peer reviewed **CASE REPORT**

Pindborg tumour of the right mandible

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Abstract

The calcifying epithelial odontogenic tumour, or Pindborg tumour, is a rare benign neoplasm arising from epithelial tissue within the mandible or maxilla. Less than 1% of all odontogenic tumours are classified as Pindborg tumours. The aetiology is idiopathic. This case report explores this pathology as diagnosed on the right mandibular region in a female in her late twenties. The aetiology, epidemiology and radiographic appearance of a Pindborg tumour are discussed.

Keywords

odontogenic, calcification, surgery, un-erupted tooth

Case report

A female in her late twenties was referred from a primary healthcare facility to the outpatient maxillofacial clinic at an academic hospital due to persistent and painless right-sided mandibular swelling. She had initially noticed the formation of a hard nodule two years before the diagnosis; however, regrettable circumstances led to the patient not receiving dental care and treatment. A physical examination was undertaken. An orthopantogram was performed to investigate the right mandibular area. Physical and radiographic findings lead to a provisional diagnosis of a Pindborg tumour, which was subsequently confirmed histologically. As part of her work up, she was referred for an unenhanced mandibular computed tomography (CT) examination. The findings were of a well-defined neoplasm. The tumour was depicted as a heterogeneous solitary expansile multilocular lesion occupying the angle of the right mandible (Figures 1 and 2). It was associated with an un-erupted molar tooth; cortical disruption of the mandible had occurred. Associated tissue masses, perisoteal reactions, other lesions or pathological fractures were not visible on the CT scan. Specific measurements of the tumour were not taken.

Orthopantomography is another effective and ideal imaging modality to assess a Pindborg tumour. The orthopantogram findings showed irregular radiopacities and radiopaque flecks within a well-defined radiolucent area. Figure 3 shows the tumour exhibiting an unilocular and honeycomb appearance. The orthopantomogram demonstrated an un-erupted molar tooth embedded in the body of the right mandible.

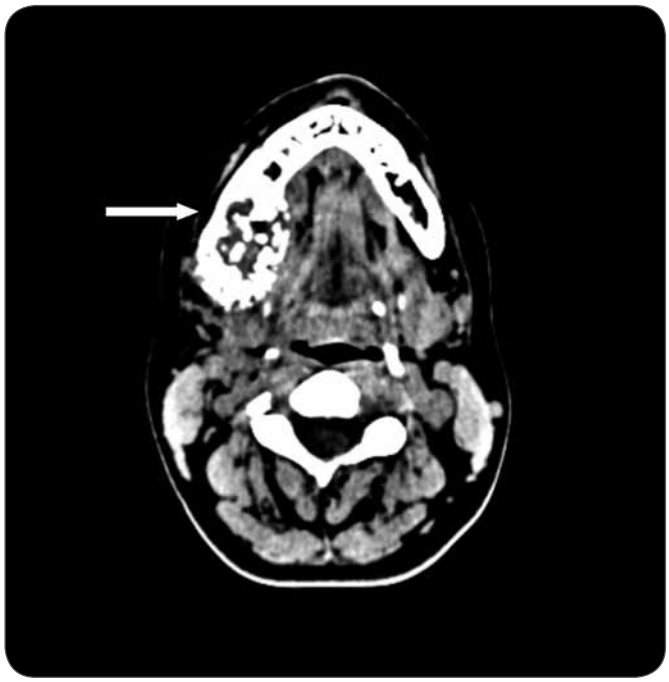

Figure 1. Non-contrast axial CT of the mandible on soft tissue window showing the heterogeneous and multilocular mass (see arrow).

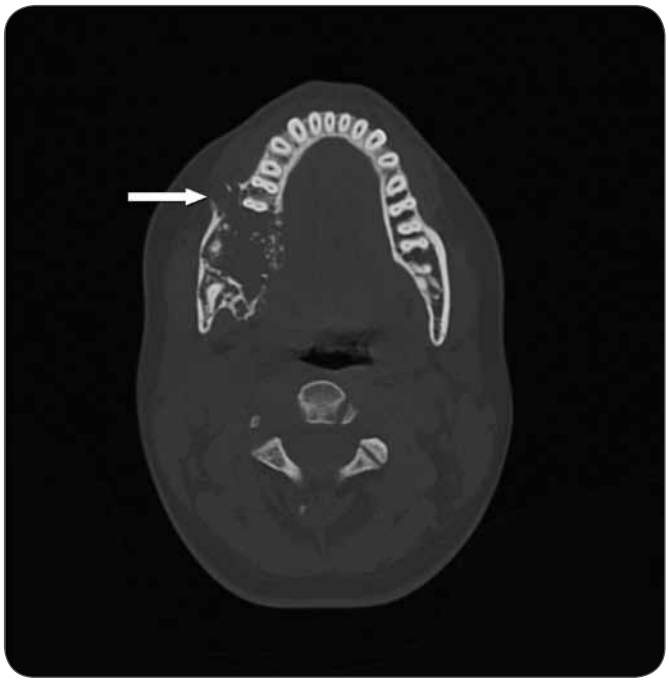

Figure 2. Non-contrast axial CT on bony window showing cortical disruption of the right mandible as a result of the tumour (see arrow).

The patient was admitted to undergo plastic surgery. The disruption of the ipsilateral mandible and the adjacent anterior molars necessitated a right segmental mandibulectomy by means of an intra-oral transbuccal approach. The pathological site was removed and a plate was inserted that stretched from the remaining right ascending ramus to the left mandibular body. A post-operative orthopantogram was performed the day after surgery to confirm correct positioning of the reconstructive plate. The patient recovered well from surgery. Appointments were scheduled to discuss the removal of the reconstructive plate and the possibility of a bone graft to aid in improving her quality of life.

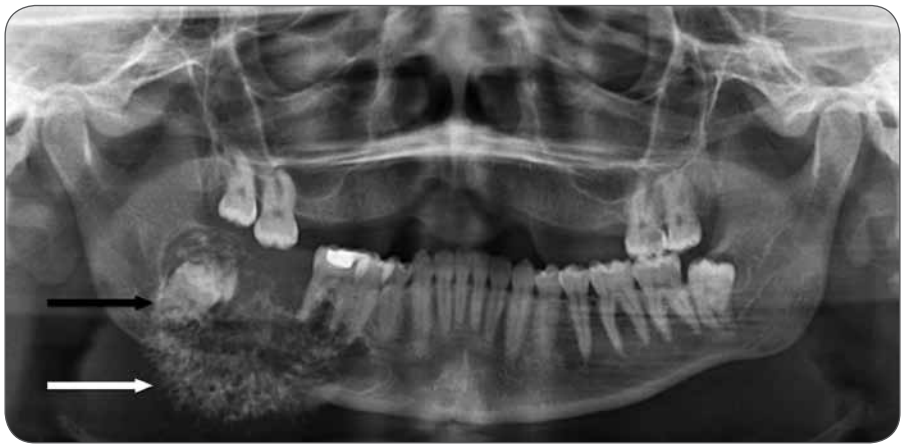

Figure 3. Orthopantogram image illustrating radiopacities within the localised radiolucent area, demonstrating a honeycomb and multilocular appearance in the body of the right mandible (white arrow). The unerupted molar tooth can also be visualised (black arrow).

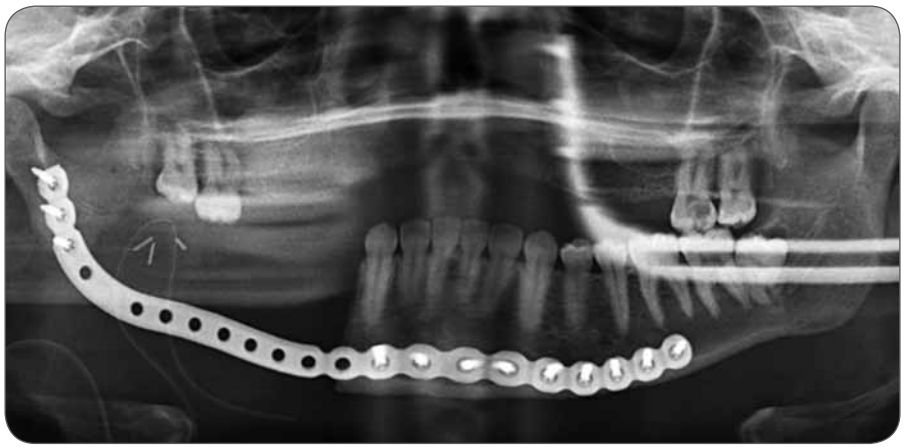

Figure 4. Orthopantogram performed post-segmental resection of the right mandible showing the reconstructive plate that stretches from the remaining right ramus to the left body of the mandible.

Discussion

A calcifying epithelial odontogenic tumour can be described as a benign, aggressive and localizing neoplasm's found primarily within the molar and premolar regions of the mandible and maxilla.[1] This tumour is closely associated with an un-erupted or impacted molar tooth; however, its true aetiology is enigmatic.[1] Less than 1% of all odontogenic tumours are classified as Pindborg tumours.[2] The incidence of an embedded or impacted tooth is commonly affiliated with such a tumour.[3] Characteristically, a Pindborg tumour includes amyloid-like materials with the potential to be calcified and it may be recognised by a painless, slow growing mass leading to mechanical complications.[4] Microscopic appearances of Pindborg tumours consist of numerous calcifications found within nuclei-centred cells arranged as sheets or islands surrounded by an eosinphilic cytoplasm.[5] In spite of these distinct appearances, cases have been reported where Pindborg tumours have developed without the presence of calcifications.

This tumour occurs predominantly in adults between 20 and 60 years old. It arises three times more often in the molar region, as opposed to the premolar region of the mandible.[1] A Pindborg tumour has no gender prediction.[6] Characteristically, it is a slow-growing mass that is typically only recognised by persistent and painless swelling leading to an asymmetrical facial appearance of the patient.[7]

Intraosseous Pindborg tumours, which occur within the mandible and maxilla, are highly associated with an un-erupted tooth. A ratio of 2:1 is used to represent the predilection for an intraosseous tumour to occur in the mandible more often than the maxilla.[2] These aggressive tumours are more common than extraosseous tumours that occur outside the jaw, ordinarily at the gingiva, and present with large amounts of bony erosion. Intraosseous tumours occur in 94% of cases with a recurrence rate of 14%, whilst extraosseous tumours are reported in only 6% of cases.[6]

Pindborg tumours have distinct histopathological factors, which can confirm the diagnosis, as well as separate it from other similar and more common odontogenic tumours. These differential diagnoses include adenomatoid odontogenous tumours, other odontogenous tumours presenting with radiopaque flecks, squamous cell carcinoma, metastatic carcinomas as well as salivary gland tumours.[4]

Cases have been reported where Pindborg tumours developed without the presence of calcifications. These tumours can be categorised into three groups; tumours presenting with Langerhans' cells; tumours displaying cementum-like material; and clear cell tumours. Clear cell Pindborg tumours, which are associated with cortical perforation, are extremely aggressive and recur at a rate of 22%.[8]

Despite the aetiology of a Pindborg tumour being idiopathic, its cellular composition indicates the involvement of the remains of the mucosa of the associated molar tooth within the oral cavity that is produced during the initial stages of odontogenesis. Metachromatic staining occurs during the tumour's exposure to crystal violet; a Congo red staining examination produces a positive result.[9] The Thioflavin T staining technique allows the cytoplasmic make-up to become fluorescent under microscopic light, demonstrating the presence of amyloid-like materials.[1]

Risk factors of this tumour include the possibility of it becoming malignant; continued growth may potentially lead to mechanical complications of the jaw.[4] The only option available for treatment of a Pindborg tumour involves surgery.

Different surgical approaches are used depending on the size and extent of the tumour.[7] Simple enucleation or curettage techniques can be used for treatment of small and intraosseous tumours; while aggressive surgery, such as a hemimandibulectomy, segmental resection or hemimaxillectomy, is used for large tumours. Surgery that involves bone disruptions requires post-surgery interventions; insertion of plates and other reconstructive procedures, such as bone grafts.[2] There may be tumour recurrence if there was incorrect or only partial surgical removal a Pindborg tumour. The prognosis is good. However, it is less favourable in clear cell variants of the tumour.[4]

The patient in this report underwent a successful segmental resection of her mandible with the insertion of a reconstructive plate (Figure 4). Her imaging and histology findings were in keeping reported cases in the literature.

Conclusion

Calcifying epithelial odontogenic tumours are rare benign locally aggressive lesions that occur within the premolar and molar regions of the mandible and maxilla. CT and orthopantogram examinations are the preferred imaging modalities. CT depicts the margins, size and extent of the tumour; orthopantogram studies provide distinct pattern appearances that assist in the diagnosis of a Pindborg tumour. Despite histopathological factors of the tumour being distinct, the aetiology is currently enigmatic. Surgical approaches are the only form of treatment. Follow-up measures are vital after treatment as there is known recurrence of the tumour.[5]

References

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