

## Case report

# Simple bone cyst associated with florid osseous dysplasia: 2 case reports

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**Abstract** – The occurrence of a simple bone cyst (SBC) is known to be more frequent among the patients affected by florid osseous dysplasia (FOD). Conventional treatment of SBC involves surgical fenestration of the cavity, which frequently leads to resolution. However, in the cases of large cavities, such as those found in cases of FOD, the advisability of surgical intervention remains in question. Two cases of SBC associated with a FOD are reported in two African women (Cameroonian 36 years old and Gabonese 72 years old). The large size of the SBC cavities appears unusual and exceptional being possibly the largest (12 cm diameter) reported case in the literature). Following surgical exploration and fenestration in these cases, the size of the SBC did not stop increasing, though a significant amount of new bone clearly appeared at the fenestration sites seen on the control CT scans (1 year later). In the light of the two presented cases, the outcome of the surgical treatment of large SBC associated with a FOD appears unconvincing, as the cavities did not decrease in volume. Though the patient did not appear to suffer any detrimental effects of surgical intervention, the benefit of such intervention remains in question and the outcome in these two case studies suggests that in the absence of symptoms, long term follow up is all that is necessary.

### Mots clés :

dysplasie osseuse  
floride / kystes  
maxillaires / kystes  
osseux solitaires /  
CT scan

**Résumé** – **Kyste osseux solitaire associé à une dysplasie osseuse floride. Présentation de 2 cas.** L'apparition d'un kyste osseux solitaire (KOS) est plus fréquente chez les patients présentant une dysplasie osseuse floride (DOF). Le traitement des KOS repose essentiellement sur la simple fenestration chirurgicale de la cavité osseuse, qui amène généralement une évolution favorable. Toutefois, dans le cas de vastes cavités, comme celles rencontrées dans les cas de DOF, on peut se demander quel peut être le résultat d'une telle intervention. Deux cas de KOS associés à une DOF sont rapportés chez deux femmes africaines (l'une Camerounaise, de 36 ans, l'autre Gabonaise de 72 ans). La grande taille des KOS semble inhabituelle. L'un d'eux (12 cm plus grand diamètre) pourrait même être le plus grand KOS publié en association avec une DOF. Malgré les fenestrations chirurgicales pratiquées, les KOS n'ont pas cessé leur expansion, bien qu'une quantité significative d'os néoformé soit nettement visible aux sites de fenestration sur les CT scans de contrôle à 1 an. Au vu de ces cas, le résultat du traitement chirurgical des grands KOS associés à une DOF apparaît peu concluant, puisque les cavités n'ont pas diminué de volume. Bien que, dans les cas rapportés, aucune des patientes n'ait souffert de complications à la suite de l'intervention, il paraît donc plus judicieux, en l'absence de manifestation symptomatique, de ne proposer qu'une simple surveillance à long terme.

## Introduction

Florid osseous dysplasia (FOD) is a relatively rare lesion, classified within bone related lesions (odontogenic tumours), as part of osseous dysplasias (OD) [1]. It is commoner in middle aged black women [2]. Radiological investigations of FOD reveal multiple radio-opaque masses, surrounded by radiolucent zones, suspended to teeth apices and located in two quadrants

or more. Simple bone cyst (SBC) is often found to be associated with FOD, especially in middle-aged black women [2].

Several surgical treatments of SBC have been proposed [3], such as curettage of the bone wall, fenestration, packing the cavity with material, aspiration, which frequently lead to resolution in the case of small cavities. However, in cases of very large cavities, the outcome of such treatment is in question.

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The results of two cases of SBC greater than 5 cm diameter in association with FOD following surgical intervention with a follow up of 10 and 12 months are presented.

### Case report 1

On presentation, a female patient of Gabonese origin, aged 72 years old, complained of pains in the lower right molar region. Her medical history was unremarkable and she was not on any medication, though revealed removal of a large maxillary cyst 10 years previously but with unconfirmed diagnosis on histopathology.

The clinical examination revealed tooth cavities, in both the first and second right lower molars.

Expansion of the mandibular lingual plate was noted in the anterior region (incisor-canine). Consequently, the mandible was enlarged in its bucco-lingual axis. There were no other signs to report, neither extraoral nor intraoral. Thermal testing confirmed the pulp vitality of all the other remaining mandibular teeth.

The orthopantomogram (OPG) revealed deformations of all lower teeth apices (Fig. 1). Secondly, mandibular osseous matrix showed heterogeneous radiolucent zones. These zones were of variable size and had clear borders. A CT scan was performed.

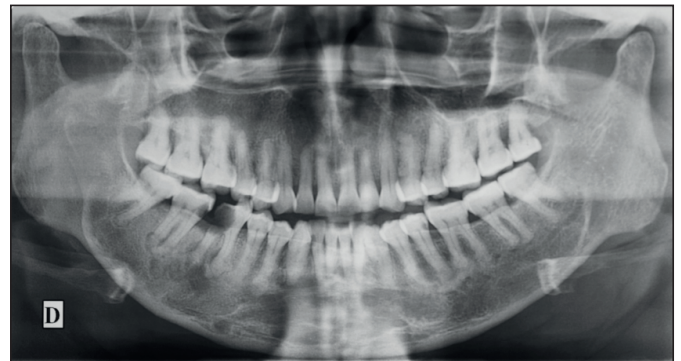
In response to the patient's dental pain, the root canal of the lower right second molar was treated, and the lower right first molar extracted. A calcified tissue was present around the distal root apex of the extracted tooth. On the macroscopic examination, this had the appearance of a cementoblastoma, which histopathology confirmed (Fig. 2).

The following week, the mandibular CT scan was performed and revealed the presence of an intra-osseous unilocular mandibular cavity. It exhibited irregular scalloped borders, was pseudo-septate and extended from the lower left first molar to the alveolus of the lower right first molar. It measured 3 cm high by 6 cm long, engulfing all the apices of the concerned teeth. The lingual cortical plate showed considerable expansion becoming particularly thin at the level of the crestal bone. The CT scan also showed mineralized masses at all the mandibular teeth apices, already visible on the OPG. The radiologist's report proposed in the differential diagnosis the possibility of an ameloblastoma.

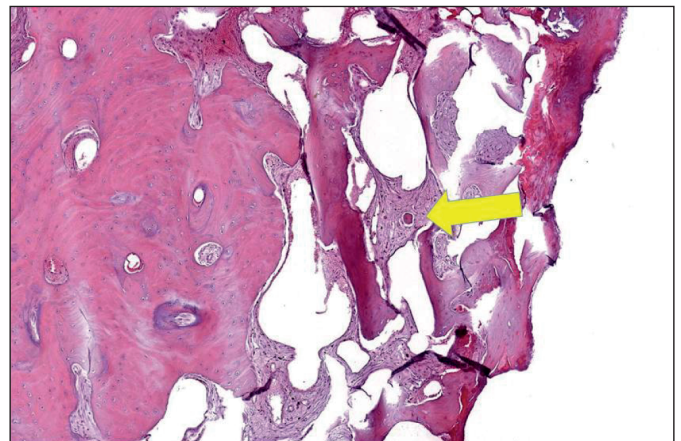
A biopsy seemed therefore indicated. On the lingual side of the lesion, the thin cortical bone was readily perforated and a biopsy of the lesion was taken from a unilocular cavity. The histology of the bone fragment was reported as atrophic, with no associated inflammatory reaction or tumourous lesion. The diagnosis of FOD associated with SBC was done.

Postoperative follow-up after 8 days showed no complications and the patient moved abroad.

The patient was seen again at 1 year. She mentioned a mild pain in the left maxillary region. Clinical examination



**Fig. 1.** Case report 1: OPG showing deformations of all lower teeth apices and aspect of abnormal mandibular osseous matrix (heterogeneous radiolucent zones); SBC cavity is not clearly apparent. *Fig. 1. Cas 1 : OPG montrant la déformation des apex de toutes les dents mandibulaires et une trame osseuse anormale (zones radioclaïres hétérogènes) ; le KOS n'est pas vraiment visible.*

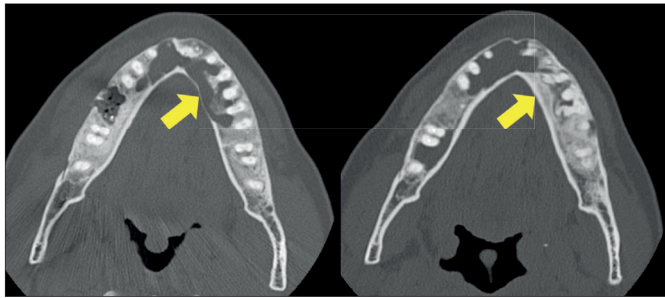


**Fig. 2.** Case report 1: thickened sclerotic bone with merging of bone trabeculae; presence of cementum spherules (arrow) (HE, x 40). *Fig. 2. Cas 1 : os sclérotique épaissi avec fusion des travées et présence de sphérules de ciment (flèche) (HE, x 40).*

was normal. A CT scan showed that the mandibular SBC cavity had been replaced by dense bone tissue at the site of the surgical access; but in contrast, the cavity had clearly developed in other locations (Fig. 3).

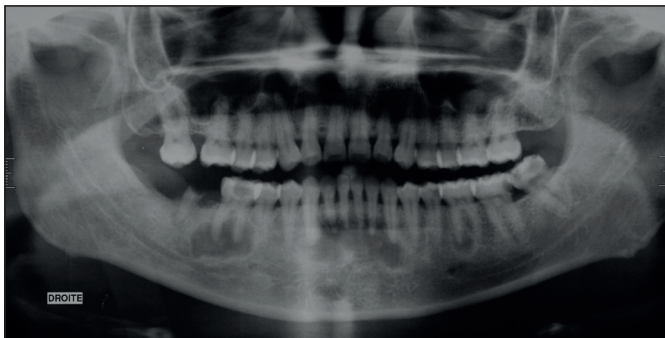
### Case report 2

A female patient of Cameroonian origin, aged 36 years old, was referred to the clinic for large heterogeneous radiolucent zones, incidentally discovered on an OPG. The patient complained of pain in the lower left molar region and of a sensation of gingival tightness. She was otherwise healthy and was not



**Fig. 3.** Case report 1: comparison between initial (left) and 12 months control (right) CT scan. The SBC cavity size has increased, and new bone has appeared at the surgical fenestration site (arrow). Note, on the initial CT, the recent avulsion site of the first right mandibular molar.

*Fig. 3. Cas 1 : comparaison entre le CT scan initial (à gauche) et le CT scan de contrôle à 12 mois (à droite). La taille de la cavité du KOS a augmenté, et de l'os néoformé est apparu au site de la fenestration chirurgicale (flèche). À noter, sur le CT scan initial, le site de l'avulsion récente de la première molaire mandibulaire droite.*



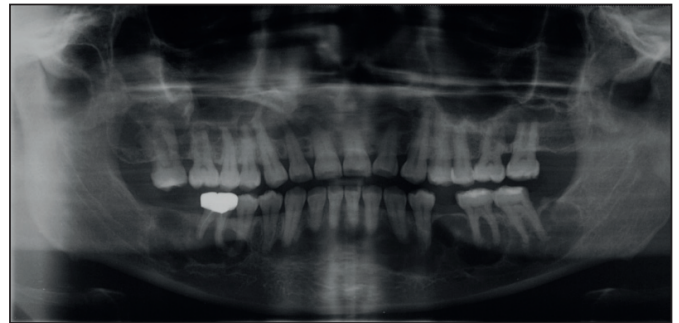
**Fig. 4.** Case report 2: OPG taken 8 years ago, showing several radiolucent zones in both maxilla and mandible.

*Fig. 4. Cas 2 : ancien orthopantomogramme (8 ans avant) montrant plusieurs zones radioclares apicales, aussi bien au maxillaire qu'à la mandibule.*

undergoing any medical treatment. Her medical history revealed the removal of mandibular "cysts" 8 years before. Histological results concluded compatibility with residual apical cysts with reactive ossification.

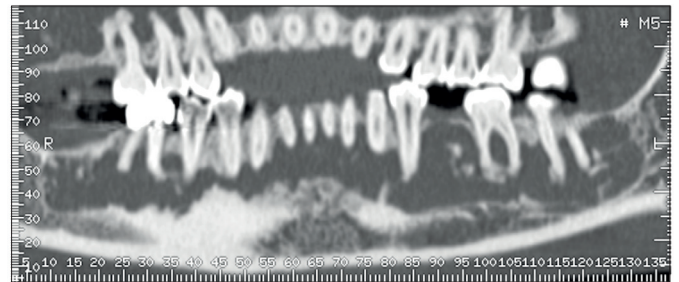
The clinical examination revealed expansion of both the maxillary and mandibular cortical bone (especially in the molar sectors). There was no pain or other symptoms to note. Thermal tests confirmed the pulp vitality of all the remaining teeth (excepting one restored tooth).

The initial radiographic report of the panoramic exam conducted 8 years before (Fig. 4) revealed radiolucent areas at the apices of both the mandibular first molars and incisors. The new OPG (Fig. 5) showed the same aspect of abnormal morphology, which seemed to extend to all upper and lower teeth



**Fig. 5.** Case report 2: recent OPG showing a right maxillary sinus lesion and larger multiloculated radiolucent zones in both maxilla and mandible.

*Fig. 5. Cas 2 : OPG récent montrant une lésion du sinus maxillaire droit et des zones radioclares au maxillaire et à la mandibule.*

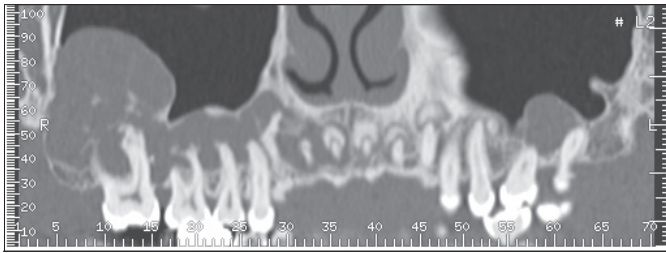


**Fig. 6.** Case report 2: CT scan (mandibular panoramic reconstruction) showing a unilocular intra-osseous cavity measuring 12 cm in the long axis and 2 cm in height.

*Fig. 6. Cas 2 : CT scan (reconstruction panoramique mandibulaire). Cavité uniloculaire intra-osseuse de 12 cm de long et 2 cm de hauteur.*

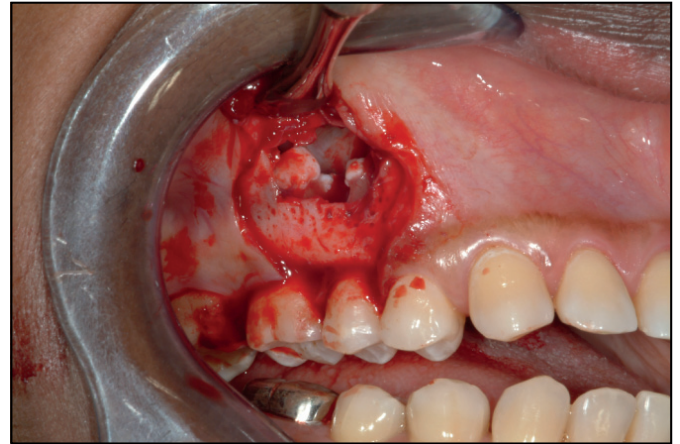
apices. It potentially resembled an ameloblastoma. Consequently, a CT scan was prescribed, which revealed the presence of intra-osseous unilocular maxillary and mandibular cavities. The mandibular unilocular cavity presented irregular scalloped borders and pseudo-septa, and extended beyond the right and left molars. It was 12 cm long, 2 cm high and 1.5 cm wide, engulfing all the mandibular apices (Fig. 6). Similar intra-osseous presentations were observed in the maxilla (Fig. 7), but did not cross the median suture. On the right side, the lesion extended into the maxillary sinus. The CT scan also revealed typical images of tooth apices surrounded by radio-opaque masses separated by a radiolucent zone, suggesting the diagnosis of FOD associated with a SBC.

Surgery exploration was performed to confirm the diagnosis and to possibly give a chance of healing of the intra-osseous cavities. Intra-osseous fluid was removed (Fig. 8) and analyzed. The thin buccal maxillary and mandibular cortical bone was therefore fenestrated, revealing unilocular pseudo-empty cavities and apices with mineralized masses (Fig. 9). The



**Fig. 7.** Case report 2: CT scan (maxillary panoramic reconstruction). Typical images of tooth apices surrounded by radio-opaque masses separated by a radiolucent zone.

*Fig. 7. Cas 2 : CT Scan (reconstruction panoramique maxillaire) ; Images typiques d'apex dentaires couronnés par des masses radio-opaques dont ils sont séparés par une zone radioclaire.*



**Fig. 9.** Case report 2: surgical access revealing pseudo-empty cavity with white masses at the tooth apices.

*Fig. 9. Cas 2 : accès chirurgical révélant, à l'intérieur de la cavité, des masses blanches aux apex dentaires.*



**Fig. 8.** Case report 2: aspiration of SBC fluid after bone cortical fenestration.

*Fig. 8. Cas 2 : aspiration du fluide du kyste osseux solitaire après fenestration de la corticale maxillaire.*

curette directly contacted the bone tissue. Osseous bone fragments were also removed for histological examination. Their analysis revealed no abnormality of the bone itself. The biochemical composition of the fluid indicated an alkaline phosphatase content of 713 UI/L.

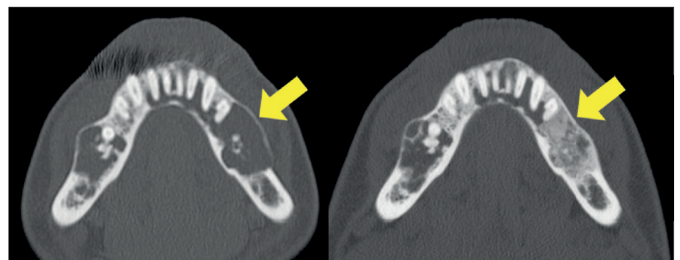
A final diagnosis of a FOD associated with a SBC was made.

The postoperative follow up 8 days later showed no complications.

The patient was seen 10 months later and remained symptom free. On the control CT scan performed, the SBC cavity did not seem to be larger than before. A significant amount of new bone appeared at the site of fenestration (Fig. 10).

## Discussion

The 2 cases reported here illustrate well the epidemiologic characteristics of FOD, known to preferentially appear among mature black females.



**Fig. 10.** Case report 2: comparison between initial (left) and 10 months control (right) CT scan. New bone has appeared at the surgical fenestration site (arrow).

*Fig. 10. Cas 2 : comparaison entre le CT scan initial (à gauche) et le CT scan de contrôle à 10 mois (à droite). De l'os néoformé est apparu au site de la fenestration chirurgicale (flèche).*

The SBC are unusual, since they measured respectively 6 cm and 12 cm at maximum dimension. Case 2 approximates the size of the SBC reported by MacDonald-Jankowski in a 47-year-old Chinese woman [4]. It could be the largest SBC ever reported in association with a FOD. Indeed, SBC rarely exceeds 5 cm in size. In a review of 44 clinical cases of SBC, of which, one was associated with "hypercementosis", and another one to a peri-apical OD, Copete et al. [5] reported only 6 cases where the lesion extended further than 5 cm, with the maximum lesion measuring 9 cm × 3.5 cm.

In these cases, the diagnostic was difficult because the size of the lesion was unusual for SBC and the content appeared heterogeneous on CT pictures. This could have also suggested an ameloblastoma, as in the case presented by Mitchell et al. [6], or a fibro-osseous lesion, as in the case of Harris et al. [7] This aspect essentially arises from pseudo-septa.

With regards to these given patients the particularities of the SBC seem mainly due to the existence of the OD. According to Melrose *et al.* [2], the SBC-OD association constructs a nosological entity, different to an isolated SBC. These authors had indeed observed a higher concentration of alkaline phosphatase in cystic fluid, when associated to OD, than in the isolated SBC (whereas the acidic phosphatase was similar). In the second case reported here, a high concentration of alkaline phosphatase in cystic fluid (713 UI/L) was found. Moreover, whereas the vast majority of SBC cases affect younger subjects [8], Kaugars *et al.* [9] suggested that the association SBC-OD is mainly found in black women, over the age of 30. These present cases (African women aged 72 and 36) seem to confirm this hypothesis.

The apparent vacuity discovered in this first reported case probably results from instantaneous aspiration of the serous fluid during the surgical exploration. The CT scan had effectively showed that the density of the cystic cavity may correspond to that of liquid content (–10 to +100 Hounsfield units), much higher than air density (–1000 Hounsfield units). The extreme fluidity of the contents of such cavities can explain its sudden disappearance because of the surgical suction. In the second reported case, the aspiration of pseudo-cystic liquid before the perforation of cortical bone ensured the detection of liquid. In an investigation carried out on 7 SBC cases, Eriksson *et al.* [10] found 4 empty cavities during surgical opening. This contradicted the MRI imaging, which suggested all the cysts contained an aqueous fluid. Suei *et al.* [3] researched gas presence in cystic cavities via a CT scanning study of 52 cases of SBC. Their findings suggest that the discovery of a void cyst during surgery is, in fact, an observational error, as liquid is always present in such cavities. In any case, the observation of an empty cavity after cortical perforation is potentially an essential diagnostic element for the clinical diagnosis of SBC [7].

In both cases, medical examination revealed a history of surgery for jaw cyst a few years earlier. It is likely that these lesions were already SBC that were not completely diagnosed as part of a SBC-OD association, which was finally obvious some years later.

The SBC in the cases reported here presented many of the identified factors that are linked to recurrence, such as the presence of scalloped contours, multiple cavities [3] and the association with the OD [2]. Furthermore, the frequency of a complete recovery of a SBC seems to decrease with increasing cyst volume [11]. And indeed, the outcome of the surgical treatment of the reported SBC appeared unconvincing, though neither patient appeared to suffer any detrimental effects of surgical intervention. The SBC cavities did not stop increasing, though it is not possible to tell whether the

surgical fenestration had an effect on their evolution. Furthermore, at all the sites where the thin cortical bone had been removed, a significant amount of newly formed bone tissue could be noted after a few months. This response suggests it might be advised to perform multiple fenestrations into the lesion to induce a better healing. Though neither patient appeared to suffer any detrimental effects of surgical intervention, the benefit of such intervention remains in question and the outcome in these two case studies suggests that in the absence of symptoms long term follow up is all that is necessary.

**Competing interests:** none

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## References

- Vazquez L, Gheddaf Dam H, Lombardi T, Bernard JP, Samson J. Classification des dysplasies cémento-osseuses: illustration par quelques cas. *Med Buccale Chir Buccale* 2007;13:118-9.
- Melrose RJ, Abrams AM, Mills BG. Florid osseous dysplasia. A clinical-pathologic study of thirty-four cases. *Oral Surg Oral Med Oral Pathol* 1976;41:62-82.
- Suei Y, Taguchi A, Nagasaki T, Tanimoto K. Radiographic findings and prognosis of simple bone cysts of the jaws. *Dentomaxillofac Radiol* 2010;39:65-71.
- MacDonald-Jankowski DS. Traumatic bone cysts in the jaws of a Hong Kong Chinese population. *Clin Radiol* 1995;50:787-91.
- Copete MA, Kawamata A, Langlais RP. Solitary bone cyst of the jaws: radiographic review of 44 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1998;85:221-5.
- Mitchell DA, Ward-Booth RP. Atypical presentation of a solitary bone cyst. *Int J Oral Surg* 1984;13:256-9.
- Harris SJ, MK OC, Gordy FM. Idiopathic bone cavity (traumatic bone cyst) with the radiographic appearance of a fibro-osseous lesion. *Oral Surg Oral Med Oral Pathol* 1992;74:118-23.
- Matsumura S, Murakami S, Kakimoto N, Furukawa S, Kishino M, Ishida T, *et al.* Histopathologic and radiographic findings of the simple bone cyst. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1998;85:619-25.
- Kaugars GE, Cale AE. Traumatic bone cyst. *Oral Surg Oral Med Oral Pathol* 1987;63:318-24.
- Eriksson L, Hansson LG, Akesson L, Stahlberg F. Simple bone cyst: a discrepancy between magnetic resonance imaging and surgical observations. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2001;92:694-8.
- Glowacki M, Ignys-O'Byrne A, Ignys I, Mankowski P, Melzer P. Evaluation of volume and solitary bone cyst remodeling using conventional radiological examination. *Skeletal Radiol* 2010; 39:251-9.