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A Nodular Type of Subcutaneous Sarcoidosis: A Case Report[1]

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Sarcoidosis is a granulomatous multisystemic disorder that rarely involves subcutaneous tissue. We describe the MR imaging findings of a subcutaneous sarcoidosis in a patient that presented with a nontender, palpable soft tissue mass on the left buttock, which was confirmed after surgical excision. The MR images showed the presence of a subcutaneous mass that breached the adjacent fascia with an irregular outline and homogeneous, slightly higher signal intensity than the surrounding muscle as seen on a T2-weighted image and with homogeneous enhancement after contrast injection. The lesion could not be differentiated from a sarcoma or a malignancy.

Index words : Sarcoidosis
Subcutaneous tissue
Buttocks
Magnetic resonance (MR)

Sarcoidosis is a chronic, multisystem granulomatous disease of unknown cause that can involve various organs. A diagnosis is usually made when clinical and radiological manifestations are supported by histological evidence of the presence of noncaseating granulomas in one or more tissues (1, 2). Sarcoidosis is well characterized by the presence of hilar lymphadenopathy and pulmonary infiltrates. Extrathoracic organs are also involved, such as the skin, eye, heart, liver, spleen, scrotum, central nervous system, joints, muscles and bone (1-5). Rarely, subcutaneous tissue (2, 3, 5-9) or the fascia (8) is involved.

We experienced a case of subcutaneous sarcoidosis that was manifested as a palpable mass that breached the adjacent fascia. The mass was seen with homogeneous and slightly higher signal than the muscle on a T2-weighted image, and the lesion mimicked a soft tissue tumor. We report here the MR imaging findings of a case of a nodular type of subcutaneous sarcoidosis that could not be differentiated from a sarcoma or a malignancy.

Case Report

A 51-year-old female without a remarkable history of disease was referred to our hospital for evaluation of a palpable soft tissue mass on the left buttock that has appeared during the past three months. On a physical examination, the mass measured approximately 5.1 cm in the longest diameter as measured with a ruler and there was no significant tenderness with the intact overlying

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skin. The erythrocyte sedimentation rate (normal 10–20 mm/hr) and level of C-reactive protein (normal, 0–0.03 mg/L) were slightly elevated at 30 mm/hr and 0.09 mg/L, respectively.

As seen on a T1-weighted spin echo MR image (Fig. 1A), the lesion, which was located in the subcutaneous layer of the left buttock, was isointense relative to muscle.

As seen on T2-weighted fast spin echo (Fig. 1B) and proton fast spin echo (Fig. 1C) images, the mass showed homogeneous signal intensity, and was slightly hyperintense than the surrounding muscle. As seen on a post-contrast T1-weighted image (Fig. 1D), the mass showed homogeneous enhancement and breached the adjacent fascia. On the next day after MR imaging was performed, the patient was admitted for further evaluation. Other soft tissue masses (4 cm in the longest diameter, respectively) on the right elbow and left knee were detected on a physical examination. Radiological evaluation of the other soft tissue masses was not performed. A chest radiograph performed two days before surgery was unremarkable. The patient underwent marginal margin excision of the subcutaneous mass on the buttock, which involved the adjacent fascia on the operative field. The histological specimen (Fig. 2) showed the presence of diffuse solid noncaseating granulomas that represented sarcoidosis. The level of serum angiotensin converting enzyme (ACE) was elevated (75.2 IU/mL) and the serum calcium level was within normal limits (2.3 mmol/L). The patient refused steroid therapy. Six months after surgery, the soft tissue masses disappeared and there was slight further progression of the pulmonary nodules as seen on a follow-up chest radiograph. We have not seen the patient for one year.

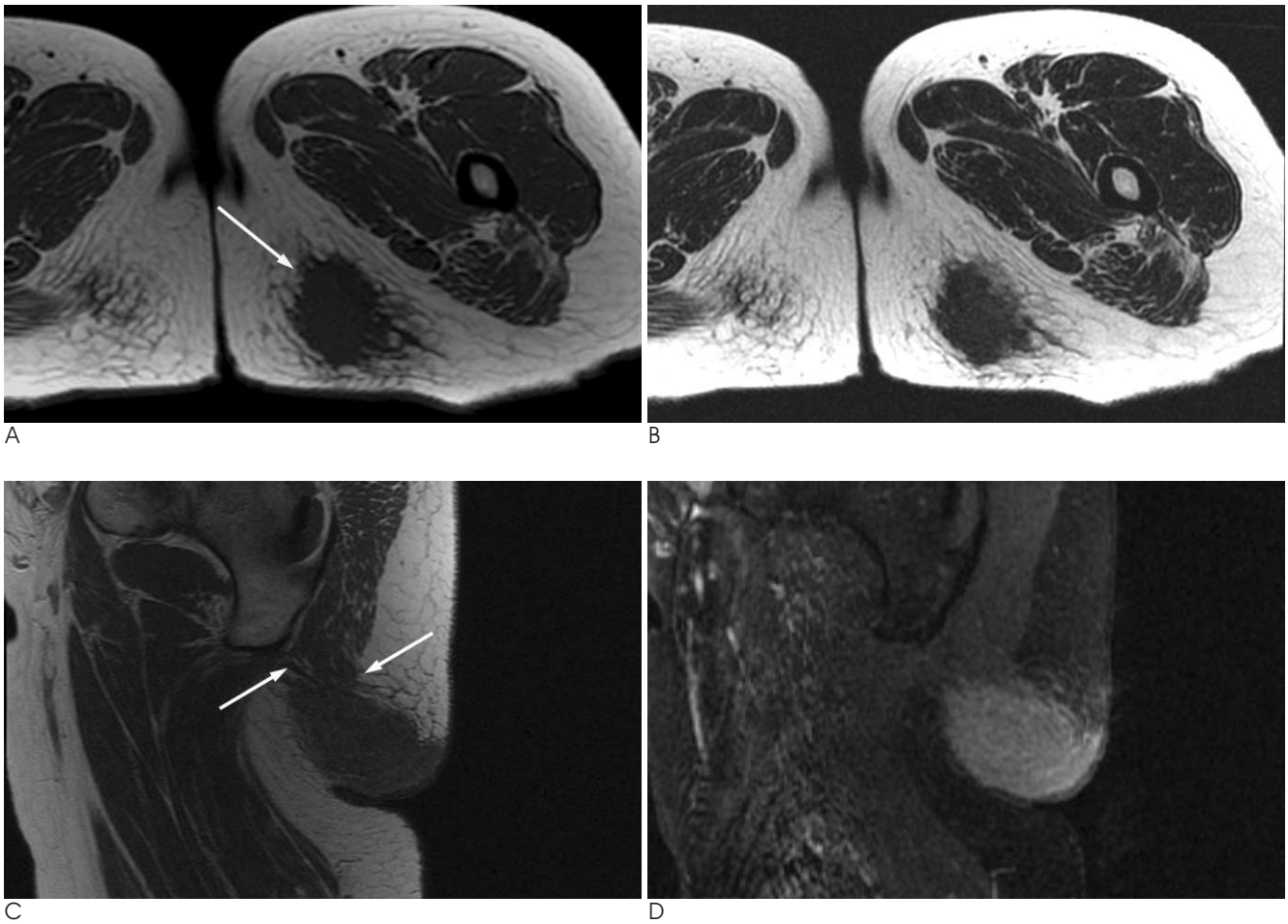

Fig. 1. A. An axial T1-weighted spin echo MR image (TR/TE: 650/9) shows a subcutaneous mass (arrow) with a spiculated border. The mass appears isointense relative to the adjacent muscle.
B, C. As seen on axial T2-weighted fast spin echo MR (TR/TE: 4000/99) and sagittal proton fast spin echo MR (TR/TE: 4000/26) images, the mass is slightly hyperintense than muscle and it abuts the adjacent fascia (arrows).
D. A sagittal post-contrast T1-weighted MR image (TR/TE: 550/14) shows homogeneous enhancement of the lesion.

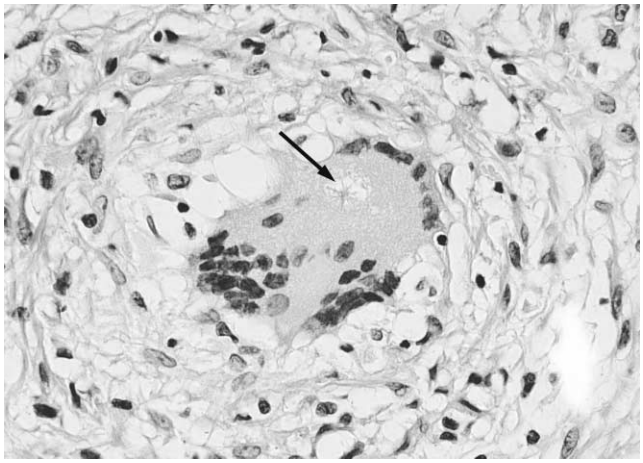

Fig. 2. A photomicrograph shows multinucleated giant cells including a typical asteroid body (arrow) with surrounding epithelioid cells and lymphocytes (hematoxylin and eosin staining, magnification × 200).

Discussion

Sarcoidosis is a relatively rare disease in Asia area that usually involves the mediastinal lymph nodes and the lungs. Subcutaneous tissue involvement is sometimes encountered although it has been reported to be rare. Moore et al. reported two cases of subcutaneous involvement among 42 sarcoidosis patients with musculoskeletal symptoms (5). In 2005, Marcoval et al. also reported that subcutaneous sarcoidosis was demonstrated in ten (2.1%) of 480 patients with systemic sarcoidosis (2). In accordance with these results, subcutaneous sarcoidosis has been reported in 6% of patients with systemic sarcoidois in one series (6) and there have been fewer than 30 documented cases in the clinical literature until 1993 (7). Although it was originally thought to occur late in the course of the disease (6), a more recent review suggests that a subcutaneous lesion usually appears at the onset of the disease and it is the presenting feature in over 40% of reported cases (2, 7). According to Marcoval et al., the presenting features of subcutaneous sarcoidosis were only the presence of a subcutaneous mass or a subcutaneous mass that was accompanied by constitutional symptoms. A mass has appeared with erythema nodosum in some cases (2). Subcutaneous sarcoidosis may be under-reported and may go unrecognized, as the disorder may be asymptomatic in cases where the only clinical feature is the presence of nontender, multiple soft tissue masses.

There have been some reported cases of subcutaneous lesions with imaging findings (8, 9). Shinozaki et al. categorized subcutaneous involvement of sarcoidosis in four cases into two patterns, i.e., nodular and diffuse types. As seen on MR images, nodular lesions associated with subcutaneous sarcoidosis showed homogeneous signal intensity similar to that of muscle on T1-weighted images, and a heterogeneous signal of higher intensity than muscle was seen on T2-weighted images. Diffuse lesions showed a stripe or mesh pattern with intermediate signal intensity as seen on both T1- and T2-weighted images. After administration of contrast material, the lesion was seen as slightly enhanced (9). In our case, the nodular subcutaneous lesion of the buttock showed similar signal intensity as seen in previously described cases (9) on a T1-weighted image and homogeneous enhancement after contrast injection, but it showed homogeneous signal intensity on T2-weighted fast spin echo and proton fast spin echo images. To the best of our knowledge, the nodular type of subcutaneous sarcoidosis that showed homogeneous and slightly higher signal than muscle on a T2-weighted image has not been previously reported.

The nodular type of the muscular sarcoidosis has a characteristic appearance as seen on MR imaging. The sarcoid nodules typically show star-shaped central areas of fibrosis that are seen with low signal intensity in all sequences and peripheral areas of granulomas that exhibit high signal intensity as seen on a T2-weighted image and intense enhancement as seen on contrast images. The central portion of the nodule is found in the chronic stage of muscular sarcoidosis (1, 5). To the best of our knowledge, a subcutaneous sarcoidosis has not been seen with a star-shaped central structure with decreased signal intensity as with muscular sarcoidosis. This finding may indicate that subcutaneous sarcoidosis is not associated with chronic fibrotic disease.

Due to the presence of the spiculated margin of the lesion and as the lesion breached the fascia, we considered the possibility of the presence of a malignant subcutaneous tumor such as malignant fibrous histiocytoma or sarcoma, and fibromatosis in the differential diagnosis. According to Galant et al., most subcutaneous masses that contact with an obtuse angle or cross the fascia are malignant, such as a malignant fibrous histiocytoma or a soft tissue sarcoma (10). These lesions show intermediate-to-low signal intensities as seen on T1-weighted images and intermediate-to-low or high signal intensities as seen on T2-weighted MR images with variable enhancement as seen on post-contrast T1-weighted images that are associated with the histological compo-

nents, which could not be differentiated in the present case. Fujimoto et al. have reported a case of subcutaneous sarcodiosis in the left buttock that breached the fascia (8). These investigators also strongly considered that the mass was an aggressive neoplasm such as a malignant fibrous histiocytoma. Whether the lesion originated from the gluteus maximus muscle, the fascia or the subcutaneous tissue was uncertain. In our case, the lesion definitely originated from the subcutaneous layer and involved the adjacent fascia, which was confirmed after surgery.

Although MR imaging findings of subcutaneous sarcoidosis are variable, the familiarity with subcutaneous sarcoidosis will be helpful to establish the diagnosis and it should be differentiated from malignant soft tissue tumors as it can breach the fascia.

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사르코이드증은 다발성으로 전신을 침범하는 육아종성 질환으로서 피하지방층에 드물게 발병한다. 좌측 둔부에 발생한 무통의 촉지성 연부 종괴를 주소로 수술을 시행하여 피하지방층 사르코이드증으로 확진 된 환자에서 사르코이드증의 자기공명영상소견을 보고한다. 자기공명영상에서 불규칙한 윤곽을 가지고 T2 강조영상에서 균질하며 근육보다 약간 고신호강도를 보이고 균질하게 조영증강되는 피하지방층 종괴는 주변 근막을 침범하는 양상을 보였으며 육종 및 악성종양과의 감별이 어려웠다.