

GSC Biological and Pharmaceutical Sciences

eISSN: 2581-3250 CODEN (USA): GBPSC2 Cross Ref DOI: 10.30574/gscbps Journal homepage: https://gsconlinepress.com/journals/gscbps/

(CASE REPORT)



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Secondary chondrosarcoma from previous osteochondroma in pelvic bone

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GSC Biological and Pharmaceutical Sciences, 2022, 19(03), 248-252

Publication history: Received on 17 May 2022; revised on 21 June 2022; accepted on 23 June 2022

Article DOI: https://doi.org/10.30574/gscbps.2022.19.3.0248

Abstract

Secondary chondrosarcoma is a known dreaded complication of osteochondral- mas, generally first suspected by newonset pain; imaging studies indicate the thick cartilaginous cap indicates possible secondary malignant transformation. In this article, we discuss a case of secondary chondrosarcoma arising from os- osteochondroma in the pelvic bone.

Keywords: Osteochondroma; Secondary Chondrosarcoma; Cartilaginous Cap; Pelvic Bone

1. Introduction

The transformation of an osteochondroma to chondrosarcoma is a well-documented, albeit rare, occurrence. The conversion usually results in low-grade chondrosarcoma, but higher-grade tumors are also possible. It is usually associated with syndromes like multiple hereditary exostoses (MHE) and is far more common after maturity, with only a few cases reported in young children [1-3].

20–50% of all benign bone tumors and 10–15% of all bone tumors are osteochondromas, making them the most prevalent bone tumors. Common locations affected by osteochondroma are metaphysis and metaphyseal equivalents. The femur bone is the single most frequently affected bone(30% of cases); flat bones such as pelvic (5%), scapula(4%), and spine (2%) are less frequently involved [4]. a rare complication of osteochondroma is Malignant transformation to chondrosarcoma, seen in 1% of solitary osteochondromas, and often noted by continued lesion growth after skeletal maturity and a hyaline cartilage cap thicker than 1.5 cm [5].

Chondrosarcoma is a relatively common primary bone tumor, and it is a primary conventional subtype in more than 90% of cases; conventional chondrosarcoma may be either de novo or arise secondary to a benign tumor with chondroid matrix, in this case, most commonly in patients with solitary or multiple osteochondromas, many of these secondary chondrosarcomas are low to intermediate grade tumors, with overall good prognosis and 5 year survival figures about 90% [6].

The most common malignant bone neoplasm is osteosarcoma, followed by chondrosarcoma and Ewing sarcoma. Clinical and radiological features are also important in diagnosing and completing bone cancer staging. Several non-specific serological or specific molecular markers for these bone neoplasms exist. Molecular markers improve diagnostic accuracy and aid in subtyping bone tumors. New advanced biomarkers, such as molecular pathologic, can diagnose benign and malignant tumors [7, 8]. Nitrotyrosine, COX-2, and CD34 were significantly associated with histological grades of chondrosarcoma, implying that these markers may play a role in patient survival [9]. QPCR

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confirmed that high COX-2 protein expression was found primarily in solitary peripheral chondrosarcoma and enchondromatosis-related central chondrosarcoma. They emphasize celecoxib's potential role in treatment [10].

This case report presents Secondary chondrosarcoma from the previous osteochondroma in pelvic bone.

2. Case Presentation

31-year-old male, with progressive low back and left lower extremity pain came to the clinic, patient symptoms lasted more than several months but became severe enough in the last month before he sought medical attention. On his clinical examination, no abnormal tenderness or limited range of motion was detected; both lower extremities forces were symmetric, and reflexes were 2+ bilaterally. A normal CBC and mild ESR elevation (37) were noted on laboratory data.



Figure 1 Frontal pelvic x-ray and axial CT scan show large lytic expansile soft tissue mass over the left sacroiliac joint with chondroid soft tissue matrix, caused sacral and iliac bone erosion, coronal PD fat-saturated image, shows thick cartilaginous cap without significant surrounding edema, axial T2 weighted image, shows a large heterogenous high

signal mass with lobulated border in left sacroiliac joint extending to paravertebral muscles and compressing left L5 nerve root

Due to concerning symptoms, a pelvic radiograph was done, which showed a large heterogeneous mass with chondroid matrix adjacent to the left sacroiliac joint with extension to the sacrum and iliac wings; in order to better define lesion location and extensions patient underwent CT, which revealed heterogenous lytic expansile lesion within left sacral ala with chondroid matrix and extension to sacroiliac joint, left iliac wing and left paravertebral muscles, further work up by MRI, showed thick cartilage cap without surrounding bone marrow or soft tissue edema, invasion of left L5, S1, and S2 neural foramina were also noted, subsequent bone scan with Tc-99-MDP showed abnormally increased uptake.

After reviewing these findings, the diagnosis of Secondary Chondrosarcoma from Previous Osteochondroma in Pelvic Bone has been made.

3. Discussion

The transformation of an osteochondroma into chondrosarcoma is a rare occurrence. However, this transformation is typically associated with syndromes such as multiple hereditary exostoses, which occur much more frequently after maturity. Considering relatively low pelvic bone involvement and also the low likelihood of secondary chondrosarcomatous transformation in solitary osteochondroma, secondary chondrosarcoma of sacral bone due to malignant degeneration of solitary osteochondroma is an exceedingly rare presentation which, to our best of knowledge, only one similar case has been reported in the literature [11] clinical findings as recent onset pain without preceding trauma or fracture. Also, a thick cartilaginous cap with recent tumor growth is alarming signs of malignant degeneration, which warrants further workup and pathologic confirmation.

The treatment of choice for high-grade chondrosarcoma, if amenable, is wide-margin complete surgical resection, however, in cases of low-grade lesions, curettage with or without local adjuvant treatment may be considered. Several novels and trending therapeutic targets for radiofrequency ablation (RFA) have been studied [12]. There is mounting evidence to support the use of this treatment in the spine as well [13]. RF ablates and creates an electromagnetic field. In thermal RF, the patient's tissue is the therapeutic target. RF-induced interactions cause heat, necrosis, and tissue destruction, relieving pain or burning the painful nerve [14]. Local recurrence is a significant complication of chondrosarcoma, particularly in secondary chondrosarcoma of pelvic, which can be problematic in 10-20% of these patients [6, 15].

Patients with osteochondromatosis are at a higher risk of developing secondary chondrosarcoma. Because of the large tumor size, local extension, and complex anatomy with proximity to major neurovascular structures, intestinal and urinary tract, chondrosarcoma in the pelvic region presents a significant challenge for orthopedic surgeons. Proper selection of patients, preoperative planning, and wide surgical margins with reconstruction provide good local control and clinical outcomes following pelvic resection. Patients with huge tumors need a large amount of blood transfusion and postoperative intensive care due to longer surgery. The surgical site infection is a common complication after pelvic resection and can be managed with debridement and antibiotic therapy [16].

Tsuda et al. reported on secondary chondrosarcomas arising from osteochondromas, reviewing 51 secondary chondrosarcomas raised from osteochondromas. The median age was 36, and follow-up was 6.9 years. Most cases affected the pelvis (59 percent). Grade I was seen in 35 (69%), II in 13 (25%), and III in 3 patients (6%). In 27% of patients, preoperative biopsy histology predicted the final grade. All patients' 10-year disease-specific survival DSS was 89.4%. Local recurrence occurred in 15 patients (29%), more often in pelvic than limb tumors (37%). (19 percent). Wide/radical margin improved local recurrence-free survival (p = 0.032), and local recurrence worsened DSS (p = 0.005). They recommend wide/radical resection margins for secondary chondrosarcoma of the pelvis. Patients with limb secondary chondrosarcomas must weigh surgery morbidity and local recurrence risk [17].

Righi et al. examined 214 secondary peripheral chondrosarcomas that arose solely from solitary osteochondromas diagnosed in a multidisciplinary setting. With a median age of 38 years, there were 66.4 % males and 33.6 % females. The rate of local recurrence was 17.3 %, while the rate of metastases was 5.1 %. Aside from age, a high histologic grade is the only factor associated with worse 5-year and 10-year overall survival. Furthermore, high histological grade and surgical debulking were linked to significantly worse disease-free survival.

They proposed that secondary peripheral chondrosarcomas have a low-grade behavior and that, when possible, wide surgical excision is the best treatment option for those that arise in solitary osteochondromas. The tumor's location

does not affect prognosis, but accurate histological grade assessment correlates with tumor aggressiveness, and longterm follow-up is required for this rare variant of chondrosarcoma [18].

Ahmed et al. reviewed 107 patients with secondary chondrosarcoma arising from a single osteochondroma (61 patients) or multiple exostoses (46 patients). Secondary chondrosarcoma patients were one to two decades younger than primary chondrosarcoma patients. There was a male predominance and a preference for flat bones. Sarcomatous degeneration was characterized by irregular margins, inhomogeneous mineralization, and an associated soft tissue mass. The tumors were generally well-differentiated. Only ten tumors were graded as Grade 2. For patients who received initial treatment at the authors' institution, the 5-year and 10-year local recurrence rates were 15.9 % and 17.5 %, respectively, and the 5- and 10-year mortality rates were 1.6 percent and 4.8 percent, respectively. Metastasis occurred in five patients, with four developing in the lung and one developing in the groin region. The majority of cancer patients died from a local recurrence. Wide excision had the lowest rate of local recurrence. Successful surgical treatment patients may have long-term disease-free survival [19].

4. Conclusion

There have only been a few small case series describing chondrosarcoma in youth. Secondary chondrosarcoma descriptions are even rarer, with only a few cases reported as part of larger case series. Chondrosarcoma is a rare and challenging diagnosis in children. The distinction between chondrosarcoma and chondroblastic osteosarcoma is frequently contested, and the treatment implications of distinguishing these diagnoses are critical. we presented a Secondary Chondrosarcoma from Previous Osteochondroma in Pelvic Bone, in a 31-year-old male, with progressive low back and left lower extremity pain.

Compliance with ethical standards

Acknowledgments

We thank the Department of Radiology, Iran University of Medical Science, Tehran, Iran, for providing the data.

Disclosure of conflict of interest

There is no conflict of interest for any of the authors

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

References

- [1] Pierz KA, Womer RB, Dormans JP. Pediatric bone tumors: osteosarcoma ewing's sarcoma, and chondrosarcoma associated with multiple hereditary osteochondromatosis. Journal of Pediatric Orthopaedics. 2001;21(3):412-8.
- [2] Gadwal SR, Fanburg-Smith JC, Gannon FH, Thompson LD. Primary chondrosarcoma of the head and neck in pediatric patients: a clinicopathologic study of 14 cases with a review of the literature. Cancer: Interdisciplinary International Journal of the American Cancer Society. 2000;88(9):2181-8.
- [3] Pierz K, Stieber J, Kusumi K, Dormans JP. Hereditary multiple exostoses: one center's experience and review of etiology. Clinical Orthopaedics and Related Research[®]. 2002;401:49-59.
- [4] Murphey MD, Choi JJ, Kransdorf MJ, Flemming DJ, Gannon FH. Imaging of Osteochondroma: Variants and Complications with Radiologic-Pathologic Correlation. RadioGraphics. 2000;20(5):1407-34.
- [5] Bernard SA, Murphey MD, Flemming DJ, Kransdorf MJ. Improved differentiation of benign osteochondromas from secondary chondrosarcomas with standardized measurement of cartilage cap at CT and MR imaging. Radiology. 2010;255(3):857-65.
- [6] Weinschenk RC, Wang W-L, Lewis VO. Chondrosarcoma. J Am Acad Orthop Surg. 2021;29(13):553-62.
- [7] Moradi Tabriz H, Obohat M, Vahedifard F, Eftekharjavadi A. Survey of Mast Cell Density in Transitional Cell Carcinoma. Iran J Pathol. 2021;16(2):119-27.

- [8] Salari S, Ghadyani M, Karimi M, Mortezazadeh M, Vahedifard F. Immunohistochemical Expression Pattern of MLH1, MSH2, MSH6, and PMS2 in Tumor Specimen of Iranian Gastric Carcinoma Patients. Journal of Gastrointestinal Cancer. 2022;53(1):192-6.
- [9] Nakagawa SA, Lopes A, De Carvalho AL, Rossi BM, Da Cunha IW, Soares FA, et al. Nitric oxide synthases, cyclooxygenase-2, nitrotyrosine, and angiogenesis in chondrosarcoma and their relation to prognosis. JBJS. 2010;92(8):1738-46.
- [10] Schrage YM, Machado I, Meijer D, Briaire-de Bruijn I, van den Akker BE, Taminiau AH, et al. COX-2 expression in chondrosarcoma: a role for celecoxib treatment? European journal of cancer. 2010;46(3):616-24.
- [11] Bani MA, Laabidi B, Gargouri F, Mansouri N, Bouziani A, Msakni I. Secondary chondrosarcoma arising in solitary sacro-iliac osteochondroma: a case report. La Tunisie medicale. 2017;95(5):386-7.
- [12] Chakravarthy K, Golovac S, Vahedifard F. Future Indications. In: Deer TR, Azeem N, editors. Essentials of Radiofrequency Ablation of the Spine and Joints. Cham: Springer International Publishing; 2021. p. 241-81.
- [13] Martel J, Bueno A, Nieto-Morales ML, Ortiz EJ. Osteoid osteoma of the spine: CT-guided monopolar radiofrequency ablation. European journal of radiology. 2009;71(3):564-9.
- [14] Vahedifard F, Malinowski M, Chakravarthy K. Mechanism of Action of Radiofrequency Ablation. In: Deer TR, Azeem N, editors. Essentials of Radiofrequency Ablation of the Spine and Joints. Cham: Springer International Publishing; 2021. p. 7-29.
- [15] Lin PP, Moussallem CD, Deavers MT. Secondary chondrosarcoma. J Am Acad Orthop Surg. 2010;18(10):608-15.
- [16] Wahyudi M, Astoguno Bayu Prakurso A. Gigantic secondary pelvic chondrosarcomas treated with pelvic resection type I and III: A case report. International Journal of Surgery Case Reports. 2020;75:327-32.
- [17] Tsuda Y, Gregory JJ, Fujiwara T, Abudu S. Secondary chondrosarcoma arising from osteochondroma. The Bone & Joint Journal. 2019;101-B(10):1313-20.
- [18] Righi A, Pacheco M, Cocchi S, Asioli S, Gambarotti M, Donati DM, et al. Secondary peripheral chondrosarcoma arising in solitary osteochondroma: variables influencing prognosis and survival. Orphanet Journal of Rare Diseases. 2022;17(1):74.
- [19] Ahmed A, Tan T-S, Unni K, Collins M, Wenger D, Sim F. Secondary Chondrosarcoma in Osteochondroma: Report of 107 Patients. Clinical orthopaedics and related research. 2003;411:193-206.