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CASE REPORT

Secondary chondrosarcoma of the iliac bone in a young woman – a rare case report and review of the literature

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ABSTRACT

Introduction and aim. Chondrosarcomas are malignant cartilage-forming tumors, with secondary cases rarely arising from osteochondromas. This report presents a case of secondary chondrosarcoma developing from an undiagnosed pelvic osteochondroma in a young female, emphasizing the importance of early detection and timely intervention.

Description of the case. A 28-year-old woman with hip and back pain underwent magnetic resonance imaging, revealing a lesion in the left iliacus muscle with sacroiliac extension. Biopsy confirmed a chondroid neoplasm, and positron emission topography-computed tomography showed minimal metabolic activity. Due to the extensive soft tissue component and recurrence risk, wide local excision was performed. Histology confirmed well-differentiated secondary chondrosarcoma, WHO grade 1 arising from an undiagnosed pre-existing osteochondroma.

Conclusion. The described case highlights the critical role of radiological and histopathological evaluation, timely surgical intervention, and multidisciplinary management for optimal patient outcomes.

Keywords. chondrosarcoma, iliac bone, malignant transformation, osteochondroma

Introduction

Chondrosarcomas are locally aggressive or malignant tumors that form a cartilaginous matrix, and make up around one-fifth of all primary malignant bone tumors. Conventional primary chondrosarcomas arise without a benign precursor, while secondary types can be central (from enchondromas), peripheral (from osteochondromas), or periosteal (on the bone surface near the periosteum). Its global incidence varies, ranging from less than 10% in India and Saudi Arabia to over 45% in Finland, Slovenia, and the Netherlands.[1]

Chondrosarcoma mainly affects middle-aged to older adults and is more common in males. It typically arises in the pelvic bones, femur and humerus, with rare occurrences in the trunk, skull, facial bones, hands and feet. Periosteal chondrosarcoma primarily affects the metaphysis of long bones, with a predilection for the humerus and distal femur.[2]

The evolution of osteochondroma into chondrosarcoma is a relatively infrequent yet well-documented occurrence, often leading to the development of low-grade tumors, although higher-grade variants can also arise. This transformation is more prevalent in adults, particularly those with conditions like multiple hereditary exostoses (MHE), with only rare instances reported in pediatric patients.[3] Osteochondromas represent a significant portion of benign bone tumors, comprising 20–50% of such cases and are among the most com-

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mon bone neoplasms. They frequently manifest in the metaphysis and metaphyseal equivalents, with the femur being the primary site of occurrence (30% of cases), while the pelvis, scapula, and spinal involvement are less common.[4] Malignant progression to chondrosarcoma, a complication seen in approximately 1% of solitary osteochondromas, is typically identified by persistent lesion growth post-skeletal maturity and a hyaline cartilage cap thickness of more than 1.5 cm.[5]

Aim

Progression of an osteochondroma in the pelvis of a young female is an uncommon occurrence, particularly without prior imaging, making the diagnosis unexpected. A prompt biopsy and appropriate management of the lesion are crucial for a thorough and effective treatment strategy.

The aim of the study was to present a case of secondary chondrosarcoma developing from an undiagnosed pelvic osteochondroma in a young female, emphasizing the importance of early detection and timely intervention

Description of the case

We present a 28-year-old Indian woman who presented with hip and backache. Notably, the severity of her symptoms prompted her to seek medical attention only recently. Clinical evaluation revealed no palpable tenderness or limitations in range of motion. Bilateral lower extremity strength was symmetric, and reflexes were within normal limits. Laboratory findings indicated a normal complete blood count (CBC). Upon observation of concerning symptoms, a pelvic magnetic resonance imaging (MRI) was conducted, which revealed a well-defined lesion within the left iliacus muscle exhibiting T1 isointensity and T2 hyperintensity, with a fusiform extension into the left sacroiliac joint with calcification and patchy peripheral and central septal enhancement. Differential considerations were proposed, including benign neurogenic tumor like schwannoma or intramuscular myxoma.

A preliminary ultrasound-guided biopsy of the lesion was done, which was reported as a chondroid neoplasm. A cartilaginous neoplasm extending into soft tissue was high on the list of differential diagnoses, even though prior imaging confirming the presence of an osteochondroma was unavailable.

Subsequently, a positron emission tomography–computed tomography (PET-CT) fusion study was done for staging and showed minimal metabolic activity within the lesion, accompanied by a discernible mass effect on the left psoas muscle *(Fig. 1)*. Both imaging modalities also revealed polycystic ovarian morphology in both ovaries.

After appropriate preoperative tests, including negative results for viral markers screening (HIV, HBsAg, and Anti-HCV), the patient was deemed fit and taken up for surgery.

Given the extensive soft tissue component and the risks of recurrence and metastasis at a young age, wide resection was chosen as the treatment approach.

Gross examination disclosed a nodular, well-circumscribed lesion within the soft tissue measuring 4×3.5×2.2 cm. The cut surface was myxoid and gray-tan, with focal gritty areas *(Fig. 2)*.

Microscopically, the lesion comprised of neoplastic chondroid tissue with minimal increase in cellularity arranged as nodules and lobules separated by both thin and thick fibrous septae and exhibiting focal cystic change. The atypical chondrocytes showed nuclear enlargement and occasional binucleation. The mitotic rate was observed to be 2–4 per 10 high-power fields. Additionally, portions of a pre-existing osteochondroma were identified, with the lesion arising from the bony trabeculae. (Fig. 3). A diagnosis of well-differentiated secondary chondrosarcoma WHO grade 1 was rendered.

The patient was counselled regarding the diagnosis and the importance of regular follow-up for monitoring and management of their condition.

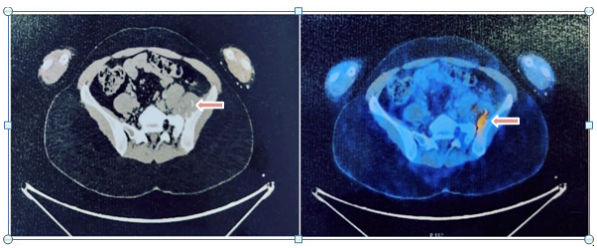

Fig.1. PET-CT FUSION study revealing a well-defined lesion with minimal metabolic activity within the substance of left iliacus muscle with fusiform extension into the left sacroiliac joint

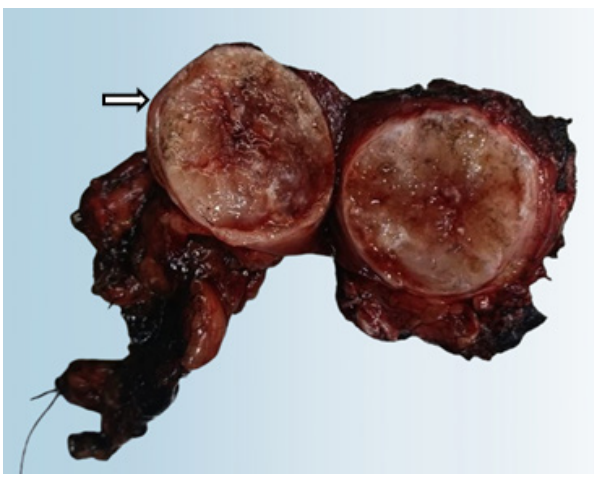

Fig. 2. Macroscopic image showing a well-circumscribed lesion in the soft tissue with myxoid appearance

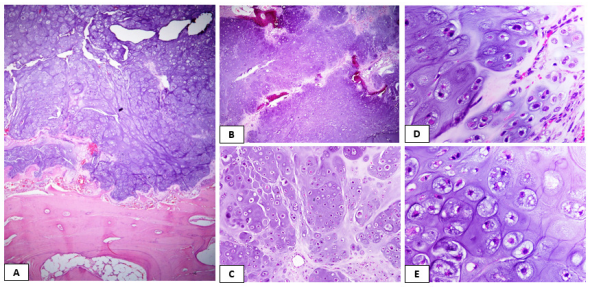

Fig. 3. Microscopic images showing A: Hematoxylin and eosin stain (H&E), 40×, bony trabeculae with thick lobulated cartilaginous cap composed of atypical cartilaginous tumor causing scalloping of the cortex, B: H&E, 40×, hyaline cartilage matrix entrapping the pre-existing lamellar bone, C: H&E, 100×, neoplastic lobules with increased cellularity and occasional binucleation, D: H&E, 400×,cells exhibiting mild nuclear pleomorphism and vacuolated cytoplasm

Discussion

Osteochondromas commonly affect the femur, with distal lesions being predominant, followed by the tibia and humerus. At the same time, those originating from flat bones are rare, with less apparent medullary continuity on radiographs.[6] The transformation of an osteochondroma into chondrosarcoma is a rare phenomenon, particularly in cases not associated with syndromes like multiple hereditary exostoses. This transformation becomes even more uncommon when considering solitary osteochondromas. Indeed, documented cases of secondary chondrosarcoma originating from solitary pelvic osteochondromas are rare. To date, few cases and series have been reported in medical literature, underscoring the exceptional nature of such occurrences and the need for further research to understand the underlying mechanisms.[7,8]

Few studies have suggested that estrogen contributes to cartilage metabolism and human growth, with its involvement in chondrosarcoma, particularly through estrogen receptor alpha, indicating a role in tumor proliferation.[8] Additionally, some studies indicate that the active estrogen-signaling pathway may not have a significant role in the development and progression of chondrosarcoma.[9] The hypothesis regarding the possible role of PCOS (polycystic ovary syndrome) in the progression of chondrosarcoma requires further in-depth research.

Alarming signs such as sudden pain without trauma, recent increase in tumor size, along with a thick cartilaginous cap may suggest malignant degeneration, needing confirmation. From a management perspective, pelvic chondrosarcoma presents surgical challenges because of its size and its proximity to critical structures. Wide-margin surgical resection is recommended for high-grade cases, while curettage alone or in combination with adjuvant therapy may be adequate for low-grade lesions.

Local recurrence is common, especially in pelvic secondary chondrosarcoma, affecting 10-20% of patients.[7,10] Despite these challenges, comprehensive approaches can lead to favorable outcomes in patients undergoing pelvic resection for chondrosarcoma.[11]

Tsuda et al.[12] analyzed 51 cases of secondary chondrosarcomas arising from osteochondromas, with a median age of 36 and a 6.9-year follow-up. In their study, the pelvis was most commonly affected (59%), with 69%, 25%, and 6% showing grade I, II, and III tumors, respectively. Preoperative biopsy accurately predicted the final grade in 27% of cases. They reported a 10-year disease-specific survival rate of 89.4%, noting a higher rate of local recurrence in pelvic tumors (37% vs. 19% in limb tumors). Wide or radical resection was linked to improved local recurrence-free survival.

Righi et al.[13] retrospectively (1943 to 2019) analyzed 214 cases of secondary peripheral chondrosarcomas from solitary osteochondromas. The median patient age was 38 years, with a male-to-female distribution of 66.4% to 33.6%. They reported a 17.3% local recurrence rate and a 5.1% metastasis rate. High histologic grade was the only factor linked to worse 5-year and 10-year overall survival, highlighting the importance of accurate histological assessment and long-term follow-up for this rare chondrosarcoma variant.

Ahmed et al.[14] conducted a study on 107 patients diagnosed with secondary chondrosarcoma, originating either from a solitary osteochondroma (61 cases) or multiple hereditary exostoses (46 cases). Compared to primary chondrosarcoma, these patients were generally younger by one to two decades and showed a male predominance, with tumors more commonly affecting flat bones. Radiologically, malignant transformation was marked by irregular margins, uneven mineralization, and the presence of a soft tissue mass. Histologically, the majority of tumors were well-differentiated, with only ten cases classified as Grade 2.

At the microscopic level, differentiating between enchondroma and atypical cartilaginous tumor relies on several growth patterns. Key features that suggest malignancy include the tumor permeating into the surrounding lamellar bone, causing bone destruction and forming resorption spaces known as Howship's lacunae. Additionally, the lack of new layers of lamellar bone forming around the edges of cartilage nodules, a process called encasement, further supports a malignant diagnosis. Additionally, a myxoid matrix comprising more than 20% of the tumor is also an indicator of malignancy.[15]

Radiology alone cannot reliably differentiate between benign and malignant cartilaginous neoplasms. Some studies suggest that PET-CT could potentially help distinguish between low- and high-grade chon-

drosarcomas based on standardized uptake values.[16] In this case, the PET-CT showed minimal metabolic activity, which corresponded to the low-grade nature of the chondrosarcoma. Due to the absence of prior radiological evidence of osteochondroma in this case, the diagnosis of a chondroid neoplasm in the preoperative biopsy of the lesion was crucial in guiding surgical decisions. Due to the predominant soft tissue component, a pre-existing osteochondroma was confirmed only in the resection specimen. This underscores the importance of both preoperative and postoperative histopathological examination in guiding further management plans.

However, the absence of long-term follow-up remains a limitation. Additionally, further research into the molecular and hormonal pathways involved in the progression of secondary chondrosarcomas is essential to improve diagnostic accuracy and develop targeted therapeutic strategies.

Conclusion

Presented case report of a 28-year-old woman diagnosed with secondary chondrosarcoma, which developed from a previously unrecognized osteochondroma of the iliac bone highlight the importance of both radiological and histopathological examination, appropriate surgical intervention and close follow-up. It should be emphasize the need for a comprehensive, interdisciplinary approach involving orthopedic, oncological, and pathological expertise.

Declarations

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Author contributions:

Conceptualization, T.D., S.L.N., A.S. and K.N.; Methodology, T.D. and S.L.N.; Validation, T.D., S.L.N., A.S. and K.N.; Formal Analysis, T.D., S.L.N., A.S. and K.N.; Investigation, T.D., S.L.N. and A.S.; Resources, T.D. and S.L.N.; Data Curation, T.D., S.L.N., A.S. and K.N.; Writing – Original Draft Preparation, T.D.; Writing – Review & Editing, S.L.N., A.S. and K.N.; Visualization, T.D. and S.L.N.; Supervision, S.L.N., A.S. and K.N.; Project Administration, S.L.N.

Conflicts of interest

The authors have no conflicts of interest to declare.

Data availability:

The data that support the findings of this study are available from the authors.

Ethics approval:

Institutional Ethical Committee approval has been obtained (AMH-C-S-051/06-24).

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