

성인 늑골에 생긴 단순성 골낭종 ^{18}F -FDG섭취의 조직학적 근거: 증례보고 및 여러 영상검사 소견과의 비교검토

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Histologic Base of Mild ^{18}F -FDG Uptake in Simple Bone Cyst of Adult Rib: A Case Report with Multi-image Correlation

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Simple bone cyst (SBC) is very rare in adult ribs. The diagnosis basically relies on conventional radiography and occasionally on CT. There has been no earlier publication on PET/CT diagnosis of SBC. We report a case of adult costal SBC diagnosed by positive ^{18}F -fluorodeoxyglucose (FDG) uptake. Histology showed the FDG uptake to be associated with reactive woven bone formation and nonspecific chronic inflammation. Correlation of PET, CT, plain radiography and sonography are also described. (Nucl Med Mol Imaging 2008;42(4):328-332)

Key Words: simple bone cyst, rib, ^{18}F -F C, PET/CT

Introduction

Simple bone cyst (SBC) is a benign tumor of the bone marrow. The absolute majority of SBC involve the humerus and femur and in the second and third decades of life, SBC is very rare in the ribs and rarer still in adult ribs.¹⁻⁴⁾ The early diagnosis of costal SBC is important because of its mimicry of serious conditions such as malignant metastasis, myeloma, tuberculosis and others.⁵⁾ There has been no earlier report on SBC in adult ribs detected by ^{18}F -fluorodeoxyglucose (^{18}F -FDG) positron emission tomography/computed tomography (PET/CT).

We report a case of adult rib SBC diagnosed by mild

^{18}F -FDG uptake. Microscopic study disclosed chronic, reactive, pericyclic tissue inflammation and woven-bone formation which were considered to have resulted from slow expansion of the cyst, accounting for FDG uptake in our case. In addition, the results of cross correlation of the findings of PET, CT, conventional radiography (CR) and ultrasound (US) are described.

Case report

A 32-year-old female patient had pain of one-month duration in the posterior arc of the right lowermost rib. Patient was otherwise healthy without known history of neoplastic disease or trauma. Vital signs and routine laboratory tests were all within normal limits.

Supine bone-technique CR of the chest showed a 10 × 6 mm ovoid lytic lesion in the medulla of the posterior arc of the right 11th rib (Fig. 1A). In this patient the ribs were 11 pairs. On magnified CR, the long axis of the lesion was transversely aligned and inferiorly eccentric in location (Fig. 1B). The superior border was

- **Received:** 2008. 8. 17. • **Accepted:** 2008. 8. 25.
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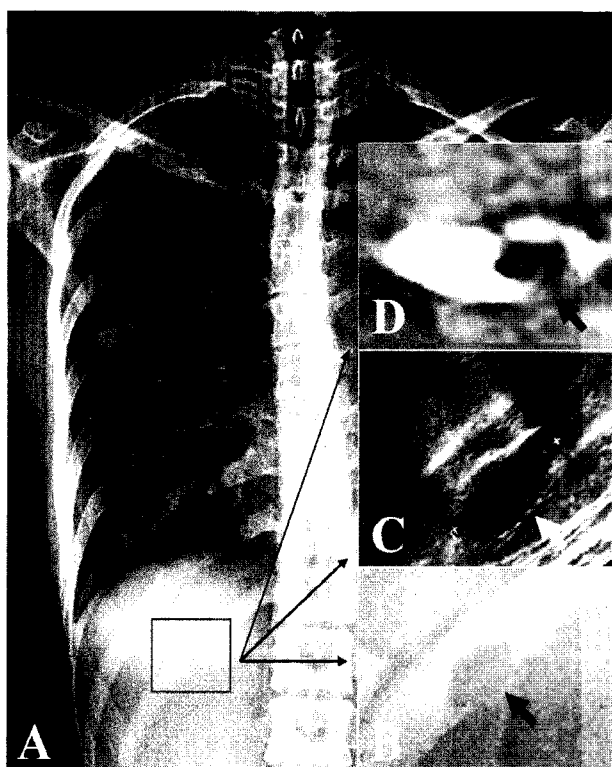


Figure 1. (A) Bone technique supine radiograph of the chest shows a small transversely coaxial lytic lesion in the middle portion of the right 11th rib arc (framed). (B) Magnified radiograph reveals inferiorly eccentric lysis with sclerotic superior border and ballooned inferior border. Thin lower cortex is barely discernible (arrow). (C) Sonograph shows lesional content to be hypoechoic with thick superior and thin inferior hyperechoic border. Note marked thinning of the inferior cortex (arrow). (D) CT shows low density lesional content with thickened superior border and thinned inferior border (arrow).

sclerotic and the inferior border ballooned with a paper thin cortex. US showed the superior and inferior borders of the lesion to be hyperechoic and content hypoechoic (Fig. 1C). On CT the lesion was radiolucent and expansive with sclerotic superior border and ballooned lower border (Fig. 1D and 2C). Differential list included eosinophilic granuloma, malignant metastasis and SBC.

For systematic survey, torso PET/CT was performed. Images were obtained 1 h and 2 h after intravenous injection of 13.8 mCi of ^{18}F -FDG using a scanner (Siemens Biograph Duo). Maximum intensity projection showed a spotty area of mild FDG uptake in the right lowermost rib (Fig. 2A). The coronal section (Fig. 2B) revealed the area to correspond to radiographic osteolysis (Fig. 1A). The maximum standardized uptake values (SUVmax) were calculated as 2.8 on the 1-h scan and 2.7 on the 2-h scan, respectively. CT performed as a

part of PET/CT revealed a small ovoid lytic lesion in the arc of the right 11th rib (Fig. 2C). Based on those imaging findings and pain the diagnosis was narrowed to SBC with possible trauma.

Patient was admitted for excision biopsy and curative resection. The tumor-bearing rib segment was removed en bloc. On cutting clear fluid flowed out and void cavity was seen to be lined with a thin, white, fibrous capsule (Fig. 3A inset). Microscopic scrutiny of cyst wall on slab section (Fig. 3A) failed to find evidence of fracture such as callus formation. Instead, the capsule was in part mantled by a layer of reactive woven bone and in part by connective tissue and thereto attached reactive bone (Fig. 3B). Of interest, the connective tissue layer was thick and infiltrated with nonspecific chronic inflammatory cells (Fig. 3C).

Discussion

According to an electron microscopic study SBC derives from the congenital rest of synovial tissue migrated into long bone metaphyses.⁶⁾ Histologically, the cyst is encapsulated just like an egg with a thin fibrous membrane that is white and shiny. It is a disease of the second and third decades of life with male predominance. SBC accounts for 3% of all biopsy-proven primary tumors of bone and over 95% of which occur in the long bones, the proximal metaphyses of the humeri and femora in particular.¹⁾ In older adults, flat or irregular bones are more commonly affected than the long bones⁷⁾ and a small fraction of SBC involve the spine, ribs, clavicle, scapula, ischium, pubis and patella.⁸⁾

Unlike aneurysmal bone cyst (ABC), SBC is very rare in the ribs. Indeed, Baker²⁾ in an analysis of 45 cases of unicameral bone cyst could not find single case in the ribs and Gayler and Donner³⁾ also could not find SBC in the ribs among 159 benign rib tumors collected from the English-German literatures. The latter authors observed that fibrous dysplasia, chondroma and osteochondroma were the three most common benign tumors and tumorous condition of the ribs. The Netherlands Committee on Bone Tumours⁴⁾ filed one case of SBC in 52 cases of primary rib tumors. Thus,

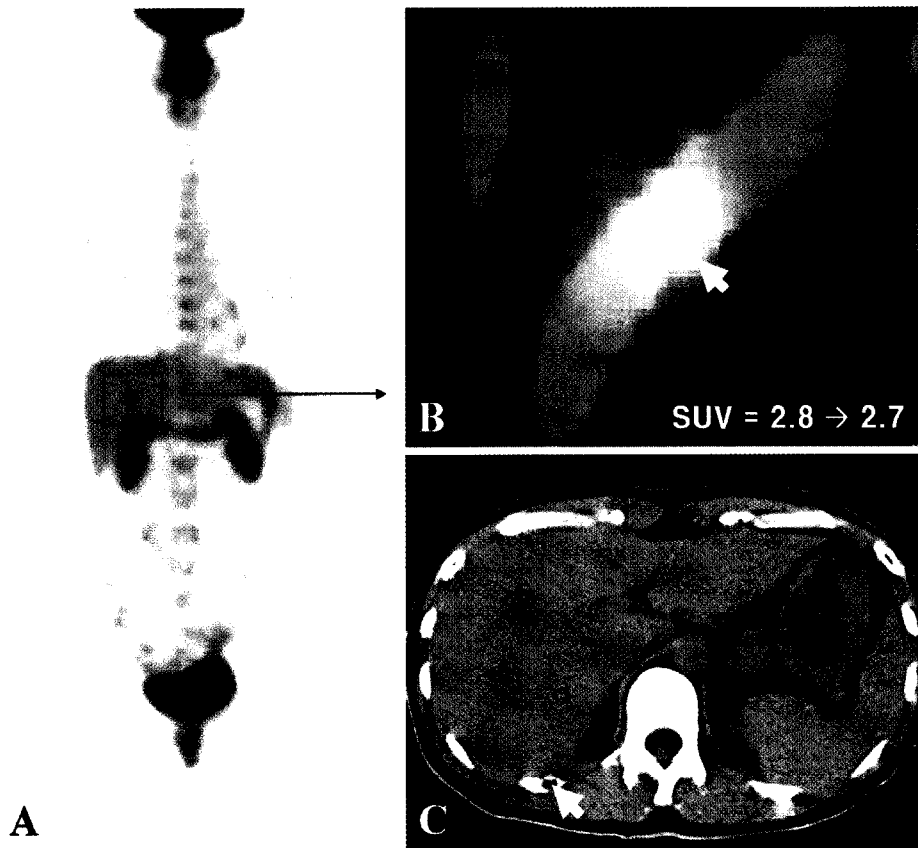


Figure 2. (A) Maximum intensity projection PET/CT of the torso shows a spotty area of mild FDG uptake in the right lowermost rib (framed). (B) Magnified coronal PET/CT scan shows the lesion with increased FDG uptake (arrow). (C) Transverse CT section shows a small lytic lesion (arrow).

SBC is very rare in the ribs and rarer still in the adult ribs.

Clinically, the accurate diagnosis of SBC in the ribs is essential especially when painful as it simulates metastasis, myeloma, giant cell tumor, eosinophilic granuloma, tuberculosis, mycosis and others.⁵ The diagnosis of SBC and other tumors of the ribs basically relies on CR and occasionally on CT and MRI⁹⁾ and US¹⁰⁾ and limitedly on PET/CT. The SBC herein reported is unique in that it occurred in the rib of a female in her fourth decade of life and was diagnosed by positive ^{18}F -FDG uptake on PET/CT. SUV was calculated as only 2.8 but enough to draw attention.

Uncomplicated SBCs in the small bones such as the ribs and phalanges insignificantly accumulate radiotracer, for example ^{18}F -FDG in PET and $^{99\text{m}}\text{Tc}$ -HDP in bone scan, and, hence, hard to detect by scintigraphy. It is because that serous fluid of SBC is metabolically inert and the fibrous capsule is thin, usually less than 1 mm,

and inert either. Metabolic activity of the capsules of even large SBCs of the femur, humerus and pelvis are subtle so that it can be detected only by pinhole scan.¹¹⁾ The rib cyst of ours was small in size (6 × 10 mm) but recognizably accumulated ^{18}F -FDG so that it could be picked up by PET/CT.

Such an unusual manifestation led us to investigate the mechanism with which FDG uptake in small SBC can be explained. At first, we suspected trauma as reasonable cause since the cyst was painful. Against expectation, however, the slab section of removed cyst showed the fibrous capsule to be preserved without evidence of fracture such as new bone or callus formation. Instead, high power scrutiny showed the capsule in part mantled with reactive woven bone and in part with fibrous tissue infiltrated with nonspecific chronic inflammatory cells and reactive woven bone thereto attached. From the findings it was inferred that active FDG metabolism of reactive woven bone and

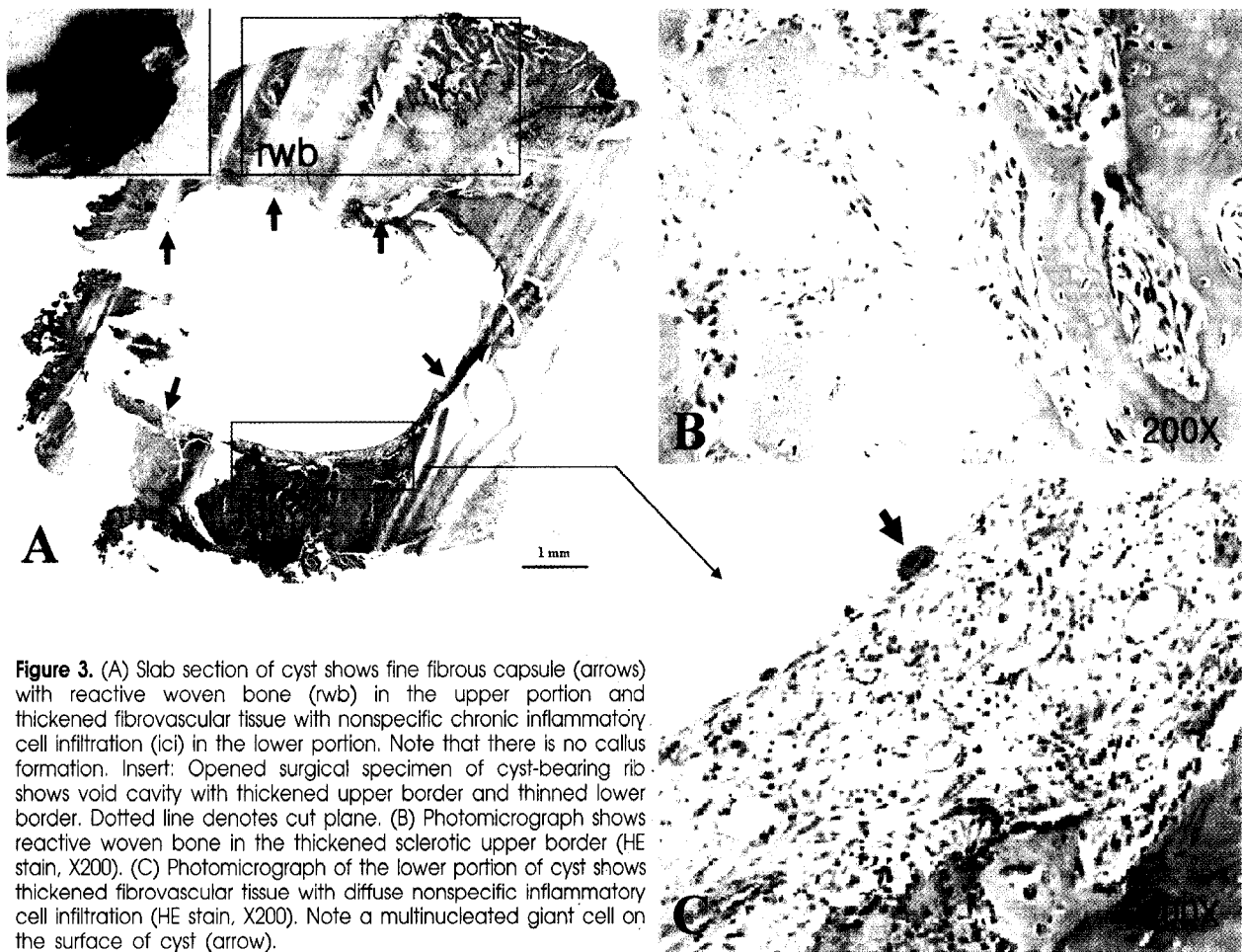


Figure 3. (A) Slab section of cyst shows fine fibrous capsule (arrows) with reactive woven bone (rwb) in the upper portion and thickened fibrovascular tissue with nonspecific chronic inflammatory cell infiltration (ici) in the lower portion. Note that there is no callus formation. Insert: Opened surgical specimen of cyst-bearing rib shows void cavity with thickened upper border and thinned lower border. Dotted line denotes cut plane. (B) Photomicrograph shows reactive woven bone in the thickened sclerotic upper border (HE stain, X200). (C) Photomicrograph of the lower portion of cyst shows thickened fibrovascular tissue with diffuse nonspecific inflammatory cell infiltration (HE stain, X200). Note a multinucleated giant cell on the surface of cyst (arrow).

chronic inflammatory cells are responsible for positive FDG accumulation in this SBC. It was also inferred that woven bone formation and nonspecific inflammation had resulted from physical stimulation of slowly expanding cyst.

Cross correlation of the image findings of the cyst revealed by CR, CT and US showed no evidence of fracture. CR demonstrated the cyst to be ballooned with thin cortex inferiorly and bordered by thickened bone superiorly and US showed cystic fluid to be hypoechoic and surrounded by hyperechoic wall. CT finding was much the same as that of CR. The integrity of the cyst wall was advantageously appreciated on US and CT scan. It is worthy of mention that FDG uptake on the color coded coronal PET scan was so distinctive.

It is concluded that FDG avidity of metabolically active reactive woven bone and nonspecific chronic inflammation resulted from slow expansion of cyst were

responsible for positive FDG uptake observed in our adult rib SBC.

Acknowledgement

Authors are grateful to Professor Jin Kim, M.D., Ph.D., Department of Anatomy, for producing slab photograph.

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