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CASE REPORT

Multiple root resorption as a presenting sign of Paget's disease of bone

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Abstract

Paget's disease is a chronic disorder of bone remodelling of uncertain aetiology. Its craniofacial manifestions may be the first indicators of disease. We present an unusual case of Paget's disease presenting with significant external root resorption. The oral manifestions and radiological features of Paget's disease as well as the implications for surgery in the mouth are discussed.

Introduction

Paget's disease of bone, or osteitis deformans, is a disease characterised by abnormal bone deposition and resorption. Craniofacial manifestations have been reported, typically involving the calvarium, maxilla and, on rare occasions, the mandible[1]. Tooth roots are affected by increased deposits of cementum (hypercementosis)[2–4]. Root resorption has previously only been reported in the mandible[3].

The condition mainly affects middle aged or elderly persons. They may complain of bone pain, or deafness or blindness due to cranial nerve compression. When the facial skeleton is affected, the entire maxilla or mandible is usually involved and this may result in alveolar ridge expansion. We report an unusual case of Paget's disease associated with extensive root resorption of the dentition.

Case report

A 72-year-old man presented to his dentist with an 8-week history of pain, swelling and purulent discharge associated with the lower left first molar tooth. Extraction of the tooth revealed severe root resorption prompting the dentist to refer the patient to the oral surgery clinic.

Past medical history and systems review was unremarkable. Clinical examination revealed extensive bucco-lingual expansion of the mandibular alveolus. Dental panoramic tomogram (DPT) (Fig. 1) and lateral skull (Fig. 2) radiographs showed typical features of Paget's disease in the maxilla, mandible, calvarium and skull base. These included hypercementosis, loss of lamina dura, altered bony trabeculation, osteolytic and osteosclerotic areas and bone enlargement with generalised loss of cortical bone. An interesting finding was that of significant external root resorption of the 35, 44, 46 and 47 teeth.

The patient was referred to a general physician for confirmation of the diagnosis of Paget's disease and further management. A bone scan was performed which demonstrated intense uptake of technetium-labelled medronate throughout the skull vault and facial skeleton with focal uptake in the 12th thoracic (T12) and 4th lumbar (L4) vertebral bodies. A serum alkaline phosphatase test showed an elevated level of 2222 U/L, with a normal adult range of 70–330 U/L. Serum calcium was within normal limits. These

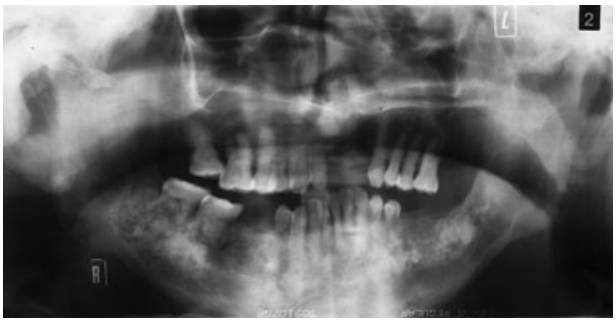

Figure 1 DPT showing Pagetoid changes of the bone of the mandible and maxilla. Note the marked resorption of the 35, 44, 46 and 47 teeth.

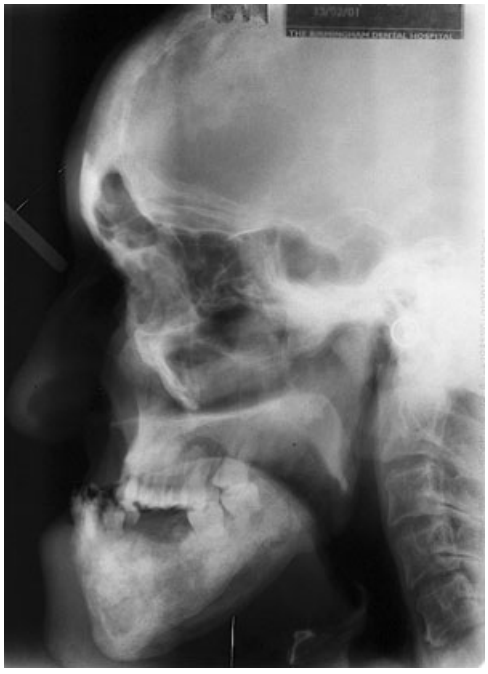

Figure 2 Lateral facial bone radiograph showing marked thickening of the anterior aspect of the mandible, skull base and frontal bone. Areas of diffuse osteosclerosis within the mandible and external root resorption of the 47 tooth is also evident.

results are consistent with a diagnosis of Paget's disease.

As the patient required multiple extractions and recontouring of the jaws, bisphosphonate therapy, to suppress disease activity, and prophylactic antibiotics were commenced prior to surgery.

Discussion

Paget's disease of the jaws appears to be less common than several decades ago. However, it is important to be aware of the features of this disorder and its dental manifestations. Its prevalence is reported to be between 0.01% and 3% in patients above the age of 40[2], increasing to 10% in patients above the age of 70 and more commonly occurring in individuals of Anglo-Saxon origin. The aetiology of Paget's disease is uncertain. It affects certain bones, typically sacrum, spine, femur, tibia and skull, usually in a symmetrical distribution[5]. In a large series[6], jaw involvement was reported to occur in 17% of Paget's cases. The maxilla is more commonly affected than the mandible. Oral manifestations of the disease include enlargement of the alveolar ridge resulting in separation of the teeth, flattening of the palate, malocclusion and pulpal calcification[2,5].

The radiographic features of Paget's disease of the skull vary with the stage of the disease. The bone progresses through an osteolytic and osteoblastic phase which can occur simultaneously but with the osteoblastic phase ultimately becoming dominant. The osteolytic resorptive phase is characterised by radiolucent lesions with a fine trabecular pattern producing a ground glass appearance[2]. In the osteoblastic phase, irregular areas of increased bone formation give rise to a distinctive cotton wool appearance on skull radiographs[2,5]. Thickening of the outer table of the skull vault leads to a loss of demarcation between the diploe[5]. In the jaws, the bone appears granular or may show a linear trabecular pattern posteriorly in the mandible.

Radiographs of the teeth may show loss of lamina dura, hypercementosis, root resorption and pulpal calcification[2,4,5]. Root resorption may result in increased mobility and migration of teeth. However, root resorption accompanied by ingrowth of pagetoid bone or hypercementosis may result in ankylosis. A surgical approach is advisable for the removal of such teeth[5].

This case is unusual because of the extensive root resorption affecting the mandibular teeth, a feature not normally associated with Paget's disease[4]. An explanation for root resorption is that the osteolytic and osteoblastic phases affecting the bone probably also affect the teeth, particularly as bone and cementum have a similar structure. Thus the osteoblastic phase is probably responsible for the formation of hypercementosis and the osteolytic phase for root resorption. In this case, it would appear that the osteolytic phase persisted resulting in extensive root resorption.

Dental surgery in Paget's disease requires careful preoperative planning. Extractions can be difficult because of hypercementosis resulting in bulbous tooth roots or ankylosis, thereby necessitating a surgical approach. In addition, the poor quality of the bone

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renders it susceptible to infection and prophylactic antibiotic therapy should be maintained during the healing phase. Reducing the activity of the abnormal bone with bisphosphonates should help reduce the likelihood of subsequent infection.

Conclusion

Extensive root resorption in Paget's disease of the mandible is an uncommon finding[2,3,5]. Gross resorption of the roots of permanent teeth should always be considered abnormal, particularly in the absence of trauma, tumour or cyst formation[4].

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