occur in the first and second decades of life, and the mean age at the time of diagnosis is 14 years.^{1 2} Odontoma is frequently associated with an impacted tooth and occasionally with a dentigerous cyst,⁷ and it has a marked predilection for the maxilla (67%) and for the anterior region of the jaw (61%). Although complex odontomas are found in the posterior jaw, Chang⁷ found them rarely in the anterior maxilla. Paul Broca was the first to coin the term odontoma in 1867.⁸ Odontomas act similarly to impacted teeth, and thus often cause disturbances in the eruption of teeth (eg, impaction or delayed eruption of the dentition, retention of primary teeth and abnormalities in the position of the teeth, tipping or displacement of adjacent teeth). Although the majority of unerupted teeth are in the permanent dentition, the problem can be identified in the early-mixed dentition.^{1 2} Complex odontomas are usually located in the first and second molar areas of the mandible. Although the compound type is equally distributed between the genders, 60% of complex odontomas occur in women. In our case, the left maxillary permanent canine was noticed to be embedded due to the odontoma.

WHO (2005) classified odontoma as a benign odontogenic tumour composed of odontogenic epithelium and odontogenic ectomesenchyme with dental hard tissue formation.

The aetiology of odontoma is not completely understood, but has been attributed to various pathological conditions such as trauma, infection and hereditary anomalies (Gardner's syndrome, Herman syndrome and basal cell naevus syndrome).⁸⁻¹⁰

Figure 3 Trans-surgical view: exposure of the lesion.

Figure 5 Enucleated complex odontome.

Figure 6 Histological slide of the tumour confirming the diagnosis of complex odontoma showing irregularly arranged eosinophilic areas with dental tubules resembling dentine-like tissue.

Clinically, three types of odontoma are recognised: intraosseous, extraosseous and erupted.

Thoma and Goldman classified odontomas as follows:

1. Geminated composite odontomes—two or more, more or less well-developed teeth fused together.
2. Compound composite odontomes—made up of more or less rudimentary teeth.
3. Complex composite odontomes—calcified structure, which bears no great resemblance to the normal anatomical arrangement of dental tissues.
4. Dilated odontomes—the crown or root part of the tooth shows marked enlargement.
5. Cystic odontomes—an odontome that is normally encapsulated by fibrous connective tissue in a cyst or in a wall of cyst.⁸

Compound odontomas seldom cause bony expansion, while complex odontomas often cause slight to marked bony expansion,⁴ although in our case we noticed expansion of the buccal cortical plate. Surgical exposure and removal of the odontoma, and elimination of the mechanical obstruction is frequently the treatment of choice, and spontaneous eruption can then be expected.⁷ However, there has been no general agreement on the best management approach for impacted teeth associated with the lesion. The treatment option comprises surgical extraction and postsurgical clinical and radiological controls to evaluate the course of the impacted tooth. In our case, we have

Figure 7 Postoperative panoramic radiograph.

planned a second surgical and orthodontic intervention to provide a better outcome for the impacted canine.¹¹

When odontomas are associated with unerupted teeth, orthodontic traction of the impacted tooth soon after removal of the lesion may be needed, but in our case since the patient is only 8 years old, the orthodontist advised follow-up with the patient for another 2 years before considering orthodontic traction.

Radiographically, compound odontomas are usually multiple, small, calcified and radiopaque masses with an anatomical similarity to normal teeth, whereas complex odontomas are calcified radiopaque masses which bear no anatomical resemblance to the teeth and are frequently associated with the posterior mandibular region, though both odontomas are surrounded by a narrow radiolucent zone.¹² In our reported case, we noticed multiple dense radiopaque masses which were suggestive of a complex odontoma.

Learning points

- ▶ An odontoma should always be removed because it contains different materials that can predispose to cystic change, interfere with tooth eruption and cause considerable bone expansion.
- ▶ The literature and clinical experience suggest that radiographic examination should be carried out for any paediatric patient who presents with clinical evidence of delayed permanent tooth eruption with or without a history of maxillofacial trauma; early diagnosis allows adoption of a less complex and less expensive treatment and ensures a better prognosis.
- ▶ An odontoma is usually separated from its adjacent teeth by the septum, hence early diagnosis and surgical removal of the odontoma will result in a better prognosis for the teeth.
- ▶ The surgical specimen should be carefully examined microscopically to rule out ameloblastic odontoma or myxofibrous hyperplasia.

Acknowledgements The author would like to thank the staff and management of People's Hospital for the support and care they provided to our patients.

Contributors All the authors assisted in the surgical procedure and the preparation of the manuscript.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Regezi JA, Sciubba JJ. *Oral pathology: clinical pathologic correlations*. 3rd edn. Philadelphia, PA: WB Saunders, 1999:350–2.
- 2 Neville BW, Damm DD, Allen CM, et al. *Oral and maxillofacial pathology*. 2nd edn. Philadelphia, PA: WB Saunders, 2002:531–4.
- 3 Amado Cuesta S, Gargallo Albiol J, Berini Aytes L, et al. Review of 61 cases of odontoma. Presentation of an erupted complex odontoma. *Med Oral* 2003;8:366–73.
- 4 Cildir SK, Sencift K, Olgac V, et al. Delayed eruption of a mandibular primary cuspid associated with compound odontoma. *J Contemp Dent Pract* 2005;6:152–9.
- 5 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.
- 6 Tomizawa M, Otsuka Y, Noda T. Clinical observations of odontomas in Japanese children: 39 cases including one recurrent case. *Int J Paediatr Dent* 2005;15:37–43.
- 7 Chang JY, Wang JT, Wang YP, et al. Odontoma: a clinicopathologic study of 81 cases. *J Formos Med Assoc* 2003;102:876–82.
- 8 John JB, John RR, Punithavathy . Indumathi elango "Compound Odontoma Associated with Maxillary Primary Tooth—A Case Report". *J IADS* 2010;1:49–51.

- 9 Sudarshan R, Annigeri RG, Vijayabala GS. Bird-headed dwarfism (Seckel syndrome) – a rare case report. *TAF Prev Med Bull* 2013;12:209–12.
- 10 Krichen G, Hentati H, Hadri R, *et al.* Odontoma associated with supernumerary and impacted teeth. *Int Dent J Students Res* 2013;1;47–52.
- 11 An S-Y, An C-H, Choi K-S. Odontoma: a retrospective study of 73 cases. *Imaging Sci Dent* 2012;42:77–81.
- 12 de Oliveira BH, Campos V, Marçal S. Compound odontoma—diagnosis and treatment: three case reports. *Paediatr Dent* 2001;23:2.

Copyright 2013 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow