Solitary synovial chondromatosis arising in the gluteus maximus bursa: computed tomography and magnetic resonance imaging findings

Kaoru Sumida1,2, Noriko Kobayashi1, Atsushi Nambu1, Masao Tago1, Isao Shibuya3 and Masashi Kawamoto4

Abstract
Chondral tumors in soft tissue are referred to as soft-tissue chondromas or extraskeletal chondromas, or as synovial chondromatosis if they arise in synovial tissue. We report the case of a 29-year-old man with synovial chondromatosis, also called synovial osteochondromatosis, which appeared in a solitary and extra-articular form. On magnetic resonance imaging (MRI) and computed tomography, the central portion of the tumor showed similar characteristics to bone marrow, despite the absence of any connection to adjacent bone. T2-weighted imaging displayed marked peripheral hyperintensity consistent with a cartilaginous area. These findings suggested the presence of enchondral ossification and were similar to those of skeletal osteochondroma, with the exception of the absence of attachment to bone. MRI is useful for distinguishing solitary synovial chondromatosis from other lesions, such as myositis ossificans, extraskeletal chondrosarcoma, and parosteal osteosarcoma.

Keywords
Synovial chondromatosis, extraskeletal osteochondroma, gluteus maximus bursa, magnetic resonance imaging (MRI)

Date received: 19 June 2015; accepted: 22 October 2015

Introduction
Synovial chondromatosis is a well-recognized benign osteochondral neoplasm that usually appears as multiple ossified or cartilaginous nodules in the synovial joints. However, solitary synovial chondromatosis outside the joints is rare (1). This pathology arising at an unusual site can potentially be mistaken for other diseases, such as myositis ossificans, extraskeletal chondrosarcoma, or parosteal osteosarcoma. Misdiagnosis may lead clinicians to an erroneous approach to synovial chondromatosis. Furthermore, biopsy of chondral masses is often inconclusive without the aid of appropriate radiological assessment. Imaging thus plays a vital role in the diagnosis of this disease. Herein, we report a case of synovial chondromatosis forming a solitary nodule that was considered to have arisen in the gluteus maximus bursa, with emphasis on the imaging findings.

Case report
A 29-year-old man presented with pain in the right buttock. He had noticed a mass in the same area 1 year earlier and began to feel pain after falling on his buttock 1 week before presentation. Physical examination revealed a firm mass on the dorsal aspect of the right

1Department of Radiology, Teikyo University Mizonokuchi Hospital, Kanagawa, Japan
2Department of Radiology, National Center Hospital of Neurology and Psychiatry, Tokyo, Japan
3Department of Orthopedics, Teikyo University Mizonokuchi Hospital, Kanagawa, Japan
4Department of Diagnostic Pathology, Teikyo University Mizonokuchi Hospital, Kanagawa, Japan

Corresponding author:
Kaoru Sumida, Department of Radiology, National Center Hospital of Neurology and Psychiatry, 4-1-1 Ogawa-higashimachi Kodaira, Tokyo 187-8551, Japan.
Email: sumida.cold@gmail.com
greater trochanter. Results of routine blood examination were unremarkable.

Plain radiographs with anterior–posterior and lateral views of the right hip showed a well-circumscribed, calcified mass that was slightly more radiolucent than the adjacent bone and showed no connection to the proximal femur (Fig. 1a, b). Computed tomography (CT) confirmed these characteristics (Fig. 2). On magnetic resonance imaging (MRI), a lobulated mass was identified anterior to the gluteus maximus muscle near the right greater trochanter. The mass showed high signal intensity similar to that of the adjacent bone marrow centrally and low signal intensity peripherally on T1-weighted (T1W) imaging (Fig. 3a), and heterogeneous moderate signal intensity again similar to the bone marrow surrounded by a layer of markedly high signal intensity on T2-weighted (T2W) imaging (Fig. 3b). The central signal was decreased on fatsaturated T2W imaging, suggesting the presence of fat within the lesion (Fig. 3c).

This lesion was followed up for 10 months, showing a slight increase in size. Subsequent needle biopsy could not rule out a malignant process such as chondrosarcoma, so total excision of the mass was performed. Intraoperative findings confirmed that the mass was of extraskeletal origin, abutting the gluteus maximus muscle. Synovial tissue encapsulated the mass, which was therefore presumed to arise in the gluteus maximus bursa. Histological examination revealed hyaline cartilage at the peripheral portion of the mass, and intracartilaginous ossification was depicted in the center. Gradual transition from fibrous cartilage to mature trabecular bone was observed (Fig. 4). The whole lesion was surrounded by synovial tissue. No evidence of malignancy was seen, such as cell atypia, necrosis, or mitotic figures. These findings were compatible with synovial chondromatosis and the final diagnosis was solitary synovial chondromatosis arising in the gluteus maximus bursa.

Discussion

The term “soft-tissue chondroma” (extraskeletal chondroma) was first introduced by Chung and Enzinger to describe a solitary cartilaginous nodule unattached to bone (2). Soft-tissue chondroma may consist of not only cartilaginous tissue, but also bone tissue due to enchondral ossification. As a result, the pathology may also be called soft-tissue or extraskeletal “osteochondroma” (3–6). When covered by synovial lining, the term “synovial osteochondroma” may be used (7). Edeiken et al. regarded it as a solitary form of synovial chondromatosis and introduced the term “solitary synovial osteochondromatosis” (8). Such cartilaginous lesions were once believed to arise through metaplasia of synovial cells, primitive cells lying within the synovial tissue, or fibroblasts via unknown stimuli (2,3), but were later reported to represent a true neoplasm because of the presence of monoclonality (9). Synovial chondromatosis has recently been reclassified
by the World Health Organization (WHO) as a benign neoplasm that can appear in primary or secondary form (10). Whether these chondroid lesions represent a true neoplasm or non-neoplastic lesion remains controversial (11) and the nomenclature is confusing, but we used the term “synovial chondromatosis” in this report in accordance with the WHO classification.

Cases of solitary synovial chondromatosis have been reported around the knee, foot, nape of the neck, wrist, elbow, and buttocks (3–7,12). To the best of our knowledge, this is the first description of solitary synovial chondromatosis considered to have arisen from the gluteus maximus bursa.

Radiologically, solitary synovial chondromatosis appears as a well-circumscribed, lobulated mass with central calcification in a ring-like, punctate pattern. Plain radiographs are often inadequate to detect calcifications or to locate the tumor (13). In addition, discontinuity with adjacent bone is more clearly demonstrated on CT. On MRI, synovial chondromatosis demonstrates low-to-intermediate signals on T1W imaging and intermediate-to-high signals on T2W imaging (5,14). Cartilaginous areas appear markedly hyperintense on T2W imaging, reflecting the water-rich chondroid matrix. Fat-containing bone marrow may be identified in the densely calcified area as signal hyperintense areas on both T1W and T2W imaging. Calcified matrix shows low signals on both T1W and T2W imaging.

Radiological differential diagnoses include myositis ossificans, tumoral calcinosis, extraskeletal osteosarcoma, and extraskeletal chondrosarcoma. Myositis ossificans is characterized by peripheral ossification, and contains cortical and medullary bone. However, a cartilaginous cap is absent in myositis ossificans. Tumoral calcinosis also appears as a well-circumscribed, lobulated calcified mass, but osseous

Fig. 3. Magnetic resonance imaging of the mass. (a) T1W imaging at the level of CT in Fig. 2 shows signal hyperintensity in the center and signal hypointensity at the periphery. (b) A layer of marked signal hyperintensity is revealed on T2W imaging (curved arrow). (c) On fat-saturated T2W imaging, peripheral signal hyperintensity is more apparent (black arrow). Areas of decreased signal suggest fat component (white arrow).

Fig. 4. Pathological specimen shows hyaline cartilage at the periphery of the mass (white arrows), and intracartilaginous ossification in the center (black arrows). Synovial tissue surrounds the mass (star).
Trabecular structures as seen in soft-tissue chondroma are not found in tumoral calcinosis (15). As densely calcified masses near the joints, extraskeletal osteosarcoma and chondrosarcoma are also important differential diagnoses. Patterns of calcification may be useful in the differential diagnosis. Osteosarcoma shows amorphous calcification described as a cloud-like pattern, which corresponds to osteogenesis in the tumor without enchondral ossification (16). Chondrosarcoma shows chondral calcifications resembling "rings and arcs", which are less dense and less conglomerated than those of osteochondroma. In addition, chondrosarcoma also lacks a cartilaginous cap (17).

Radiological findings may distinguish between synovial chondromatosis and osteosarcoma, or chondrosarcoma. However, close pathological investigation is still necessary, as chondrosarcoma may arise from synovial chondromatosis (18).

The tumor in our case lay on the dorsal aspect of the proximal femur, representing the expected location of the gluteus maximus bursa. Knowledge about bursal anatomy may also help in differential diagnosis. If a mass lesion with the above-mentioned imaging characteristics lies at the expected location of an extra-articular bursae, the likelihood of synovial chondromatosis would be increased.

Our case was difficult to correctly diagnose preoperatively due to its extraordinarily large size and unexpected site of occurrence. In our case, the biopsy specimen comprised chondral tissue that could be interpreted as low-grade chondrosarcoma. It should be noted that without appropriate radiological assessment, accurate pathological diagnosis of chondral lesions cannot be made (19).

In conclusion, solitary synovial chondromatosis in our case showed a similar appearance to skeletal osteochondroma except for the extraskeletal location, as a centrally ossified mass containing bone marrow surrounded by a thin cartilaginous layer showing a bright signal on T2W imaging. Bursal anatomy may also be helpful for this diagnosis. Familiarity with these imaging characteristics will avoid unnecessary biopsy or excessive surgery.

Acknowledgements
We thank Prof. Masaharu Fukunaga (Department of pathology, Jikei University Daisan Hospital) for diagnostic advice.

Declaration of conflicting interests
The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

References
1. Sim FH, Dahlin DC, Ivins JC. Extra-articular synovial chondromatosis. J Bone Joint Surg Am 1977;59:492–495.
2. Chung EB, Enzinger FM. Chondroma of soft parts. Cancer 1978;41:1414–1424.
3. Lim SC, Kim YS, Kim YS, et al. Extraskeletal osteochondroma of the buttock. J Korean Med Sci 2003;18:127–1304.
4. Li C, Arger PH, Dalinka MK. Soft tissue osteochondroma. A report of three cases. Skeletal Radiol 1989;18:435–437.
5. Sowa DT, Moore JR, Weiland AJ. Extraskeletal osteochondromas of the wrist. J Hand Surg Am 1987;12:212–217.
6. Singh R, Sharma AK, Magu NK, et al. Extraskeletal osteochondroma in the nape of the neck: a case report. J Orthop Surg (Hong Kong) 2006;14:192–195.
7. Aydin N, Gokkus K, Topal C, et al. Solitary synovial osteochondroma of the knee: mimicking a giant loose body. Int Med Case Rep J 2012;5:83–86.
8. Edeiken J, Edeiken BS, Ayala AG, et al. Giant solitary synovial chondromatosis. Skeletal Radiol 1994;23:23–29.
9. Sciot R, Dal Cin P, Bellemans J, et al. Synovial chondromatosis: clonal chromosome changes provide further evidence for a neoplastic disorder. Virchows Archiv 1998;433:189–191.
10. Sciot R, Bridge JA. Synovial chondromatosis. In: Fletcher CDM, Bridge JA, Hogendoorn P, et al. (eds). World Health Organization Classification of Tumours. WHO Classification of Tumours of Soft Tissue and Bone. Lyon: IARC Press, 2013, p.261.
11. Steiner GC, Meushar N, Norman A, et al. Intracapsular and paraarticular chondromas. Clin Orthop Relat Res 1994;303:231–236.
12. Al-Najjim M, Mustafa A, Fenton C, et al. Giant solitary synovial osteochondromatosis of the elbow causing ulnar nerve neuropathy: a case report and review of literature. J Brachial Plex Peripher Nerve Inj 2013;8:1.
13. Crotty JM, Monu JU, Pope TL Jr. Synovial osteochondromatosis. Radiol Clin Am 1996;34:327–342.
14. Kramer J, Recht M, Deely DM, et al. MR appearance of idiopathic synovial osteo-chondromatosis. J Comput Assist Tomogr 1993;17:772–776.
15. Palmer PE. Tumoural calcinosis. Br J Radiol 1966;39:518–525.
16. Yarmish G, Klein MJ, Landa J, et al. Imaging characteristics of primary osteosarcoma: nonconventional subtypes. Radiographics 2010;30:1653–1672.
17. Sundram M, Perceflay S, McDonald DJ. Case report 799. Extraskeletal chondrosarcoma. Skeletal Radiol 1993;22:449–451.
18. Evans S, Boffano M, Chaudhry S, et al. Synovial chondrosarcoma arising in synovial chondromatosis. Sarcoma 2014;2015:447593.
19. de Andrea CE, Kroon HM, Wolterbeek R, et al. Interobserver reliability in the histopathological diagnosis of cartilaginous tumors in patients with multiple osteochondromas. Mod Pathol 2012;25:1275–1283.