

The atypical clinical case of infected odontome in geriatric patient: a case report

Amit S. Date^{1^}, Pooja Kamath^{2^}, Treville Pereira^{2^}

¹Department of Oral and Maxillofacial Surgery, D. Y. Patil School of Dentistry, Navi Mumbai, India; ²Department of Oral Pathology, D. Y. Patil School of Dentistry, Navi Mumbai, India

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Correspondence to: Dr. Amit S. Date, MDS, PGCOI, FIBOMS. Professor, Department of Oral and Maxillofacial Surgery, D. Y. Patil School of Dentistry, Sector 7, Highway Road, Dr D. Y. Patil Vidyanagar, Nerul, Navi Mumbai 400706, India. Email: asdate2001@yahoo.co.in.

Background: Odontomas are odontogenic tumors that occur in 2nd–3rd decade of life. They are almost unheard of in 7th–8th decade. They seldom cause symptoms or get infected, and are discovered as incidental radiological findings in majority of cases. Here we report a geriatric case of infected odontome. This case is unique in terms of patient's age and uncommon presentation in the form of intra-oral sinus on alveolar mucosa.

Case Description: We report an 81-year-old woman who was referred to us for evaluation of fistula on alveolar ridge by her general dental practitioner. Due to past medical history of oral bisphosphonate use, medication-related osteonecrosis of the jaws (MRONJ) was our working diagnosis. The fistula failed to resolve following a course of oral antibiotics (co-amoxiclav 625 mg bid for 7 days) and antibacterial mouth rinses with chlorhexidine. However, upon excision and histopathologic examination, it turned out to be infected odontoma. This was a surprising diagnosis at this age. She was successfully treated under local anesthesia in dental office and is disease free till 30th April 2023.

Conclusions: Developmental anomalies like odontome which usually manifests in 2nd or 3rd decade should also be at the back of mind even when evaluating geriatric patients.

Keywords: Fistula; geriatric; mandible; odontome; case report

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Introduction

Background

The odontome is a developmental tumour-like mass consisting of disorderly arranged dental tissues. It has a limited growth potential and can be considered as a dental hamartoma rather than true neoplasm. This is because although epithelial and ectomesenchymal tissues appear

normal morphologically, they have disturbed structural arrangement (1). Odontomas are of two types: complex odontoma and the compound odontoma. The distinction between them is based on either the appearance of well-organized tooth like structures (compound odontomas) or on a mass of disorganized odontogenic tissues (complex odontomas).

They grow slowly, can expand the jaw, and are painless

[^] ORCID: Amit S. Date, 0000-0001-8787-0452; Pooja Kamath, 0000-0001-8368-023X; Treville Pereira, 0000-0001-5662-5235.

mostly. They are often detected on routine radiographs or diagnosed through a failed eruption of a permanent tooth. Based on 137 cases from the odontoma survey the mean age at the time of diagnosis was 19.9 years (range, 2 to 74 years). Almost 84% of cases occurred before the age of 30. Only 1 case was found in the 70–79 years age group. Incidence of associated pain and inflammation was seen in only 4% patients (2). Conservative complete local excision is considered curative for this lesion.

Rationale and knowledge gap

Odontome is a developmental odontogenic anomaly, most cases were observed in young patients in 2nd and 3rd decades of life. Thus they are not included in differential diagnosis when evaluating a geriatric patient in 7th and 8th decades. They seldom cause symptoms or get infected, and are discovered as incidental radiological findings in the majority of cases.

Objective

We report a unique case of infected odontoma in an

81-year-old woman. She was referred to us for evaluation of fistula on alveolar ridge by her dentist. Due to past medical history of oral bisphosphonate use, medication-related osteonecrosis of the jaws (MRONJ) was our working diagnosis. Due to the failure of lesion to resolve following medical management, we explored the area. To our surprise, the fistulous tract was connected to hard calcified mass in the alveolar bone. Upon excision and submission for histopathology, it was reported as complex odontoma. Healing was uneventful and the patient is under regular follow-up. We present this article in accordance with the CARE reporting checklist (available at <https://fomm.amegroups.com/article/view/10.21037/fomm-23-43/rc>).

Case presentation

An 81-year-old woman was referred to us by her general dental practitioner for evaluation of fistula on mandibular alveolar mucosa on 1st March 2022. She sought his consultation for replacement of missing teeth which were removed many years earlier uneventfully. The dentist upon examination found a fistulous opening on the crest of the alveolar ridge on the left premolar area (*Figures 1,2*). The patient was unaware of its existence. She did complain of occasional local dull ache. There was no lip paresthesia or intra-oral discharge. Her past medical history revealed consumption of oral bisphosphonates (osteofos 70 mg once a week) for 3 months to prevent fractures subsequent to post-menopausal severe osteoporosis. Intra-oral periapical radiograph revealed radiopaque mass with irregular outline in the alveolar bone surrounded by thin radiolucent line at places. Computed tomography (CT) scan revealed this lucency continuous with crest of ridge (*Figure 3*).

Due to bisphosphonate use, a working diagnosis of MRONJ was made. We thought it had resulted in sequestrum formation seen on imaging and fistulous opening on mucosa. Other differential diagnoses included infected root pieces, sclerosing osteomyelitis of mandible, bone scar, sclerotic cemental mass, idiopathic osteosclerosis and dense bone island (enostoma). The fistula failed to resolve following a course of oral antibiotics (co-amoxiclav 625 mg bid for 7 days) (3). and antibacterial mouth rinses with chlorhexidine. Therefore the area was explored under local anesthesia. Upon raising mucoperiosteal flap, we found calcified yellowish mass within the bone with surrounding scanty granulation in continuity with fistulous tract (*Figure 4*). This was unlike bony sequestrum which would be found in a case of MRONJ. The calcified

Highlight box

Key findings

- An 81-year-old woman was referred to us for management of fistula on left mandibular alveolus. Due to a history of consumption of bisphosphonates, our working diagnosis was medication-related osteonecrosis of the jaws. The lesion failed to resolve on administration of co-amoxycylav and chlorhexidine rinses. Upon surgical exploration, a calcified mass was found connected with fistulous tract. Histopathological diagnosis was infected odontome.

What is known and what is new?

- Odontomas are odontogenic tumors that occur in 2nd–3rd decade of life. They are almost unheard of in 7th–8th decade. They seldom cause symptoms or get infected, and are discovered as incidental radiological findings in majority of cases.
- Bisphosphonates can cause osteonecrosis of jaw bone in some patients. These are initially managed with antibiotics and antiseptic mouth rinses. Those which fail to resolve are managed surgically.
- As the fistula failed to resolve with antibiotics, it was explored under local anesthesia. However, upon excision and histopathologic examination, it turned out to be infected odontoma. This was a surprise diagnosis at such an advanced age.

What is the implication, and what should change now?

- Odontome can be included as one of the differential diagnoses even when evaluating a geriatric patient.



Figure 1 Frontal facial view. This image is published with the patient consent.



Figure 2 Initial presentation showing fistula (black arrow) on mandibular alveolus.

mass was excised in multiple pieces to avoid fracture of atrophied jaw. Wound healing was uneventful. Sutures were removed at one week. The authors disclose no adverse and unanticipated events during or after surgery. No medication was administered following suture removal. Subsequent histopathological exam revealed soft tissue capsule consisting of loose connective tissue containing strands and islands of odontogenic epithelium. Some areas showed cell rich zone of soft tissue formation of dentin and enamel, not resembling tooth morphology. The lesion appeared



Figure 3 Computed tomography scan panoramic view with arrow showing radiopaque mass involving left alveolus.



Figure 4 Intra-operative view showing hard calcified mass connected with crestal fistula.

as a mass of primary tubular dentin which enclosed hollow circular or oval structures with empty spaces from decalcified mature enamel. The connective tissue showed inflammatory cell infiltration and blood vessels. Diagnosis of complex odontoma was made (*Figure 5A,5B*). Thus consideration of MRONJ which was considered initially upon clinical evaluation was ruled out following histopathological diagnosis. The patient is being followed up regularly and is asymptomatic at her recent 1-year post-operative follow-up (*Figure 6*).

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent

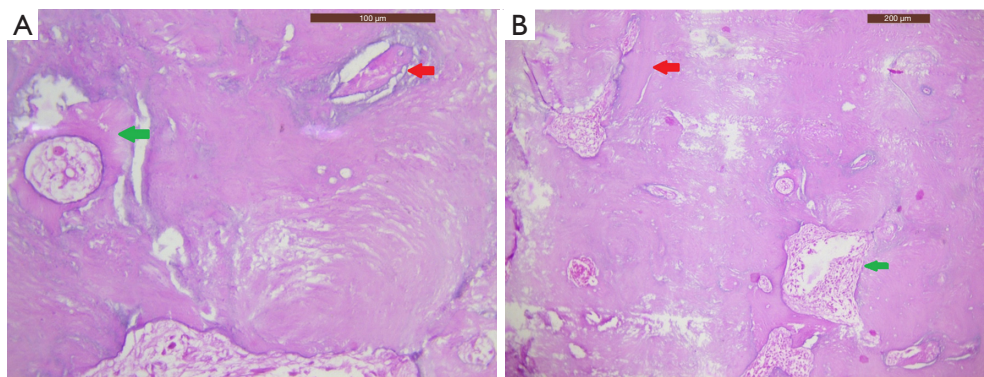


Figure 5 Photomicrograph showing features suggestive of odontome. (A) Green arrow pointing to dentin like tissue enclosing soft pulp tissue surrounded by cementoid. The tissues are arranged disorderly and bear no resemblance to arrangement of tissues in a tooth. Red arrow shows enamel space (empty crescent like space left due to demineralization of enamel) (H&E $\times 400$). (B) Disorganized arrangement of hard and soft tissues of tooth development. Green arrow shows soft tissue resembling pulp. Red arrow shows dentinoid, cementoid- and pulp-like tissue jumbled up with no resemblance to tooth like arrangement (H&E $\times 100$).



Figure 6 Follow-up picture showing complete healing.

was obtained from the patient for the publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

Discussion

Key findings

The 81-year-old lady presented with fistula on mandibular alveolar ridge. It failed to resolve with antibiotics. Upon

surgical exploration, fistulous tract was connected with hard calcified mass within alveolar bone. MRONJ was considered distinct possibility in our case due to a history of consumption of oral bisphosphonates in the past. The drug inhibits osteoclast action and thus interferes with normal bone turnover. This makes jaw bone more susceptible to infection and loss of vitality. American Association of Oral and Maxillofacial Surgeons has published their position paper on this subject recently in 2022 (4). It gave detailed account of medications resulting in bone necrosis, patients at risk, pathophysiology, preventive aspects and management strategies. With reference to this paper, the following is the pertinent information as regards this case.

Clinically they present with dull, aching bone pain in the jaw, which may radiate to the temporomandibular joint region in the earlier stages. In the later stages, regions of osteosclerosis involving the alveolar bone and/or the surrounding basilar bone are seen. There is exposed and necrotic bone intra-orally or fistula that probes to the bone in patients who are asymptomatic and have no evidence of infection/inflammation. The majority of patients on bisphosphonates take a low dose orally for osteoporosis prevention. Their risk of developing MRONJ is less in patients consuming oral bisphosphonates than those patients on a high dosage taken intravenously for malignancy affecting the bones. Nevertheless, it remains a possibility. MRONJ is more likely to appear in the mandible (75%) than the maxilla (25%) (5).

Other diagnostic possibilities considered

- (I) Infected dental root piece. The outline of the mass on CT scan was quite irregular and did not resemble that of premolar root. Hence this was considered unlikely.
- (II) Chronic sclerosing osteomyelitis (sclerosing osteitis) is observed as proliferative osteoblastic response around carious tooth of children and young adults with high tissue resistance. Root or socket outline is clearly visible which is absent in present case (6).
- (III) Sclerotic cemental masses of the jaws. A total of 38 cases were reported by Waldron and his coworkers under the termsclerotic cemental masses of the jaws which produced pain, drainage, or localized expansion. The radiographic appearance was also similar to that of diffuse sclerosis. However these are multiple, symmetric lesions hence ruled out in present case (7).
- (IV) Idiopathic osteosclerosis is generally accepted as developmental intraosseous anatomic variation and characterized by the occurrence of asymptomatic, round, elliptical, or irregular radiopaque mass, in the bicuspid, molar region of the mandible similar to this case (8). It is asymptomatic and may remain static or demonstrate slow growth that usually stops when the patient reaches skeletal maturity. In our case, it is ruled out due to fistula formation and dull ache.
- (V) Dense bone island (enostoma) are incidental findings, consisting of failure of resorption of secondary spongiosa within the trabecular bone (9-11). It has no specific relationship with the dentition unlike the present case.
- (VI) Bone scar. Osteosclerosis commonly follows chronic peri-apical inflammation. It often remains as a sclerotic area of bone after extraction. It does not lead to fistula formation. Hence it was considered unlikely diagnosis (6).

Strengths and limitations

This case is unique for its occurrence of infected odontome in an 81-year-old female in the form of non-healing fistula on mandibular alveolus. A comparison with existing literature revealed no similar cases in English language publications. We believe the odontome within the alveolus may have become secondarily infected upon contact with the oral environment. However, the mechanism remains uncertain. As it is a single case report, further studies may be needed to investigate these findings.

Conclusions

The present case is unusual as odontoma which turned out to be final diagnosis is virtually unheard of at such an advanced age. It seldom presents with secondary infection and fistula formation as in this case. Regardless of the patient's age, odontomas should be ruled out as a potential etiology even in geriatric cases.

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Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at <https://fomm.amegroups.com/article/view/10.21037/fomm-23-43/rc>

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Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for the publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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