CASE REPORT

The importance of cone beam CT in the radiological detection of osteomalacia

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Although osteomalacia is one of the most common osteometabolic diseases among the elderly, there is no case in the literature that presents the effects of osteomalacia in detail using cone beam CT (CBCT). While thin and porous bones are the most common radiographic sign of the disease, the radiological hallmarks are pseudofractures (Looser's zone). We coincidentally detected osteomalacia in a 23-year-old female and we showed the pseudofracture on CBCT images. In the present case, we aim to present the images of osteomalacia that were detected by CBCT in detail. CBCT has an important value in screening for osteomalacia.

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Introduction

The bone tissue is a highly dynamic structure throughout life. Bone mineral metabolism moves toward bone formation and modelling, promoting skeleton growth through changes in bone dimensions, geometry and density.¹ The bone remodelling process occurs continuously throughout life and consists of substituting old bone with new bone.¹ Inadequate extracellular levels of calcium and inorganic phosphate, minerals which are necessary for new bone to properly calcify, results in rickets and osteomalacia.² In these diseases, osteoid (the matrix for new bone formation) builds up in excessive amounts because of its failure to mineralize properly. The age of the patient is the principal difference between these two diseases. Osteomalacia in children is known as rickets, and because of this the use of the term osteomalacia is often restricted to a milder, adult form of the disease.² Rickets and osteomalacia can result from vitamin D deficiency (nutritional or metabolic disturbances), calcium and/or phosphorus deficiency and primary bone mineralization defects (hereditary hypophosphatasia).¹ A common cause of the disease is a deficiency in vitamin D, which is normally obtained from the diet and/or sunlight exposure.² In nutritional osteomalacia, failure to absorb dietary vitamin D may be a result of various gastrointestinal malabsorption

problems, chronic kidney disease or some types of liver disease.² There are other types of osteomalacia that are familial (genetic) diseases, such as hypophosphatasia and hypophosphatemia. Hypophosphatasia usually has the pattern of inheritance that is characterized by an autosomal dominant mode of disease transmission. Hypophosphatemia (also called vitamin D-resistant rickets) is inherited in an X-linked dominant fashion, thus males tend to be more severely affected than females.² Because of this, in the diagnosis of this disease it is important to remember inheritance.¹ The diagnosis of osteomalacia is based on the clinical picture, laboratory and radiograph alterations.¹ In laboratory findings, it produces abnormal biochemical findings, including elevated alkaline phosphatase values with normal or decreased serum calcium and phosphorus and parathyroid hormone values (secondary hyperparathyroidism).³ In most cases, the disease can be diagnosed by assaying for 25-OH-vitamin D.³ A biopsy is required for a definitive diagnosis.⁴ Osteomalacia is one of the most common osteometabolic diseases among the elderly.³ Patients affected by osteomalacia frequently complain of diffuse skeletal pain and their bones are susceptible to fracture with relatively minor injury.⁵

Although patients with osteomalacia have thin bones as a rule, this fact contributes little to routine diagnosis because cortical thickness *per se* is a variable feature within different people. Thus, the radiological hallmarks of the disease are pseudofractures. Radiologically, pseudofractures are seen most commonly in all stress-bearing

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Figure 1 Conventional panoramic radiography showed rarefaction and porosity in the jaws

Figure 2 Osteomalacia; axial cone beam CT image. Note the hypodense appearance of trabeculae and cortex of mandible and Looser's fracture (white arrows)

bones *e.g.* the pelvis, spine and long bones in the legs.² In addition, pseudofracture in the jaws near the angle has also been reported.⁶ Osteomalacia in the jaws can be detected by conventional radiographic techniques such as panoramic and intraoral periapical radiographs in

dentistry.² In addition, it can be detected by cone beam CT (CBCT), which is used in dentistry to image high contrast objects such as teeth and bone.^{2,7} CBCT has advantages compared with conventional radiographic techniques.

This case report focuses on the radiological diagnosis of osteomalacia in the jaws by using CBCT in an adult person.

Case report

A 23-year-old female had visited the Department of Oral Diagnosis and Radiology, Dentistry Faculty, Ataturk University for routine dental care. There were no problems with her general medical history. She had not undergone medical therapy for a systemic disease. No fractures were noted in her history. In anamnesis, there was no history of systemic skeletal disease in her family. The patient was found generally in a good condition. Both extraoral examination and intraoral examination revealed no significant abnormalities. She had good oral hygiene. To detect her general dental status, panoramic radiography was performed. In the panoramic radiography, we detected an overall radiolucent

Figure 3 Osteomalacia was shown in the sagittal and coronal cone beam CT images. Note the hypodense appearance of mandible, maxilla (a) (large white arrows) and C2, C3, C4 vertebrae (b,c) (small white arrows)

Figure 4 Osteomalacia; coronal cone beam CT image. Cortical and trabecular bone destructions (Looser's fracture) in the mandibular bone (white arrows)

porous appearance in the bone (Figure 1). We decided to perform a CBCT scan for a more detailed examination of the jaw bones. The images were obtained by using CBCT (NewTom-FP; Quantitative Radiology, Verona, Italy) scans with 0.2 mm slices in the axial planes, 2 mm slices in the coronal planes and 2 mm slices in the sagittal planes and three-dimensional (3D) images (Figures 2–6). The CBCT scanning was done with the patient positioned supinely and the head position adjusted in such a way that the hard palate was parallel to the floor, while the sagittal plane was perpendicular to the floor. The CBCT unit has an automatic exposure control system (AEC). Imaging parameters were as follows: 110 kV; 3.5 mA; field of view, 130 × 170 mm; voxel size, 0.125 mm³. On the CBCT images, we detected a hypodense appearance of the jaw bones, except for both sides of the mandibular ramus region. In addition, the lamina dura had completely disappeared around the roots of the teeth (except for 38). Cortical and trabecular bone destructions was also observed (Figures 2–6). We thought that the patient had a systemic skeletal disease such as osteoporosis or osteomalacia. The patient was referred to clinical chemistry

and laboratory medicine for a laboratory exam. The laboratory test results showed that the serum calcium was low (6.4 mg dl⁻¹—reference range 8.6–10.2 mg dl⁻¹); phosphate was normal (3.8 mg dl⁻¹—reference range 2.7–4.5 mg dl⁻¹); bone alkaline phosphatase was high (44 U L⁻¹—reference range 0–1 U L⁻¹); parathyroid hormone (PTH) was high (76 pg ml⁻¹—reference range 7–62 pg ml⁻¹); and serum 25-OH-vitamin D level was low (9.9 ng ml⁻¹—reference range 11–43 ng ml⁻¹). After the laboratory test results, these findings led us to hypothesize that the patient had osteomalacia. The patient was referred to physicians for medical evaluation and treatment.

Discussion

In osteomalacia there is a re-modelling of bone in the absence of adequate calcium that results in a softening and distortion of the skeleton and an increased tendency towards fracture in the disease.^{3,8} Radiologically, there are severe asymmetric deformities of all stress-bearing bones.⁹ Essentially, the radiological sign of osteomalacia is pseudofractures, also called Looser's zones. Pseudofractures are seen as poorly calcified, ribbon-like zones extending into bone at approximate right angles to the margin and are seen most commonly in the weight-bearing bones.² Osteomalacia in which a pseudofracture was observed in the jaw near the angle has also been described.⁶ Looser's zones are bilaterally near-symmetrical bands of uncalcified tissue.⁶ Lemay and Blunt suggested that Looser's zones were caused by continuous pulsation beating against a softened cortex.¹⁰ Steinbach *et al* claimed to confirm this theory by arteriography, showing some of the pseudofractures to be in proximity to an abnormal obturatory artery in one case.¹¹ Jackson

Figure 5 Osteomalacia; cone beam CT, sequential cross-sectional images. Note the hypodense appearance of mandible and maxilla; (a) and (f) molar region, (b) and (e) premolar region, (c) and (d) incisive region

Figure 6 Osteomalacia; three-dimensional cone beam CT reconstructed images. Cortical destruction in jaws: (a) anterior view, (b) right sagittal view, (c) left sagittal view and (d) inferior view. Inferior depression areas on the mandibular bone are shown (white stars)

et al criticized this theory and concluded that the zones were not in close connection with pulsating blood vessels.⁶ But in their study, they also suggested that the arterial origin of Looser's zone was only partly true in the mandible.⁶ In our case, we observed that the zones in the mandible were bilaterally near-symmetrical bands and appeared to have possible arterial relationships (Figure 4). In addition, this possible relationship was seen especially in the mandibular symphysis region in 3D images because of its intensive arterial content, such as the lingual artery, sublingual artery, submental artery and mylohyoid artery, when compared with the mandibular ramus region. However, to confirm this possible relationship for mandibular osteomalacia, specific studies designed for this aim should be performed in the

future. In this case, we also observed the pseudofracture (Looser's zone) in the left and right premolar region on CBCT images (Figure 4). On panoramic projections, there may be an overall radiolucent appearance.² Also, trabeculae of bone may be sparse and unusually coarse. The lamina dura may be especially thin in individuals with long-standing or severe osteomalacia. The teeth are not altered in this condition in as much as they are fully developed before the onset of osteomalacia. In most cases of osteomalacia, it is useful to remember that no radiographic manifestations are apparent in the jaw bones.² However, in our case, thinning mandibular cortical structures such as the inferior mandibular border, the walls of the mandibular canal and the lamina dura were observed on CBCT images (Figures 2–5). In

addition, effects of osteomalacia were observed in the upper jaw, especially in maxillary left and right tuberosity regions. Zygomatic bone and cervical vertebrae (C1–C4) were also affected by this condition (Figure 3). Within the spongy portion of the jaws, the fine trabeculae became reduced in number. CBCT is well suited for imaging the craniofacial area,⁷ providing images with submillimetre resolution.⁷ The true nature of the alveolar bone topography around teeth may be assessed by CBCT. In the CBCT, the exact nature and severity of bone destruction can be assessed from just one scan.⁷

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Conclusions

CBCT has advantages compared with conventional radiological techniques such as panoramic radiography. Unfortunately, detailed examinations as in this case may not be performed with conventional radiological techniques because of the superimpositions of the anatomical structures of maxillofacial area. The use of CBCT, especially in complex bone destruction, gives more true and detailed information for diagnosis when compared with conventional radiographic techniques.