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Case Report

Osteochondrolipoma: a lipoma with cartilaginous and osseous differentiation of the ischium

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Abstract: Osteochondrolipoma is an extremely rare histological variant of lipoma with osseous and chondroid differentiation, which occurs mostly in the head and neck area and the upper half of the body. In this paper, we present a case of osteochondrolipoma, which displayed components of both cartilage and bone in lipoma, located on the right side of the ischium of a 31-year-old male. On thorough review of literature, no such site has been reported to date. Peculiarities of this case and the diagnostic challenges are discussed.

Keywords: Osteochondrolipoma, ischium, osteochondroma

Introduction

Lipomas are very common benign soft tissue neoplasms. They are usually slow-growing and may occur anywhere in the body. According to their localizations, lipomas are distinguished into superficial and deep lipomas. The latter are further subclassified as intramuscular, intermuscular, intraosseous, or parosteal [1]. Lipomas can rarely have areas of bone formation or grey cartilage nodules and are mostly associated with a parosteal localization of the neoplasm. In this study, we present a case of osteochondrolipoma showing not only major adipocytic differentiation but also areas of bone formation and cartilaginous cell differentiation located in the ischium.

Case summary

Physical examination of a 31-year-old male revealed a mass at the right ischium, which had been present for at least 1 year, without apparent discomfort and limited joint activity. Eight months previously, partial biopsy of the tumor was pathologically diagnosed as osteochondroma in a local hospital. Recently, the mass was increasing, and there was still no pain and other discomfort. The patient visited our hospital for further diagnosis and treatment. He did not have any documented significant past medical history, and no other remarkable medical history could be elicited. On exam, there was a single well-defined mass, which was surrounded by normal skin without ulceration, tenderness, erythema, or swelling of the skin. No dysfunction of the lower extremities was found. Additional examination of the inguinal lymph nodes revealed no remarkable lesion.

Radiographically, computed tomography showed a broad-based, well-demarcated and heterogenous bony mass (8×8×5 cm) in the right ischium, which was deeply localized with attachment to right ischium (**Figure 1B**) and connected with the surrounding bone cortex, and was not contiguous with the marrow space. The adjacent bone cortex was thickened. Inside the mass, fat density was surrounding (**Figure 1A**, **1B**), and was separated by strips of isodensity. Enhanced scan showed no significant enhancement in fat-dense area. No bone abnormalities were found in other bones, and the joint relationship was normal. Based on these features, a provisional diagnosis of angiolipoma was considered, and osteochondroma could not be excluded.

Grossly, en bloc resection of the mass from the right sciatic bone was performed. The base of

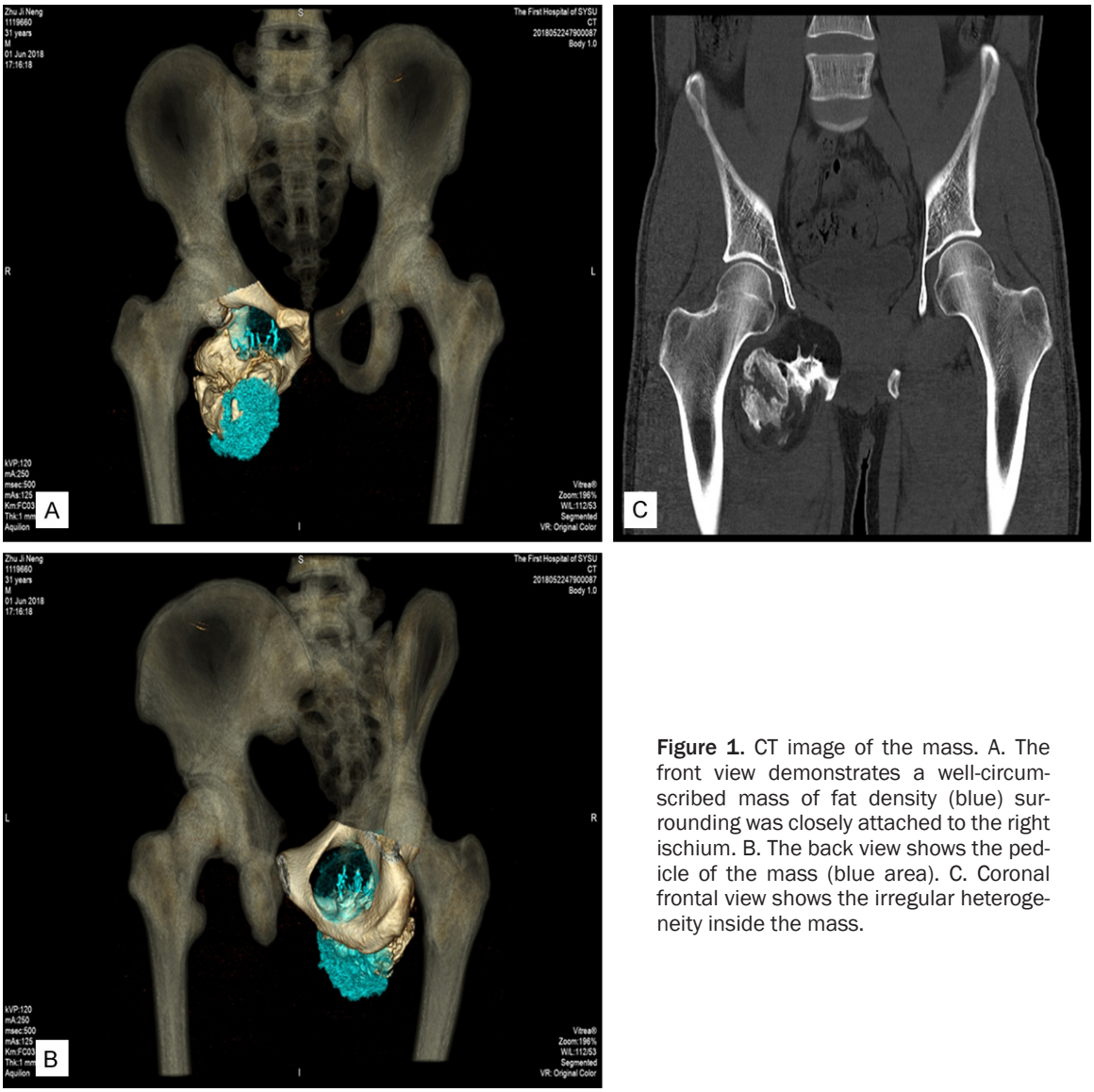

Figure 1. CT image of the mass. A. The front view demonstrates a well-circumscribed mass of fat density (blue) surrounding was closely attached to the right ischium. B. The back view shows the pedicle of the mass (blue area). C. Coronal frontal view shows the irregular heterogeneity inside the mass.

the mass adhered strongly to the underlying ischium. The mass measured 8×8×5 cm and was partly encapsulated by a thin, fibrous membrane. The cut surface of the specimen was yellowish with a mostly homogeneous appearance, with grey white and transparent regions interlaced without any restricted border (**Figure 2**). The gray white or transparent areas were hard, and the gray yellow areas were soft.

Microscopic examination revealed an partly encapsulated lesion mainly comprising of lobules of mature univaculated adipocytes and bony trabeculae, with scattered patchy islands of benign cartilage (**Figure 3**). Additionally, proliferating fibroblasts (paucicellular fibrous stroma with benign fibroblasts) and numerous thick-walled blood vessels were seen among above main components (**Figure 3B**). The bony trabeculae were surrounded by osteocytes and osteoclasts were also apparent (**Figure 3B**). Also, seen in between were foci of mature hyaline cartilage (**Figure 3C**). None of the major components showed any nuclear pleomorphism or immaturity. No intramedullary extraskeletal trilineague hematopoiesis was found, and there was no cartilaginous cap and evidence of zonation. All above ruled out a diagnosis of mesenchymal hamartoma, osteochondroma, myositis ossificans, and well differentiated ossifying liposarcoma. A final diagnosis of osteochondrolipoma (lipoma with bone and cartilaginous differentiation) was made. Considering its rare site, the clinicians and patholo-

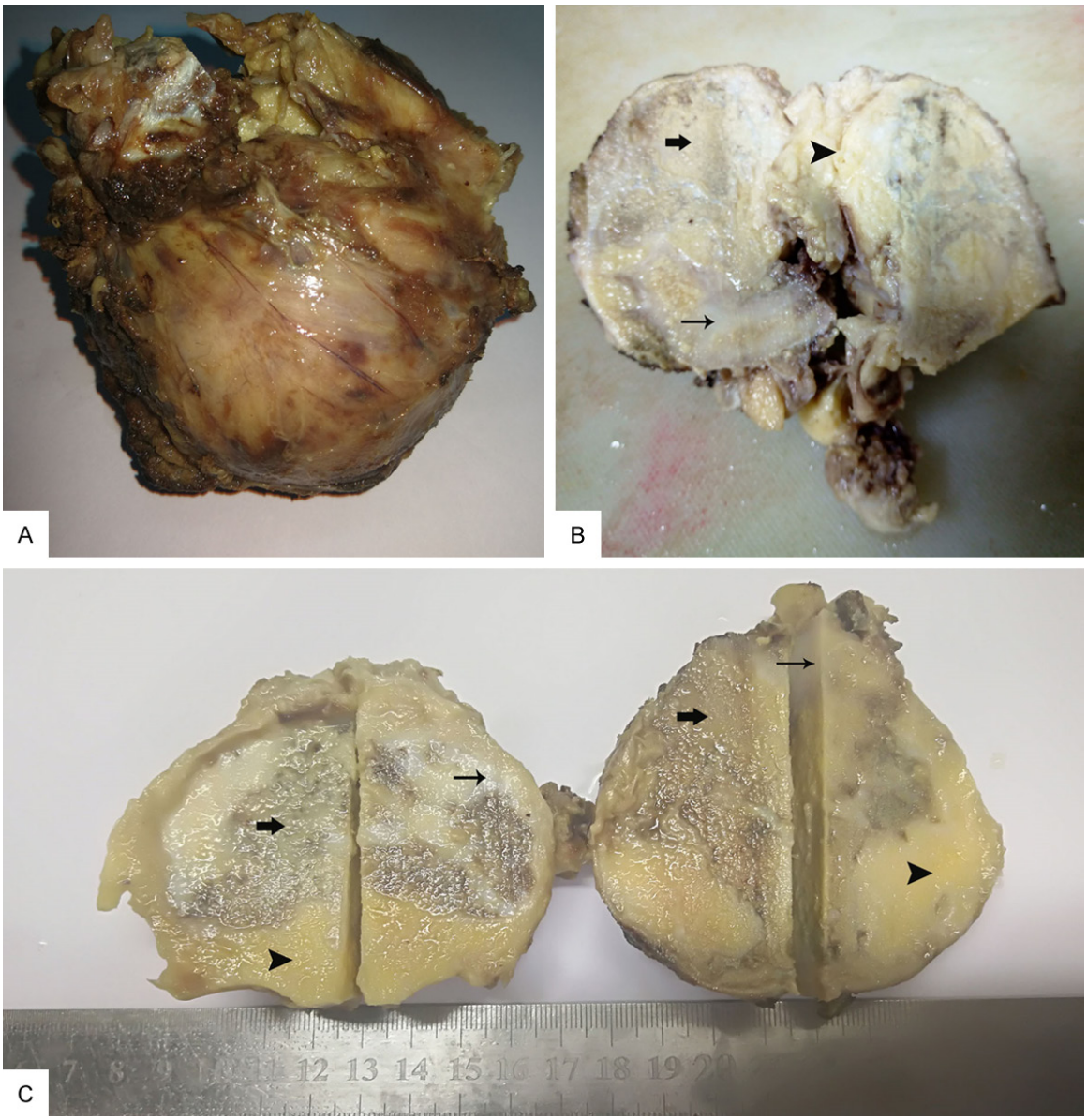

Figure 2. Gross photographs of excised tumor tissue. A, B. The appearance of a well-circumscribed and enveloped mass. C. Cut section showing heterogeneity of grey white (thick arrow), yellow (arrowhead) and focally transparent (fine arrow) interlaced.

gists should regard osteochondrolipoma as a possible diagnosis for a well-defined, calcified mass in the lower half of the body.

Discussion

The term "osteochondrolipoma" was defined by Rau et al. [2]. Osteochondrolipoma is a benign lesion, which is a variant of lipoma that shows an osseous and cartilaginous differentiation. So far, osteochondrolipoma have been reported in very few cases, among which most occured in the head and neck region and the upper half of the body, including the submandibular region [3], tongue [4], the subcutaneous region of the chest wall [5], the scapular region [6], intracranial interhemispheric region [7], the mandible region [8, 9] and intratentorial region [10]. Extremely rarely, they occurred in lower half of the body except the distal leg region reported by Manjula [11], the thigh in the parosteal area [2], and none described in the ischium as our report.

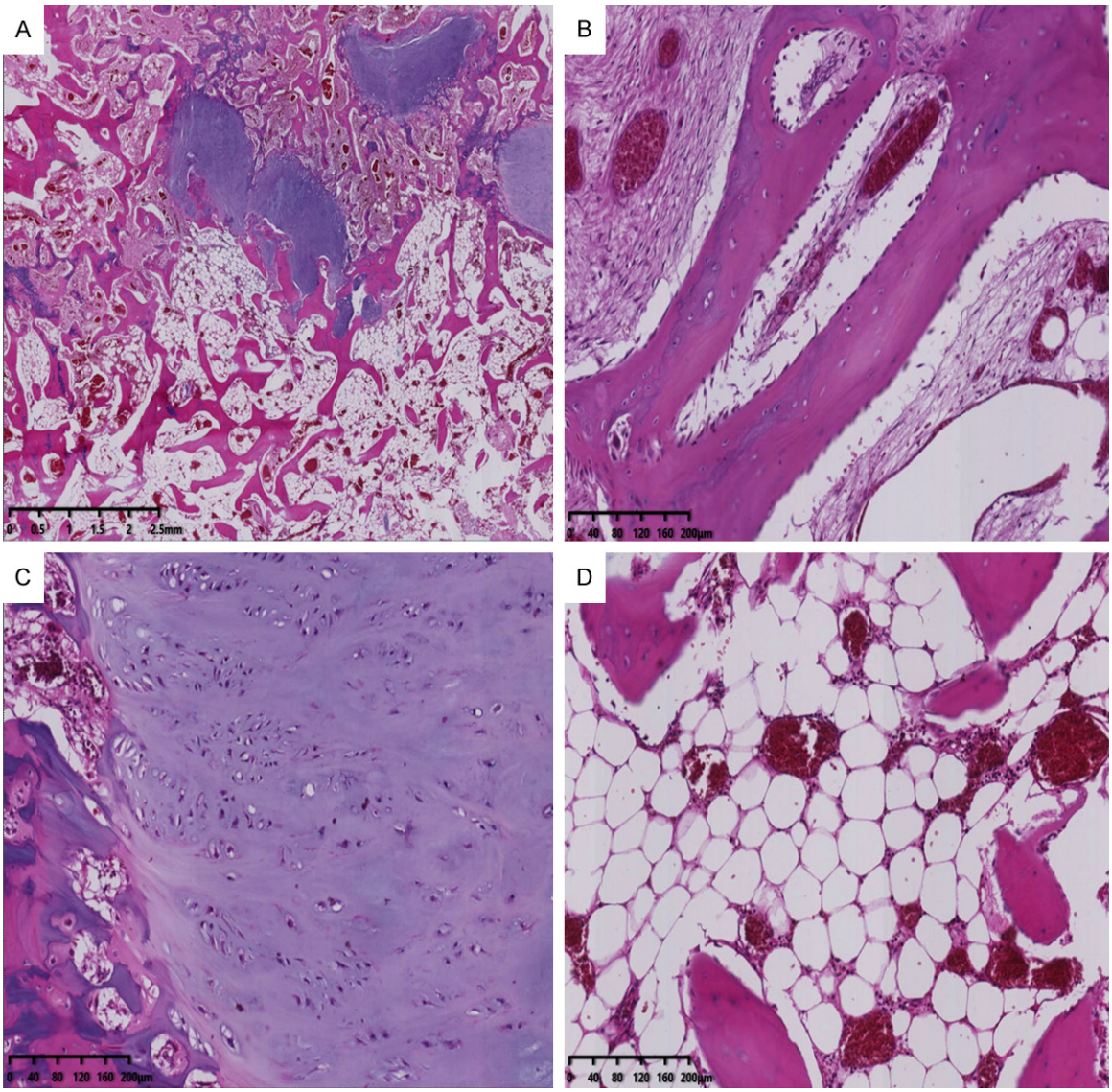

Figure 3. Microscopic examination of excised tumor tissue. A. Low power view photomicrograph of excised tumor tissue shows that components of the tumor including osteoid tissue, hyaline cartilage, and fat tissue arranged disorderly (H&E staining). B. Bony trabeculae showing osteoblastic rimming, which can be seen with osteoblasts inside and surrounded by osteoclasts. C. The hyaline cartilage appears hypocellular. The chondrocytes have uniform and isomorphic nuclei without nuclear hyperchromasia. D. Sheets of mature adipose tissue with interspersed bony trabeculae.

Conventional lipoma is composed of lobules of mature adipocytes. Lipomas can occasionally have areas of bone formation, which can be seen in osteolipoma. Grey cartilage nodules may be seen in chondrolipoma. Histopathologically, osteochondrolipoma is a lipoma with osseous and cartilaginous components. Sometimes, intramedullary extraskeletal trilineague hematopoiesis with a normal cellularity for the patient age was found within the bone. Normal hematopoiesis was composed of erythroid and myeloid precursors with scattered megakaryocytes [5]. In our case, no intramedullary extraskeletal trilineage hematopoiesis was found. As there are at least 4 cellular components of the tumor (lipocytes, fibroblasts, chondrocytes, and osteocytes), some authors hold different opinions about the pathogenesis. All these components may originate from multipotent undifferentiated mesenchymal cells, and cartilaginous and osseous metaplasia changes in a preexisting lipoma itself or the tumor stro-

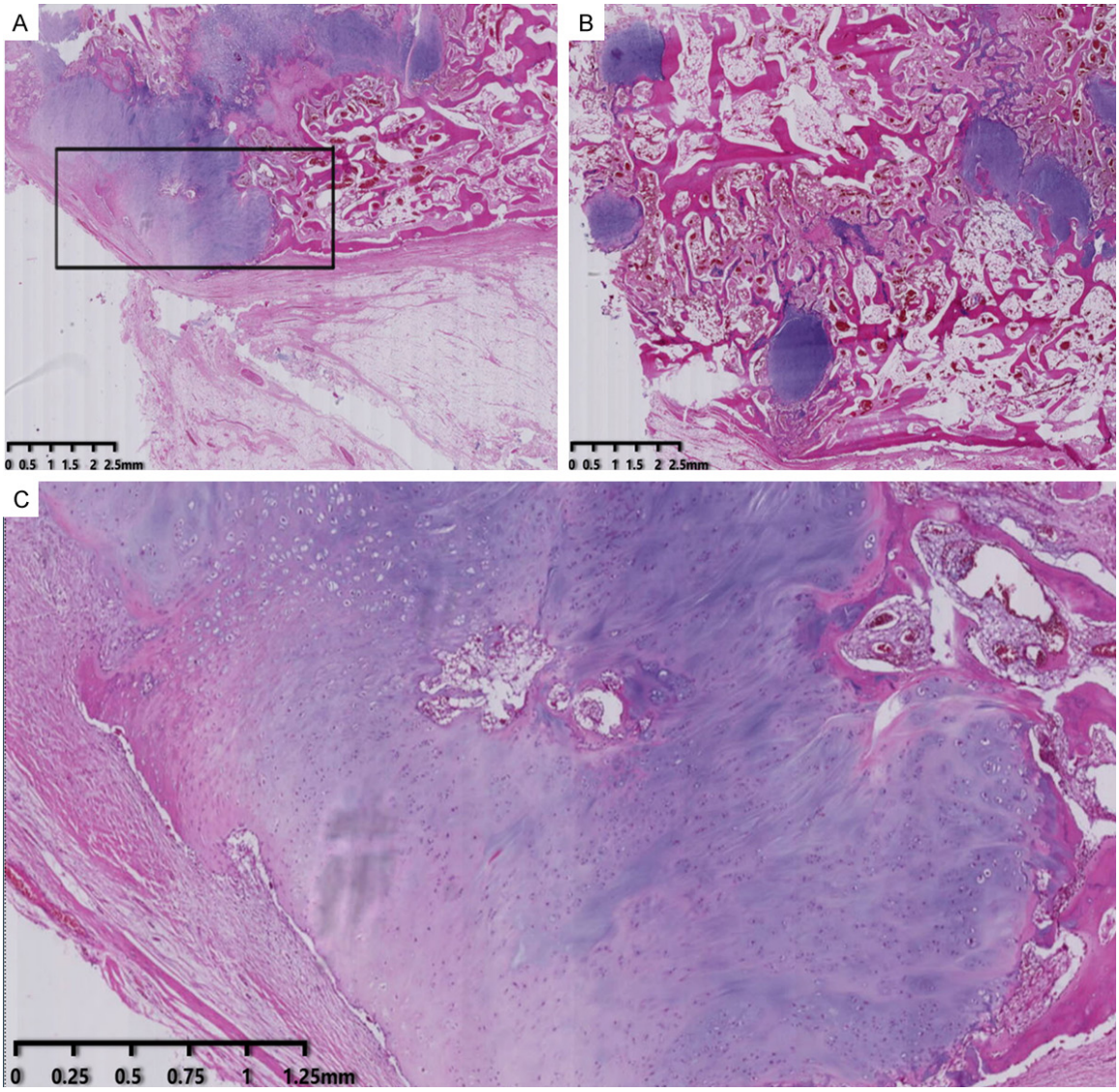

Figure 4. Partial biopsy of the osteochondrolipoma led to the misdiagnosis of osteochondroma. (A, B) Some regions of the tumor are composed mainly of bone and cartilage tissues. (C) Partial region of the tumor with abudant bone and hyaline cartilage without adipose tissue that mimics the photomicrograph of osteochondroma (Magnification of A).

ma also could be responsible [12]. To our knowledge, different differentiation lines of multipotent stem cells have been shown to exist also in adult differentiated fat tissue [13]. In vitro and animal models showed a multidirectional differentiation capacity of adipose tissue-derived stem cells, which allowed formation of bone, cartilage, fat, and fibrous tissue from the same precursor cells [13].

Previous reports showed that osteochondrolipoma could be mobile and non-adherent to the bone or muscle [5] or be firmly attached to the bone [2]. Occasionally, osteochondrolipoma presents as a cyst in popliteal region as reported by Choi [14]. Initially, in the local hospital, CT images showed that the mass contained osseous and cartilage tissue and was closely connected to the periosteum with a broad attachment to the underlying ischium, which is similar to those of osteochondroma in growth pattern, shape, components and appearance to a great extent. Subsequently, part of the tumor was biopsied and only osseous and cartilage tissue were found (**Figure 4**). Both CT images and microscopic changes lead to support a diagno-

sis as osteochondroma in the local hospital. Recently, the patient came to our hospital for excision of the tumor. Computed tomography was redone and the consultation opinion of multidisciplinary case discussion hold that: (1) To be similar to osteochondroma, the present tumor was deeply localized with attachment to right ischium and connected with the surrounding bone cortex. Whereas, the present tumor was not contiguous with the marrow space, which could be distinguished from osteochondroma. (2) Mature fat tissue was not the only predominant component of the tumor, but also the cartilage and bone structures encapsulated within the lipoma were part of the tumor itself, in our opinion, a diagnosis of osteochondrolipoma most specifically.

Besides osteochondroma discussed above, osseous structures and cartilaginous areas raising the differential diagnosis of osteochondrolipoma include secondary hyperostosis of the underlying bone and chondroid lipoma, respectively. Here, osseous structures are not part of the tumor itself [15], whereas in our case, bone structures encapsulated within the lipoma as one of the main components. Chondroid lipoma also shows focal hyalinization of the tumor matrix but has an immature aspect with multivacuolated cells and myxoid changes [16]. Other tumors including teratoma, osteoma, ossifying fibroma, and myositis ossificans should be taken in consideration in the differential diagnosis as well. However, mature fat is not a main component in these entities. Considering its rare site of osteochondrolipoma in our case, the clinicians and pathologists should regard osteochondrolipoma as a possible diagnosis for a exogenous, calcified mass in the lower half of the body.

To conclude, osteochondrolipoma has never been reported in the ischium region. We report this case due to its rare occurrence and being the first case reported in the ischium region. Osteochondrolipoma has the same prognosis as a simple lipoma [6]. Treatment of choice for an osteochondrolipoma is complete surgical excision. So far, recurrences have not been reported [4].

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Disclosure of conflict of interest

None.

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