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內文:

Abstract

- ♦ Paget's disease is a chronic disorder of bone remodeling of uncertain etiology. We present an unusual case of Paget's disease presenting with significant external root resorption.
- ♦ The oral manifestations 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 (osteitis deformans*), is a disease characterized by abnormal *bone deposition* and *resorption*.
- ♦ Craniofacial manifestations have been reported, typically involving the *calvarium*, *maxilla* and, on rare occasions, the *mandible*1.
- ♦ 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*.
- → Tooth roots are affected by increased deposits of cementum (hypercementosis)2–4.
- ♦ We report an unusual case of Paget's disease associated with extensive root resorption of the dentition.

Case report

- 1. A **72-year-old** man presented an 8-week history of pain, swelling and purulent discharge associated with the lower left first molar tooth.
- 2. Extraction of the tooth revealed severe *root resorption*
- 3. Past medical history and systems review was unremarkable.
- 4. Clinical examination revealed extensive *bucco-lingual expansion* of the mandibular alveolus.
- 5. 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 generalized loss of cortical bone.
- 6. An interesting finding was that of significant external *root resorption of the 35*, 44,46 and 47 teeth.
- 7. A bone scan was performed which demonstrated intense uptake of *technetium-labelled*(鎝; Tc) medronate throughout the skull vault and facial skeleton with focal uptake in the 12th thoracic (*T12*) and 4th lumbar (*L4*) vertebral bodies.
- 8. 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 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

Characteristics

- ♦ Its prevalence is reported to be between 0.01% and 3% in patients above the age of 402, increasing to 10% in patients above the age of 70 and more commonly occurring in individuals of <u>Anglo-Saxon</u> origin.
- ♦ The <u>etiology</u> of Paget's disease is uncertain. It affects certain bones, typically sacrum, spine, femur, tibia and skull, usually in a symmetrical distribution5.
- ♦ In a large series6, 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.
 - A. The osteolytic resorptive phase is characterised by radiolucent lesions with a fine trabecular pattern producing a *ground glass appearance*2.
 - B. In the osteoblastic phase, irregular areas of increased bone formation give rise to a distinctive *cotton wool appearance* on skull radiographs2,5.
- → Thickening of the outer table of the skull vault leads to a loss of demarcation between the diploe5.
- ♦ In the jaws, the bone appears granular or may show a *linear trabecular pattern* posteriorly in the mandible. (*horizontal direction in the mandibuble but randomly oriented in the maxilla*)
- ❖ Radiographs of the teeth may show loss of lamina dura, hypercementosis, root resorption and pulpal calcification2,4,5. 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 teeth5.
- 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.

Management

- ♦ 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 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.
- ♦ Diet and Exercise

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 formation4.

References

- 1. Swartz JD, Vanderslice RB, Korsvik H, Saluk PH, Popky GL, Marlowe FI *et al*. High resolution computed tomography: part 6, craniofacial Paget's disease and fibrous dysplasia. Head Neck Surg 1985;8:40–7.
- 2. Barnett F, Elfenbein L. Paget's disease of the mandible a review and report of a case. Endod Dent Traumatol 1985;1:39–42.
- 3. Gergely JM. Monostotic Paget's disease of the mandible. Oral Surg Oral Med Oral Pathol 1990;70:805–6.
- 4. Smith NHH. Monostatic Paget's disease of the mandible presenting with progressive resorption of the teeth. Oral Surg 1978;46:246–53.
- 5. Smith BJ, Eveson JW. Paget's disease of bone with particular reference to dentistry. J Oral Path 1981;10: 233–47.
- 6. Stafne EC, Austin LT. Study of dental roengenograms in cases of Paget's disease (osteitis deformans), osteitis fibrosa cystica and osteoma. JADA 1938;25:1212–14.

key words

<u>BISPHOSPHONATES</u> (also called: diphosphonates) are a class of drugs that inhibit <u>osteoclast</u> action and the <u>resorption of bone</u>. Its uses include the prevention and treatment of <u>osteoporosis</u>, <u>osteitis deformans</u> ("Paget's disease of bone"), bone <u>metastasis</u> (with or without <u>hypercalcaemia</u>), <u>multiple myeloma</u> and other conditions that feature bone fragility.

<u>ANGLO-SAXON</u> is the term usually used to describe the peoples living in the south and east of <u>Great Britain</u> from the early <u>5th century</u> AD to the <u>Norman conquest</u> of 1066. **POSSIBLE FACTORS:**

- A. Infection factor: Paget's disease may be caused by a <u>slow virus</u> infection (i.e., <u>paramyxoviruses</u> such as <u>measles</u>, <u>Canine distemper virus^[2]</u>, and <u>respiratory</u> syncytial virus);
- B. Hereditary factor: Prevalence of familial Paget's disease (where more than one family member has the disease) ranges from 10 to 40 percent in different parts of the world. Because early <u>diagnosis</u> and treatment is important, after age 40, siblings and children of someone with Paget's disease may wish to have an <u>alkaline phosphatase</u> blood test every 2 or 3 years. If the alkaline phosphatase level is above normal, other tests such as a bone-specific alkaline phosphatase test, bone scan, or x-ray can be performed.

DIAGNOSIS:

Pagetic bone has a characteristic appearance on \underline{x} -rays. A $\underline{skeletal\ survey}$ is therefore indicated.

An elevated level of <u>alkaline phosphatase</u> in the blood in combination with normal <u>calcium</u>, <u>phosphate</u>, and <u>aminotransferase</u> levels in an elderly patient are suggestive of Paget's disease.

Bone scans are useful in determining the extent and activity of the condition. If a bone scan suggests Paget's disease, the affected bone(s) should be x-rayed to confirm the diagnosis.

Diet and Exercise

In general, patients with Paget's disease should receive 1000-1500 mg of <u>calcium</u>, adequate <u>sunshine</u>, and at least 400 units of <u>vitamin D</u> daily. This is especially important in patients being treated with bisphosphonates. Patients with a history of kidney stones should discuss calcium and vitamin D intake with their physician. <u>Exercise</u> is very important in maintaining skeletal health, avoiding <u>weight gain</u>, and maintaining joint mobility. Since undue stress on affected bones should be avoided, patients should discuss any exercise program with their physician before beginning.

beginning.		
題號	題目	
1	Paget's disease最不常involve到的骨頭為以下何者?	
	(A) Pelvis	
	(B) Spine	
	(C) Tibia	
	(D) Radius	
答案(D)	出處:different diagnosis of oral & maxillofacial lesions 5 th edition p.407	
題號	題目	
2	下列何者不適合做為Paget's disease的診斷依據?	
	(A) Alkaline phosphatase test過高	
	(B) Smear slides檢查口腔中有真菌菌落	
	(C) Panorax, LA films 上有skull, maxilla, mandible骨頭沉積	
	(D) Bone scan 上有多處骨頭高活性反應	
答案(C)	出處: different diagnosis of oral & maxillofacial lesions 5 th edition p.511	