Unusual presentation of more common disease/injury

Infected complex odontoma: an unusual presentation

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Summary

Odontomas represent a hamartomatous malformation. They are usually asymptomatic and are diagnosed on routine radiological examination. The eruption and infection of odontoma are uncommon. Till now, only 20 cases of erupted odontoma are reported in the literature. We report an unusual case of infected complex odontoma with eruption of odontoma in the oral cavity and perforation of the cheeks with a tooth impacted upon which makes the presentation unusual that has not been reported earlier. The dentist should be familiar with common and unusual appearance of odontoma for proper diagnosis and management.

BACKGROUND

The term odontoma was first coined by Broca in 1866, who defined it as a tumour formed by overgrowth of complete dental tissue.¹ Histologically, they are composed of different dental tissues, including enamel, dentine, and cementum and, in some cases, pulp tissue.²⁻³ According to the latest WHO classification (2005) of odontogenic tumours there are two types of odontomas—compound odontomas and complex odontomas. The compound odontomas are usually unilocular and contain multiple teeth-like bodies with enamel-capped crowns known as denticles.⁴ In complex odontomas there is no morpho-differentiation and the lesion appears as a single complex mass with haphazard distribution of enamel, dentine and cementum. The compound odontomas are twice more common than complex odontomas.

The odontomas can also be classified as central or intraosseous (occurring within the bone) and peripheral or extraosseous variety (occurring within soft tissues covering tooth-bearing portions of the jaws).⁵ The central odontomas may erupt into the oral cavity and are known as erupted odontoma. The peripheral odontomas are rare and tend to exfoliate. The eruption and infection of odontomas are uncommon. A case of infected complex odontoma with unusual presentation is reported here.

CASE PRESENTATION

A 14-year-old male patient without systemic disease reported a painless swelling on the left side of the face since childhood and perforation of the left cheek with pus drainage since the last 3 years.

An extra-oral examination revealed a hard swelling on the left side of the lower jaw. The left cheek was perforated with well-formed molar with pus discharge. There was sinus opening and pus discharge anterior to perforation (figure 1). The intra-oral examination revealed missing 35 36 37 with enlarged and lobulated gingiva in edentulous region with exposure of hard, yellowish-white dental tissue-like material with pus discharge (figure 2).

INVESTIGATIONS

The orthopantomograph showed an irregular radio-opaque mass of calcified material in the left mandibular posterior region surrounded by a radiolucent band with smooth outer periphery. The calcified mass shows an affected 38 at postero-inferior aspect of the mandible (figure 3). The clinical–radiological findings are suggestive of infected complex odontoma.

The H&E-stained decalcified section showed an admixture of dentinal tissues arranged in a haphazard manner with serrated borders confirming the clinical–radiographical diagnosis of complex odontoma (figure 4).

DIFFERENTIAL DIAGNOSIS

- **Cemento-ossifying fibroma**—odontomas are more radiopaque than this lesion and are associated with unerupted molar teeth.

Figure 1 Clinical photograph showing molar perforating cheek and pus discharge.

Figure 2 Intraoral photograph with missing 35 36 37 and erupted, infected odontomas posteriorly.

- **Ameloblastic fibro-odontoma** and odontomas are associated with missing tooth. Differentiation from developing odontoma may pose difficulty but this tumour has greater soft-tissue component than odontoma. Mature lesion of complex odontoma will have one mass of disorganised tissue in the centre while this tumour will show multiple scattered dental hard tissue.
- **Periapical cemental dysplasia** may resemble complex odontomas but are usually located in the periapical region of teeth and are multiple in number. However, if a solitary lesion is present, differentiation can be done on the basis that this entity has wider uneven sclerotic margin while odontoma has well-defined cortical margin and uniform soft-tissue capsule.

TREATMENT

The lesion was surgically excised (figure 5) and the molar which was perforating the cheek was extracted.

OUTCOME AND FOLLOW-UP

The healing was uneventful and no postoperative complications were noted. The patient was followed up for 1 year; however, there was no recurrence.

DISCUSSION

Odontomas are considered as hamartomas of aborted tooth formation rather than true neoplasm.⁶ They contain various component tissues of the teeth. They are the most common odontogenic tumours constituting 22%

Figure 4 Photomicrograph showing admixture of dental tissues arranged in a haphazard manner.

of all odontogenic tumours of the jaws.³ The exact aetiology of odontomas is uncertain. It has been attributed to various pathological conditions such as local trauma, inflammatory and/or infectious process, odontoblastic hyperactivity, hereditary anomalies and persistence of a portion of dental lamina, growth pressure and developmental influences.^{7 8}

Odontomas are also seen in hereditary anomalies such as Gardner's syndrome and Hermann's syndrome, suggest that alteration of the genetic component for controlling dental development might be responsible.⁷

The complex odontomas are usually located in the posterior mandible while compound odontomas are more common in the anterior maxilla between roots of erupted teeth or over the crowns of impacted teeth. The complex odontoma of unusual size involving maxillary sinus has been reported in one case.⁹

In general, majority of odontomas are asymptomatic and are discovered on routine radiographic examination. Only few cases are reported with swelling, delayed eruption and in severe cases infection or lymphadenopathy. Sometimes odontoma becomes exposed through soft tissues, that is, erupted odontoma. Until now, only 20 cases of erupted odontoma are reported in the literature.¹⁰

In 70% of odontomas, pathological anomalies are observed in relation to the neighbouring teeth such as devitalisation, malformation, aplasia, malposition and affected teeth.¹¹ The present case also shows the eruption of odontoma in the oral cavity with aplasia of 35 36 37 and affected 38. Also, there was perforation of the cheek

Figure 3 Panoramic radiograph showing radio-opaque mass involving left body and ramus of the mandible.

Figure 5 Enucleated surgical specimen.

with impacted tooth which makes the presentation unusual which has not been reported earlier.

The radiographic characteristics of odontomas are always diagnostic. Radiographically, odontomas present as well-defined, radio-opacities situated in the bone with density greater than bone and equal to that of a tooth. A radiolucent halo, typically surrounded by a thin sclerotic line, surrounds radio-opacity. The radiolucent zone represents a connective tissue capsule similar to that of the normal tooth follicle. In compound odontoma, multiple teeth-like structures of varying size and shape are seen. Complex odontomas are seen as irregular radio dense masses with no resemblance to teeth. Radiographically three different development stages are seen depending on the degree of odontoma calcification. In the first stage, the radiolucent lesion appears due to the lack of calcification, the intermediate stage is characterised by partial calcification and in the final stage the odontoma appears radio-opaque which is surrounded by a radiolucent halo.^{3 7 12 13} A developing odontoma may be discovered on the routine radiographic examination and may present difficulty in diagnosis because of lack of calcification.

Odontomas are well-capsulated lesions and have less chances of recurrence. The management comprises conservative surgical removal. The affected teeth associated with odontomas should be preserved and followed-up clinically and radiologically for at least 1 year. If there is no change in position of the tooth, fenestration followed by orthodontic traction is indicated. The extraction of tooth is indicated if tooth is ectopic, heterotrophic, with morphological alterations or associated with cystic transformation.¹⁴ The healing of these lesions may take 9–12 months in young patients.

The histological appearance of the odontoma is not spectacular. Odontoma contains normal appearing enamel or enamel matrix, dentin, pulp tissue and cementum which may or may not exhibit a normal relation to one another. If the morphological resemblance to teeth does exist, the structures are usually single rooted. The connective tissue capsule around the odontoma is similar in all respects to the follicle surrounding a normal tooth.

Learning points

- ▶ The odontomas constitute a large portion of jaw tumours.
- ▶ Majority of odontomas are asymptomatic and are discovered on routine radiographic examination.
- ▶ Odontomas which erupt into the oral cavity and become infected pose moderately serious condition.
- ▶ The dentist should be familiar with both common and unusual appearances of odontomas for proper diagnosis and management.

Competing interests None.

Patient consent Obtained.

REFERENCES

1. **Cohen DM**, Bhattacharyya I. Ameloblastic fibro-odontoma and odontoma. *Oral Maxillofac Surg Clin N Am* 2004;**6**:375–84.
2. **Philipsen H**, Reichart P, 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;**32**:86–99.
3. **Amado CS**, Gargallo AJ, Berini AL, *et al.* Review of 61 cases of odontoma: presentation of an erupted complex odontoma. *Med Oral* 2003;**8**:366–73.
4. **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.
5. **Junquera L**, de Vicente JC, Roig P, *et al.* Intraosseous odontoma erupted into the oral cavity: an unusual pathology. *Med Oral Pathol Oral Cir Bucal* 2005;**10**:248–51.
6. **Shekar SE**, Rao RS, Gunasheela B, *et al.* Erupted compound odontome. *J Oral Maxillofac Pathol* 2009;**13**:47–50.
7. **Vengal M**, Arora H, Ghosh S, *et al.* Large erupting complex odontoma: a case report. *J Can Dent Assoc* 2007;**73**:169–72.
8. **Hitchin AD**. The etiology of calcified composite odontoma. *Br Dent J* 1971;**130**:475–82.
9. **Mupparapu M**, Singer SR, Rinaggio J. Complex odontoma of unusual size involving the maxillary sinus: report of a case and review of CT and histopathologic features. *Quintessence Int* 2004;**35**(8):641–5.
10. **Serra-Serra G**, Berini-Ayres L, Gay-Escoda C. Erupted odontomas: a report of three cases and review of literature. *Med Oral Pathol Oral Cir Bucal* 2009;**14**:E299–303.
11. **Kaneko M**, Fukuda M, Sano T, *et al.* Microradiographic and microscopic investigation of a rare case of complex odontoma. *Oral Surg Med Oral Pathol Oral Radiol Endod* 1998;**85**:131–4.
12. **Ferrer Ramirez MJ**, Silvestre Donat FJ, Estelles Ferriol E, *et al.* Recurrent infection of a complex odontoma following eruption in the mouth. *Med Oral* 2001;**6**(4):269–75.
13. **Garcia-Consuegra L**, Junquera LM, Albertos JM, *et al.* Odontomas. A clinical–histological and retrospective epidemiological study of 46 cases. *Med Oral* 2000;**5**(5):367–72.
14. **Frank C**. Treatment options for impacted teeth. *J Am Dent Assoc* 2000;**131**:623–32.

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