Case Report
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A case report of calcifying epithelial odontogenic (Pindborg) tumour in the mandible

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The calcifying epithelial odontogenic tumour (CEOT), also known as Pindborg tumour, is an uncommon lesion that included <1% of all odontogenic tumours. This is a rare benign, but locally aggressive odontogenic tumour, usually seen in the posterior area of the mandible and is mostly found in patients between 30 and 50 years of age, without sex predilection. The tumour has a recurrence rate of 10–15% and rare malignant transformation.
Keywords odontogenic tumour, Pindborg tumour, mandible

Introduction

Calcifying epithelial odontogenic tumour (CEOT), also known as Pindborg tumour, is an uncommon tumour, including <1% of all odontogenic tumours.[1–3] The tumour was first described in 1956 by the Late Dr. Jens J Pindborg.[1] It is usually intraosseous and mostly occurs in the jaw bones. The mandible is affected twice more than the maxilla. The most involved site is the mandibular posterior area.[4] The tumour shows a variable radiographic view based on its development; mixed radiolucent–radiopaque feature is the most prevalent, seen in 65% of cases.[5,6] The mean occurrence age is 40 years with a range between 2nd and 6th decades.[4] Here, we present a rare case of huge Pindborg tumour in the mandibular body of a 48-year-old man.

Case Report

A 48-year-old male patient was referred to Babol dentistry school, Babol, Iran, for fixed prosthetic treatment. During examinations, an asymmetry with a swelling at posterior area of the left side of his mandible was observed (Fig. 1). The patient was aware of the lesion from 15 years ago, but, as there were no sign or symptom, he did not seek treatment. His past medical and habitual history was otherwise clear. On extra oral examination, we found a well-defined bony hard swelling in the mandibular molar area extending to the angle. The overlying skin was intact with no tenderness. On intra-oral examination, the mass was palpated with intact mucosal coverage (Fig. 2). In his panoramic view, the multilocular lesion measuring was 7 × 3 cm, with coarse trabeculae and radiopaque foci and impacted molar tooth was detected in the left mandibular body. The inferior border of mandible and alveolar crest in the lesion area were expanded and the third molar tooth was displaced to the border of the mandible (Fig. 3). An incisional biopsy under local anaesthesia was performed (Fig. 4) and the sample was sent to the pathology laboratory for histopathological evaluations. The microscopic views showed an odontogenic tumour composed of nests and islands of epithelioid cells with oeosinophilic cytoplasms. The production of amyloid-like material was evident, so Pindborg tumour was considered as a diagnosis (Figs. 5, 6). The patient was scheduled for a surgical excision and reconstruction as treatment plan.

Discussion

The CEOT, which also known as Pindborg tumour, is an uncommon lesion that included <1% of all odontogenic tumours. Approximately about 200 cases have been reported to date.[7] Pindborg tumour was previously described in the literature as adenoid adamantoblastoma, ameloblastoma of unusual type with calcification.[8] Thoma and Goldman described the tumour as a neoplasm arising from the odontogenic epithelium; subsequently, the German pathologist Jorgen Pindborg recognised it as a separate entity in 1955, and in honour of him, this lesion was termed as the Pindborg tumour.[4] In 1967, Abrams and Howell reported the first case of CEOT consist of clear cells.[9] The term 'CEOT' has been generally accepted by the WHO in the first edition of 'Histological Typing of Odontogenic Tumours, Jaw Cysts and Allied Lesions', where it was recognised as a distinct entity. For more than 30 years, the CEOT has been known widely as 'Pindborg tumour'.[10]

CEOT is a rare benign, but locally aggressive odontogenic tumour.[11] It is a slow-growing neoplasm, which has a recurrence rate of 10–15% and rare malignant transformation.[12] This neoplasm is mainly intraosseous with a strong tendency to the mandible.[13] Peripheral tumours usually arise in the anterior gingiva and account for <5% of cases. Tumour histogenesis is not exactly clear, but it is believed to arise from remnants of dental lamina and stratum intermedium.[13,14] Odontogenic tissue is able to produce dentin and enamel because of the interactions between odontogenic mesenchyme and epithelium. Thus, when odontogenic tissue undergoes tumoural changes, it can produce abnormal calcifications resembling enameloid, dentinoid and cementum in histologic features.[15] Clinically, Pindborg

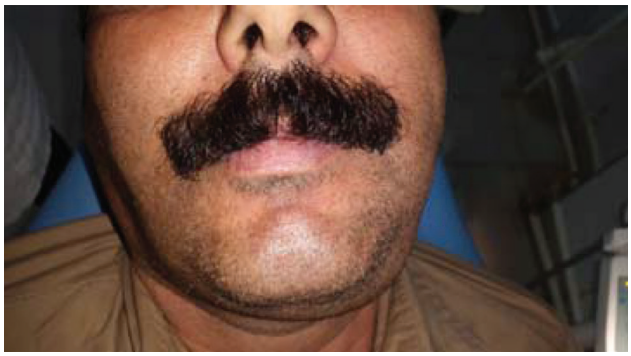

Fig. 1 **Clinical view showing facial asymmetry.**

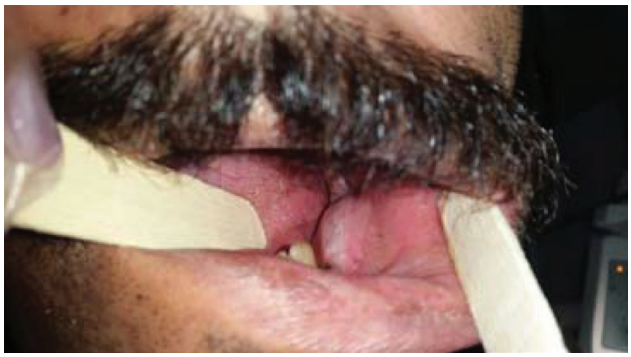

Fig. 2 **Exophitic growth on the left side of mandibular ridge.**

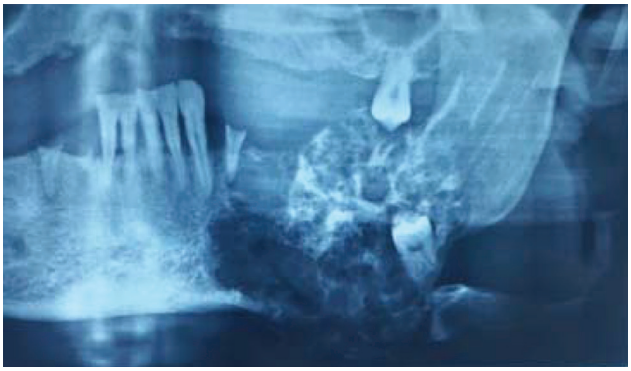

Fig. 3 **A multilocular mixed radiolucent–radiopaque lesion.**

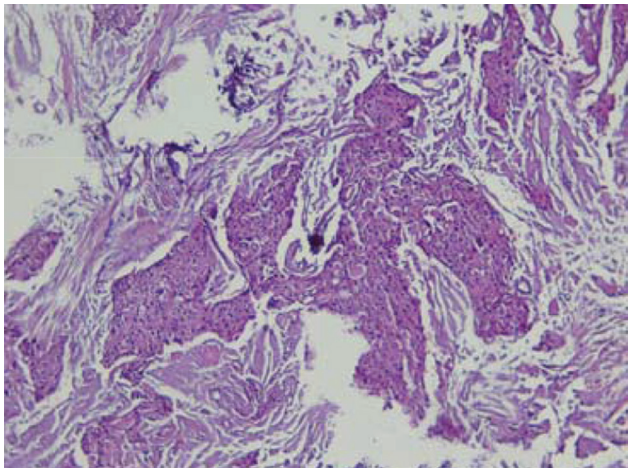

Fig. 5 **Histopatholoic view showing tumoural nests (×100).**

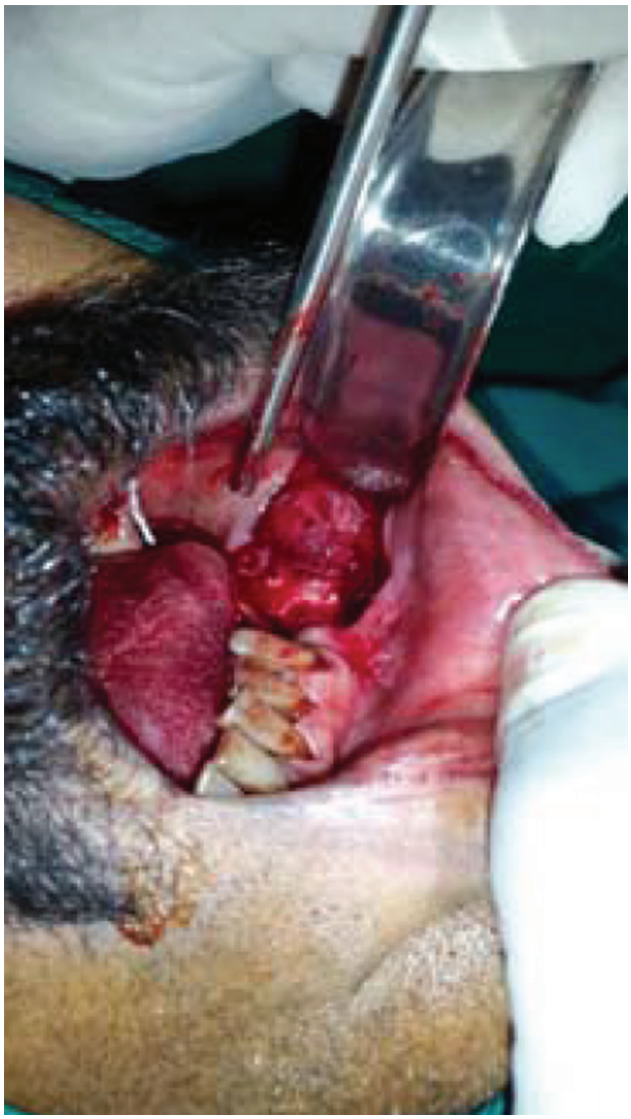

Fig. 4 **Surgical view.**

tumour usually is seen in the posterior area of mandible and is mostly found in patients between 30 and 50 years of age, without sex predilection.[16] This case was also seen in mandibular premolar molar area of a 48-year-old male patient. The most common clinical features of CEOT, when detectable, are a localised swelling of the involved jaw. Pain or paresthesia may exist which is depended to the size of the tumour, the growth pattern or its location, and proximity to the neurovascular structures.[17] Our case had no pain or paresthesia, despite the noticeable size of the tumour and its long duration. Radiographically, CEOT is characterised as a unilocular or multilocular radiolucent lesion that often exhibits a mixed radiopaque–radiolucent pattern. The mixed pattern shows areas of scattered flecks of calcification (driven snow pattern).[5,6] However, calcifications sometimes, may not be observed on radiographs.[17] Our case also revealed a radiolucent–radiopaque mass

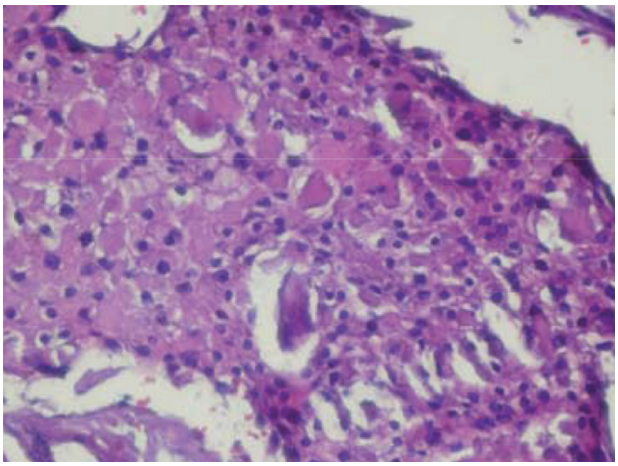

Fig. 6 **Histopathologic view showing amyloid-like material production (×400).**

without driven snow appearance. Association with impact teeth are often seen,[13] but was not seen in our case. CEOT is microscopically characterised by cords and nests of round to polygonal oeosinophilic epithelial cells with nuclear pleomorphism and conspicuous intercellular bridges in a fibrous stroma that typically contains variable amounts of the Congo red positive amyloid-like material and calcified structures.[18–20] Epithelial Island, pleomorphism and amyloid-like material were seen in our case, and amyloid presence was confirmed by Congo red staining.

In the mandible, the treatment of choice is marginal resection with a rim of normal tissue. Wide resection seems to be unnecessary in typical cases. In the maxilla, more aggressive treatment should be done because of the rapid-growth potential of the neoplasms.[18] Our treatment plan was an excisional surgery with reconstruction, and the patient was asked to attend follow-up sessions to monitor the recurrence. ■

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