



Parosteal Lipoma Associated with a Growing Osteochondroma of the Right Ilium

우측 장골능선의 골연골종과 연관된 방골성 지방종의 증례 보고

Min Jae Myung, MD¹ , Ji Seon Park, MD^{1*} , Kyung Nam Ryu, MD¹ ,
Yong-Koo Park, MD² , Myung-Won You, MD¹ , Jong Hun Baek, MD³

Departments of ¹Radiology, ²Pathology, and ³Orthopaedic Surgery, Kyung Hee University Hospital, College of Medicine, Kyung Hee University, Seoul, Korea

Parosteal lipoma is a rare type of lipoma, the incidence being approximately 0.3% of all lipomas. Moreover, parosteal lipoma coexisting with osteochondroma is extremely rare. A few cases with coexistence of osteochondroma and parosteal lipoma have been reported and they were thought to be reactive changes of adjacent bone by parosteal lipoma. However, temporal relationship of these tumors could not be explained. Here, we report a case of parosteal lipoma associated with osteochondroma of the right ilium developed over 6 years, with follow-up radiographs.

Index terms Lipoma; Osteochondroma; Radiography; Multidetector Computed Tomography; Magnetic Resonance Imaging

INTRODUCTION

Lipoma is the most common benign soft tissue tumor and is composed of mature adipose tissue (1). Depending on its location, it is categorized into subcutaneous, intramuscular, intermuscular, intraosseous, intracortical or parosteal (1). Parosteal lipoma only accounts for approximately 0.3% of all lipomas, most of which are found in the extremities, such as the femur, tibia and radius (1). Parosteal lipoma has been reported mainly as a solitary tumor (2). However, there are a few case reports describing this benign tumor coexisting with osteochondroma (2-4). Till date, only one case has reported on the characteristics of parosteal lipoma underlying osteochondroma with different imaging modalities; however, it could not demonstrate the relationship between the

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*Corresponding author

Ji Seon Park, MD
Department of Radiology,
Kyung Hee University Hospital,
College of Medicine,
Kyung Hee University,
23 Kyungheedae-ro,
Dongdaemun-gu, Seoul 02447,
Korea.

Tel 82-2-958-8622

Fax 82-2-968-0787

E-mail francesca@daum.net

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ORCID iDs

Min Jae Myung

[https://
orcid.org/0000-0002-8901-0461](https://orcid.org/0000-0002-8901-0461)

Ji Seon Park

[https://
orcid.org/0000-0002-7842-4407](https://orcid.org/0000-0002-7842-4407)

Kyung Nam Ryu

[https://
orcid.org/0000-0002-8145-4295](https://orcid.org/0000-0002-8145-4295)

Yong-Koo Park

[https://
orcid.org/0000-0001-7209-864X](https://orcid.org/0000-0001-7209-864X)

Myung-Won You

[https://
orcid.org/0000-0001-6262-5784](https://orcid.org/0000-0001-6262-5784)

Jong Hun Baek

[https://
orcid.org/0000-0001-8631-9570](https://orcid.org/0000-0001-8631-9570)

two benign tumors owing to the absence of interval change in imaging findings. Here, we report a case of a parosteal lipoma in a 31-year-old woman associated with osteochondroma of the right ilium over a period of six years with interval radiographs. Moreover, our case shows CT, MRI, and pathologic findings that suggest that the parosteal lipoma developed from the growing osteochondroma.

CASE REPORT

A 25-year-old woman complaining of right buttock pain presented as an outpatient to the Department of Rehabilitation Medicine. Her initial pelvis plain radiograph at that time showed only a small bony excrescence superior to the right anterior superior iliac spine (ASIS) (Fig. 1A). The bony lesion showed medullary and cortical continuity, which suggested an osteochondroma. Avulsion injury of gluteal aponeurotic origin could also be considered because the lesion was too high and posterior to be originating from the ASIS. However, she had no history of trauma and was not an athlete, so the possibility of repeated minor trauma was low. She managed her symptoms with conservative management such as taking pain killers.

Six years later, even with the use of pain killers, her pain persisted and she noticed a palpable enlarged mass in the same location. Consequently, she was referred as an outpatient to the department of orthopedic surgery. On the follow-up pelvis plain radiograph, the small

Fig. 1. A 31-year-old woman with a parosteal lipoma associated with a osteochondroma of the right ilium. **A.** Plain radiograph 6 years ago shows a bony mass (arrow) that protrudes from the superior to the right anterior superior iliac spine of the pelvis, which is connected with the cortex and marrow cavity. **B.** Recent plain radiograph shows enlargement of the bony mass (arrow) with a thick radiolucent lesion (asterisk) partially enveloping the bony mass. However, no radiolucency is observed around the bony mass on the previous radiograph.

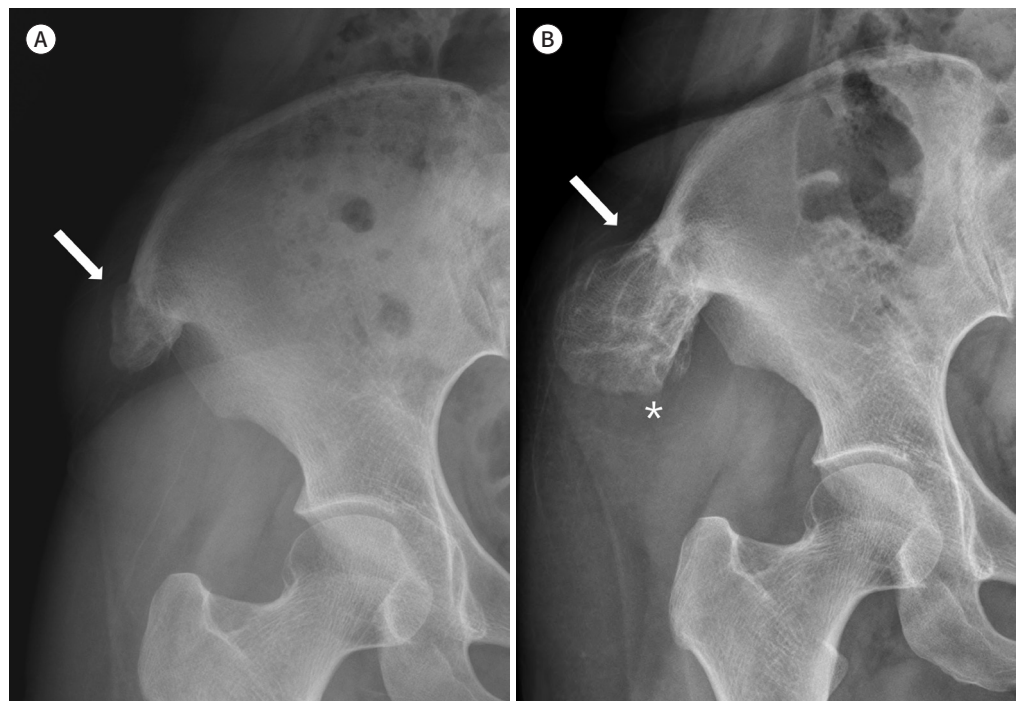
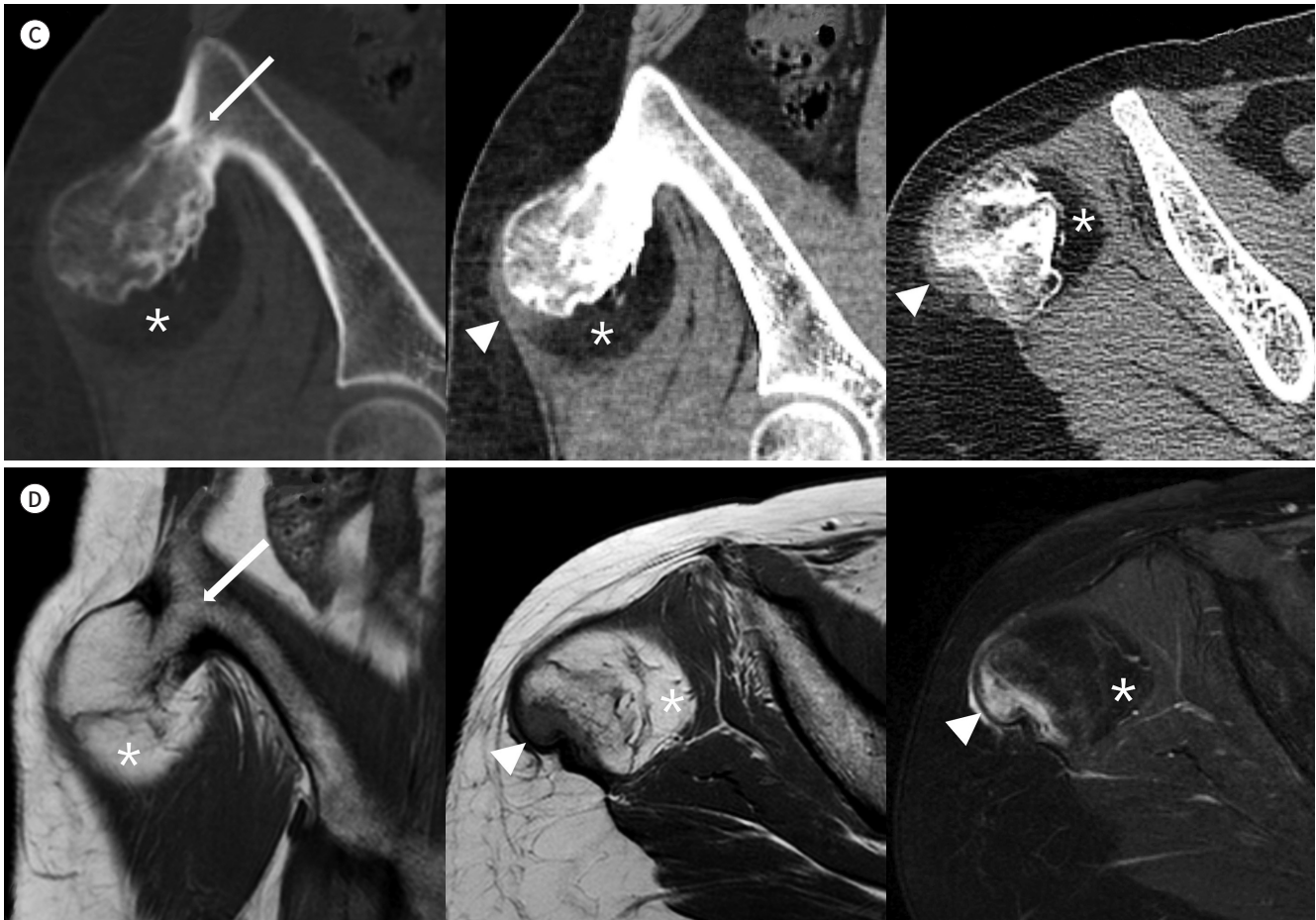


Fig. 1. A 31-year-old woman with a parosteal lipoma associated with an osteochondroma of the right ilium.

C. Pelvic 3-dimensional CT (coronal, coronal and axial images) shows a bony mass. The mass shows connectivity with the cortex and marrow cavity across the right ilium (arrow). The anteromedial aspect shows a fat-attenuated lesion (asterisks), and the posterolateral aspect shows a soft-tissue attenuated lesion (arrowheads).

D. Pelvic bone MRI (coronal T1WI, axial T1WI, and axial FST2) also shows connectivity with the cortex and marrow cavity across the right ilium (arrow) and fat components (asterisks) suggestive of a parosteal lipoma, and soft tissue components (arrowheads) suggestive of a cartilage cap surrounding the bony mass. On axial FST2, the fat component of the parosteal lipoma shows signal drop while the suggested cartilage cap shows high signal intensity.

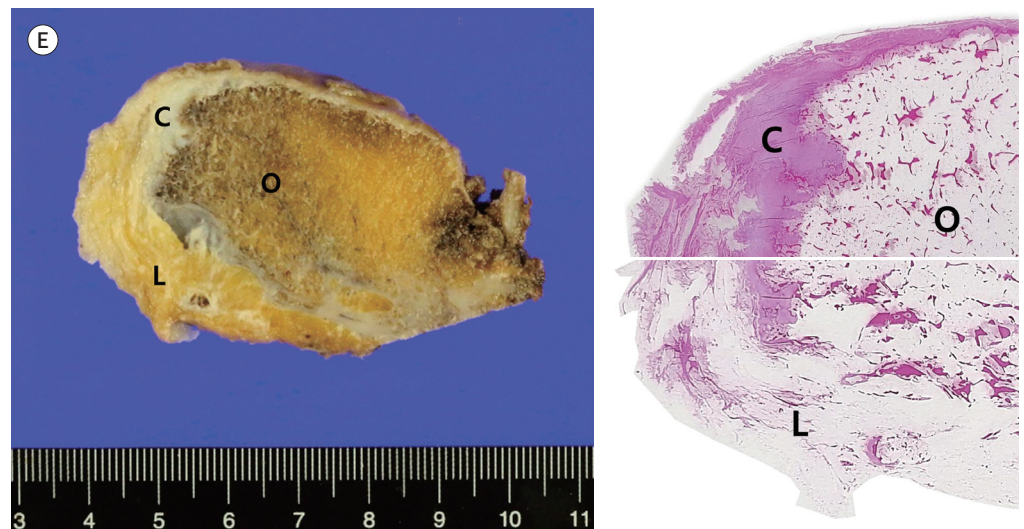
FST2 = fat suppression T2-weighted image, T1WI = T1-weighted image



bony excrescence from the previous film had not only grown in size, but also showed a lesion with thick perilesional radiolucency and the irregular margins of an adjacent bony lesion (Fig. 1B). This perilesional radiolucency seemed like a fat component rather than other soft tissue component. Hence, her doctor recommended a subsequent CT and MRI for further evaluation.

The 3-dimensional CT scan of the pelvis revealed an approximately 5.4 cm × 5.0 cm × 4.8 cm sized protruding bony mass slightly superior to the right ASIS, while maintaining the continuity of the marrow cavity and cortex of the original bone (Fig. 1C). This protruding bony lesion was suggested osteochondroma. The margin of the protruding bony lesion, on the other hand, was irregular, suggesting an overlying expansile soft tissue component (Fig. 1C). This soft tissue component was composed of two distinctive portions, a fatty hypoatten-

Fig. 1. A 31-year-old woman with a parosteal lipoma associated with an osteochondroma of the right ilium. **E.** Photograph of the coronally sectioned gross specimen (left image) shows mass formation with the following three different components: osteochondroma (O), fibrocartilage cap (C), and parosteal lipoma (L). In the low-power photomicrograph of resected lesion (right, hematoxylin and eosin stain, $\times 40$), an osteochondroma (O) composed of the trabecular bone is visible in the center. Surrounding the osteochondroma is a cartilage cap (C) and the outer region is the parosteal lipoma (L), composed of mature fat tissue.



uated portion anteromedially (approximately 3.5 cm \times 4.1 cm \times 2.0 cm in size) and an isoattenuated portion posterolaterally (coronal maximal thickness of 1.8 cm). The isoattenuated portion was not detected on plain radiography, which suggests a cartilaginous cap (Fig. 1C). In particular, the overlying fatty component can be thought of as a parosteal lipoma that envelops most of the protruding bony lesion. The surrounding muscles, including the gluteus medius and minimus, were compressed.

Subsequent MRI also revealed typical findings of osteochondroma with medullary and cortical continuity (Fig. 1D). The suggested cartilage cap was at the posterolateral aspect, which shows intermediate signal intensity on T1-weighted images (Fig. 1D) and high signal intensity on fat suppressed T2-weighted images (Fig. 1D). Previously interpreted as the parosteal lipoma on CT, the anteromedial aspect of the soft tissue showed a high signal intensity on both T1- and T2-weighted images (Fig. 1D), and low signal intensity on corresponding fat suppression images (Fig. 1D). On MRI, subtle soft tissue edema was present between the osteochondroma and overlying fascia lata and proximal iliotibial tract (Fig. 1D). The edema was thought to be due to impingement of the overlying structures and suggested osteochondroma.

Because our patient complained of her lesion, marginal resection was performed. The excised specimen underwent pathologic examination. Grossly, the mass showed osseous components with blue gray fibrocartilage cap and yellowish fatty areas (Fig. 1E). Histologically, the osseous area showed mature trabecular bones and fatty marrow covered by a fibrocartilage cap, which are characteristic features of osteochondroma. The fatty area was composed of mature adipose tissue that tightly attached to the periosteum, consistent with parosteal lipoma (Fig. 1E). The margin between the cartilaginous portion and parosteal lipoma did not form a transition zone, so the possibility that cartilage is affected by metaplastic change in the parosteal lipoma is reduced. She recovered her usual health and her symptoms subsided postoperatively.

DISCUSSION

Parosteal lipoma, an extremely rare benign tumor, is highly associated with the periosteum (1). Parosteal lipoma occurs mainly in the thigh, around the femur, and in the upper extremity around the proximal radius, along with tibia, humerus, and scapula (5). Patients with parosteal lipoma are usually in their 40s and 60s, and they complain of a slow growing, large, painless, and immobile mass (1). While the exact etiology of parosteal lipoma is unknown, the periosteum does not contain adipose tissue, which makes it impossible to be the source of such tumors (6). However, lipoblasts are distributed throughout connective tissue septa and blood vessels and they can occur anywhere in connective tissue (4). Parosteal lipoma around the periosteum causes a reactive change in the periosteum of the surrounding bone, and this characteristic is related to its radiographic appearance (5).

In radiologic examinations, parosteal lipoma is seen as a well-circumscribed fat-containing mass, which is located close to the bone and is characterized by a reactive change in the abutting bone (5). These reactive changes include osseous bowing, cortical erosion, and focal cortical hyperostosis (7). The most common finding is osseous projection. In Murphey's study, all cases showed bone production and the majority showed large bone excrescence (5). These bone excrescences may be confused with osteochondroma, yet connectivity between the cortex and marrow cavity can differentiate between them. The lesions seen on the bony margins of the parosteal lipoma do not show any connectivity with the cortex or marrow cavity (1). On the contrary, osteochondroma has a stalk, that shows direct continuity with the underlying cortex and medullary canal (8).

On the initial plain radiograph, a bony mass is seen in our patient, beyond the excrescence on superior to ASIS of the pelvis, which shows connectivity to the marrow cavity. This finding is specific to osteochondroma. Osteochondroma can have associated complications that are clinically important. Of those, the most feared complication is malignant transformation, which occurs in 1% of osteochondromas in total (9). Imaging findings that suggest malignancy include growth of the osteochondroma in a skeletally mature patient, irregular or indistinct lesion surface, focal radiolucency in the internal lesion, and erosion or destruction of the adjacent bone (9, 10). In addition, there is a report of cartilage cap thickness differences between benign osteochondroma (0.1–3.0 cm; average, 0.6–0.8 cm) and those with secondary chondrosarcoma (1.5–12 cm; average, 5.5–6.0 cm). Therefore, a cartilage cap more than 1.5 cm thick in a skeletally mature patient should be viewed with great suspicion of harboring malignant transformation (9). In our case, the osteochondroma grew in size over 6 years and the margin of the tumor became irregular with newly developed radiolucent areas on serial plain radiographs. Except for fat density lesions, the other two findings could suggest malignant transformation of the osteochondroma.

The new radiolucent portion was characterized as a parosteal lipoma on CT and MRI. The cartilage cap was at the posterolateral aspect of the tumor, which could not be seen on the plain radiograph. The cartilage cap was approximately 1.8 cm thick, around the cut-off value for malignant transformation.

In addition, there are some reasons for the conclusion that we reach. First, in the view of the temporal relationship between two tumor occurrences, only osteochondroma was identi-

fied on initial radiographs, and parosteal lipoma was seen later as a thick radiolucency adjacent to the growing osteochondroma on follow-up radiographs. Of course, the first radiograph cannot completely rule out the presence of parosteal lipoma that is not visible on X-rays. However, although there was no cross sectional image, because the peripheral cortical margin was smooth, clean, and no radiolucency, it is more likely that there was no parosteal lipoma at the first time. Second, the osteochondroma in our case was accompanied by irregularity of the margin in contact with the parosteal lipoma. Although an irregular cortical margin in osteochondroma is known to be indicative of malignant transformation, the cortical changes in our case were thought to be a secondary change caused by the surrounding parosteal lipoma rather than malignant transformation. Third, the fat component of parosteal lipoma forms one mass contour with the osteochondroma as a whole. For these three reasons, we finally concluded that parosteal lipoma with underlying osteochondroma is more plausible than a simple coexistence of two solitary masses. Furthermore, it is different from ordinary osseous excrescence due to reactive changes caused by parosteal lipoma. Of course, it is less likely, but parosteal lipoma from previous avulsion injury of the gluteal aponeurosis can also be considered.

There are few reported cases of parosteal lipoma and combined osteochondroma (2-4). In those previously reported cases, only one case showed evidence of osteochondroma in the imaging study. However, even in that case report, there was no definite mass formation as in our case. Moreover, in our case, the temporal relationship between osteochondroma and parosteal lipoma was clearly observed because there was a plain radiograph obtained 6 years previously. The initial radiograph showed that the osteochondroma was not the result of a reactive change caused by the parosteal lipoma, instead the osteochondroma occurred first and the parosteal lipoma appeared as a second tumor on the original tumor.

In conclusion, we report plain radiograph, CT, and MR findings of parosteal lipoma associated with osteochondroma located on the right ilium with interval change on follow up radiographs.

Author Contributions

Conceptualization, M.M.J., P.J.S., R.K.N.; data curation, M.M.J., P.J.S., R.K.N., P.Y., B.J.H.; investigation, M.M.J., Y.M.; methodology, M.M.J., P.J.S.; resources, R.K.N., P.Y., B.J.H.; supervision, P.J.S.; visualization, M.M.J., P.J.S., P.Y.; writing—original draft, M.M.J.; and writing—review & editing, M.M.J., P.J.S., Y.M.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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우측 장골능선의 골연골종과 연관된 방골성 지방종의 증례 보고

명민재¹ · 박지선^{1*} · 류경남¹ · 박용구² · 유명원¹ · 백종훈³

방골성 지방종(parosteal lipoma)은 모든 지방종의 약 0.3%를 차지하는 드문 유형의 지방종이다. 더구나, 골연골종(osteochondroma)과 공존하는 방골성 지방종은 극히 드물다. 이전에 골연골종과 방골성 지방종이 같이 발견된 증례들이 보고된 적 있으나, 이들은 방골성 지방종에 의해 주변 뼈에 유발된 반응성 변화로 보고되었고, 시간의 변화에 따른 종양들의 선 후관계를 파악하지 못했다. 본 저자들은 6년간의 X선 사진 변화와 함께 골연골종과 연관된 방골성 지방종의 컴퓨터단층촬영, 자기공명영상, 병리학 소견에 대해 보고하고자 한다.

경희대학교 의과대학 경희대학교병원 ¹영상의학과, ²병리학과, ³정형외과