An egg in the leg: Case report of an osteochondrolipoma

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Abstract – Osteochondrolipomas, a very rare combination of chondroid and osseous differentiation within lipomas, are typically found in the neck and head area. We present the case of an osteochondrolipoma in the thigh of a 54-year-old female, with matching histological and cytological correlation. To the best of our knowledge, this atypical location has only been reported once in the literature.

Key words: Lipoma, Osteochondrolipoma, Upper leg, Case report.

Introduction

Lipomas are the most common benign mesenchymal neoplasm in humans. They usually present as painless, slow-growing masses. History is mostly long but sometimes only recently noticed. Their peak incidence occurs in the 5th to 7th decades of life. Lipomas are found almost anywhere in the body. One can distinguish superficial (subcutaneous) and deep lipomas (muscular tissue) depending on their localization. The latter are further divided into intermuscular, intramuscular, parosteal, or intrasosseous. Treatment of choice is a marginal excision, and local recurrence is almost nihil.

Conventionally lipomas are composed of mature adipose tissue, but sometimes they can contain other mesenchymal elements such as fibrous tissue, blood vessels, and, less frequently, cartilage or bone. These variations in composition do not affect treatment nor prognosis [1].

The present paper reports an unusual example of an osteochondrolipoma, which is a mixture of distinct osseous and cartilaginous areas within a bigger adipocytic differentiated mass. Besides this rare combination, its location in the thigh makes it a very uncommon finding. Features and differential diagnosis of osteochondrolipoma are discussed.

Case report

A 54-year-old woman was referred to the University Hospitals Leuven with the preliminary diagnosis of a soft tissue tumor in the right upper leg identified as a mass of 43 mm × 62 mm on plain radiograph and echography.

She had been complaining of irradiating pain on the lateral aspect of the right leg for 4 months and a one-week history of a palpable mass in the right thigh. There was no remarkable medical history and no recent significant trauma to the affected region.

Clinical evaluation revealed a well-defined nodular mass in the deeper aspect of the right tight, measuring about 5 cm. No concomitant tenderness, swelling, erythema, or other superficial skin lesions were found. The range of motion of the right hip and knee were unaffected and palpation for aberrant inguinal lymph nodes was negative.

At the time of referral, plain radiographs of the hip and pelvis showed crumbly calcification within a soft tissue mass posterior to the trochanteric region without a sign of bone erosion.

Echography concluded on a non-growing, well-demarcated inhomogeneous muscular tumor without an argument for myositis (Figure 1). Based on the clinic and first radiological characteristics, one decided for further investigation by means of magnetic resonance imaging (MRI) and a diagnostic ultrasound-guided needle biopsy. T1 sequences showed a high-intensity signal equal to that of the subcutaneous fat, mainly present in the periphery, confirming the fatty characteristic of this lesion. T2 sequences revealed diffuse dystrophic calcification deep in the mass (Figure 2). Despite the proximity of the mass to the femur, no clear continuity with the adjacent femoral cortex or bone marrow was shown. Oedema and adenopathy were absent. Compared to a 2-years old computed tomography (CT) where the mass was overlooked, a minimal volume increase was noted with a current size of 94 mm × 53 mm × 66 mm. The retrospective protocol describes a lesion fully encapsulated by the right musculus quadratus femoris. Biopsy of the lesion demonstrated fibromyxoid to chondroid
mesenchymal cells of unknown origin with no evidence of malign morphologic signs.

Based on this information, a tentative diagnosis of pleomorphic/round cell lipoma was made, and marginal resection was performed. We used the Kocher Langenbeck-type approach with an incision placed slightly more posterior to facilitate exposure and dissection of the sciatic nerve. After muscle splitting of the gluteus maximus fibers with the division of the proximal half of the tendon at 1 cm of its insertions, the sciatic nerve was carefully dissected out and protected. The mass was fully encapsulated by a thin transparent membrane and strongly attached to the underlying femur. The surface of the specimen was stone-hard but smooth, with a yellow shine covering a homogeneous pattern of white and gray dots. The resected tumor was sent for histopathological analysis. Postoperatively, the patient showed no vascular or neurological complications. A mild limp was noted due to dissection of the abductors, which quickly resolved with physiotherapy. At the latest consultation (12 months post-operative), the patient reported manifest improvement of the original discomfort. Clinical examination and ultrasound scans did not show any signs of local recurrence. Plain radiographs did not show any calcified density to suggest local recurrence.

Microscopic examination revealed a well-circumscribed mesenchymal tumor consisting of mature hyaline cartilage islands contiguous with bony trabeculae embedded in mature adipose tissue (Figure 3). No mitotic figure or cytological atypia was observed. Based on these results, we conclude on an osteochondrolipoma, completely excised with clear margins.

Discussion

Generally, when equally divided, a tumor consisting of more than one type of mesenchymal tissue is called a mesenchymoma. The term “osteochondrolipoma” refers to the different
Table 1. Previously reported cases of osteochondrolipomas.*

| No. | Authors                  | Age (y) | Sex | Localisation                | Periostal adhesion | Size (cm) | Duration |
|-----|--------------------------|---------|-----|-----------------------------|--------------------|-----------|----------|
| 1   | Katzter                  | 55      | F   | Ischial region              | Not mentioned      | 9.5 x 7 x 4.5 | ...      |
| 2   | Rau et al.               | 19      | F   | Left forearm                | +                  | 1.9       | ...      |
| 3   | Tasic et al.             | 41      | M   | Left groin                  | –                  | 8 x 5 x 4   | ...      |
| 4   | Rau et al.               | 70      | M   | Left femur                  | –                  | 8         | ...      |
| 5   | Kuyama et al.            | 59      | M   | Lower lip                   | Not mentioned      | 0.9 x 0.5 x 0.5 | 2 mo   |
| 6   | Tasic et al.             | 60      | F   | Tongue                      | –                  | 2.0 x 1.7  | 5 y      |
| 7   | Soullard et al.          | 61      | M   | Submandibular region        | +                  | 4.5 x 4.5 x 4 | >20 y  |
| 8   | Gültekin et al.          | 64      | M   | Mandibular symphysis region | +                  | 2         | 2 mo     |
| 9   | Ensat et al.             | 73      | M   | Left palm                   | –                  | 6.5 x 6.4 x 5 | 5 y    |
| 10  | Sunohara et al.          | 59      | F   | Left axilla                 | +                  | 7.9 x 7.6 x 9.0 | 5 y   |
| 11  | Nisio et al.             | 49      | M   | Left scapular region        | –                  | 3.0 x 3.0  | 1 mo     |
| 12  | Tomonaga and Kudawara    | 58      | F   | Left thigh                  | –                  | 3 x 4      | 3 y      |
| 13  | Choi et al.              | 63      | F   | Left popliteal fossa        | –                  | 4 x 5 x 3  | >1 y     |
| 14  | Kitazawa and Shiba       | 72      | M   | Mandibular symphysis region | –                  | 2 x 1.5 x 1.5 | < 20 y  |
| 15  | Gru and Santa Cruz [4]   | 36      | M   | Chest wall                  | Not mentioned      | –         | < 1 y    |
| 16  | Zhu et al. [5]           | 31      | F   | Right ischium               | +                  | 8 x 8 x 5  | >1 y     |

* Table from Kitazawa and Shiba [2], copied and augmented with accordance of the authors.

amounts of cartilage and bone present in the specimen. This combination of those two rare subtypes has been documented only a few times, and among them, mostly in the head and neck region. Presence in the lower half of the body is uncommon, as previously allighted by Kitazawa and Shiba (Table 1) [2]. To the best of our knowledge, only one other case was reported to occur in the region of the thigh.

Until now, no clear consensus exists about the pathogenesis of osteochondrolipomas. Two main theories have been proposed to explain cartilaginous and osseous differentiation in lipomas. The first one suggests that fatty, osseous, and cartilaginous tissue may arise from multipotent undifferentiated mesenchymal cells. This hypothesis has been reinforced over the last years by several studies showing at the microlevel the presence of different differentiation lines of multipotent stem cells in mature human fatty tissue, and at the second place the presence of cartilage, bone, and fat components (of a calcified mass) located in the thigh of a middle-aged female. Microscopic examination confirmed the presence of cartilage, bone, and fat components within the tumor. After marginal excision, no recurrence occurred at 12 months. Pathogenesis of osteochondrolipoma remains speculative. We reported the case due to its exceptional rarity as being the second described in this region.

In conclusion, we exposed the clinical, radiological, and histological findings of an osteochondrolipoma located in the thigh of a middle-aged female. Microscopic examination confirmed the presence of cartilage, bone, and fat components within the tumor. After marginal excision, no recurrence occurred at 12 months. Pathogenesis of osteochondrolipoma remains speculative. We reported the case due to its exceptional rarity as being the second described in this region.

Conflict of interest

RD, RS, FS, DH, and HW certify that they do not have a financial conflict of interest (e.g., consultancies, stock ownership, equity interest, patent/licensing arrangements, etc.) in connection with this article.

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Ethical approval was not required.

Informed consent

Written informed consent of the patient was obtained for this publication.

Authors contributions

RD wrote the manuscript with support from HW and FS. The operation was carried out by HW and RD. The mass was characterized by RS, DH, SF and HW. HW supervised the project.

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