

Chondromyxoid fibroma of the pubic ramus: a case report and literature review

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Chondromyxoid fibromas (CMF) are benign cartilaginous bone tumors which are found most frequently in the metaphyses of long bones. They comprise less than 1% of primary bone neoplasms. We report an interesting incidental case of a 43-year-old woman with a CMF of the left pubic ramus, presenting with complaints of gradual onset of left groin pain over a period of 2 years. According to radiological examination, a malign chondroid bone tumor was excluded, and histopathological examination confirmed the diagnosis of CMF. The patient underwent aggressive curettage and bone grafting 6 years ago. Pelvic bones are encountered as rare localizations for CMFs. Pubic ramus is accepted as an exceptional site for this benign bone tumor of cartilaginous origin. To our knowledge, no any other CMF case in this localization has been reported in the literature. In atypical regions such as the pelvis and pubic ramus, CMF must be considered for differential diagnosis of malign tumors.

Keywords: Benign cartilaginous tumors; chondromyxoid fibroma; computed tomography; magnetic resonance imaging; pelvic tumors; pubic ramus; radiographs.

Chondromyxoid fibroma (CMF) was first described in 1948 by Jaffe and Lichtenstein^[1] as a benign cartilaginous bone tumor. Since then, CMFs have accounted for fewer than 1% of all primary bone tumors and are currently the rarest reported neoplasm of cartilaginous origin.^[2–4] Pathologic examination shows lobulated areas of spindle-shaped or stellate cells with abundant myxoid or chondroid intercellular material.^[5–7] Radiographic appearance of CMF varies but typically looks like well-circumscribed, eccentric geographic pattern of bone destruction with a round and ovoid shaped.^[1,8] The tumor is more common in males than females (2:1) and is located mostly in the metaphyseal areas of the lower extremity,^[9,10] although the tumor is found in flat bones, such as the iliac bone, in up to 25% of cases.^[11]

There have been sporadic reports of CMF in other anatomical sites. In decreasing order of frequency, CMF has been reported in the hands, feet, vertebrae, and ribs. Pubic ramus involvement reported in our case presentation appears to be a unique site of involvement for this rare tumor.

Case report

A 43-year-old woman was referred to our clinic with a history of left groin pain, of which she had experienced a gradual onset 2 years prior and which had intensified in the last 3 months; a slight limping on her left side related to the pain was also present. Written informed consent was obtained from the patient for publication of this case report and any accompanying images. On

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physical examination, a slight limitation of active hip motions on the left side and a tender firm mass was seen on deep palpation over the left groin area. Clinical examination was otherwise normal. Pelvic radiograph of the patient showed a fusiform shaped lobulated mass in the left superior pubic ramus bone. This radiolucent septated chondroid lesion had a thin sclerotic rim surrounding the peripheral part of the lesion and extending through the edge of the symphysis pubis (Figure 1a, b).

Computed tomography (CT) studies showed scattered calcifications and septations in this expansile irregular radiolucent lesion. Bony destruction was noted at the superior edge of the tumor, and soft tissue extension was present (Figure 1c). Magnetic resonance imaging (MRI) showed a well-defined expansile bone lesion which was hypointense in T1- and hyperintense in T2-weighted images, with heterogeneous uptake of contrast material (Figures 2a, b). The presence of bone destruction and uptake of contrast material on MRI gave rise

to suspicion of a malignant cartilaginous bone tumor; however, CMF could not be ruled out. MRI also demonstrated cystic cavities and nodularity in sagittal FS-T1-weighted images (Figure 2c).

An open incisional biopsy was performed. Pathologic examination showed a lesion with stellate or spindle-shaped cells in a myxoid background (Figure 3a). There was a lobular pattern with hypocellular centers and hypercellular peripheries (Figure 3b). No atypical mitoses or necrosis were detected. Immunohistochemically, the cells were stained with S100 focally. A diagnosis of CMF was confirmed. A subsequent marginal curettage of the lesion with removal of the small mass of soft tissue extension was performed in the destructed part of the superior pole of the pubic bone. The walls of the cavity were curetted with a high-speed burr and destructed with the help of electrocautery as adjuvants. Figure 1b shows anteroposterior pelvic radiography demonstrating the lesion after a thorough curettage and chip bone grafting after the operation.



Fig. 1. (a) Preoperative anteroposterior pelvic radiograph of the patient; a fusiform-shaped lobulated mass in the left superior pubic ramus bone. (b) Anteroposterior pelvic radiograph demonstrating the lesion after thorough curettage and chip bone grafting after the operation. (c) Computed tomography sample of the patient; studies showed that this expansile irregular radiolucent lesion showed scattered calcifications and septations.

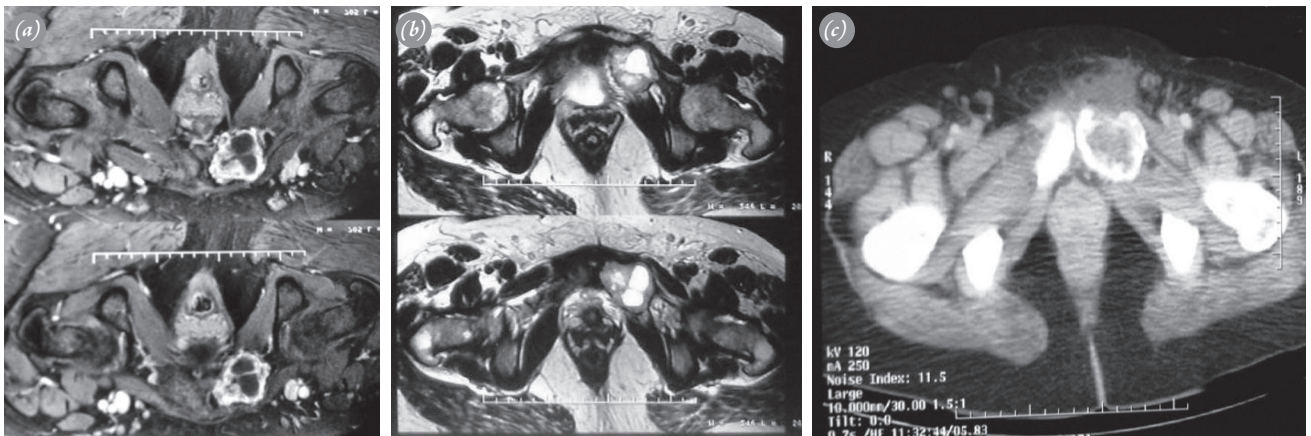


Fig. 2. (a) Magnetic resonance imaging showed a well-defined expansile bone lesion which was hypointense in T1-weighted images, with heterogeneous uptake of contrast material. (b) Magnetic resonance imaging showed a well-defined expansile bone lesion which was hyperintense in T2-weighted images, with heterogeneous uptake of contrast material. (c) Magnetic resonance imaging demonstrated cystic cavities and nodularity in sagittal FS-T1-weighted image.

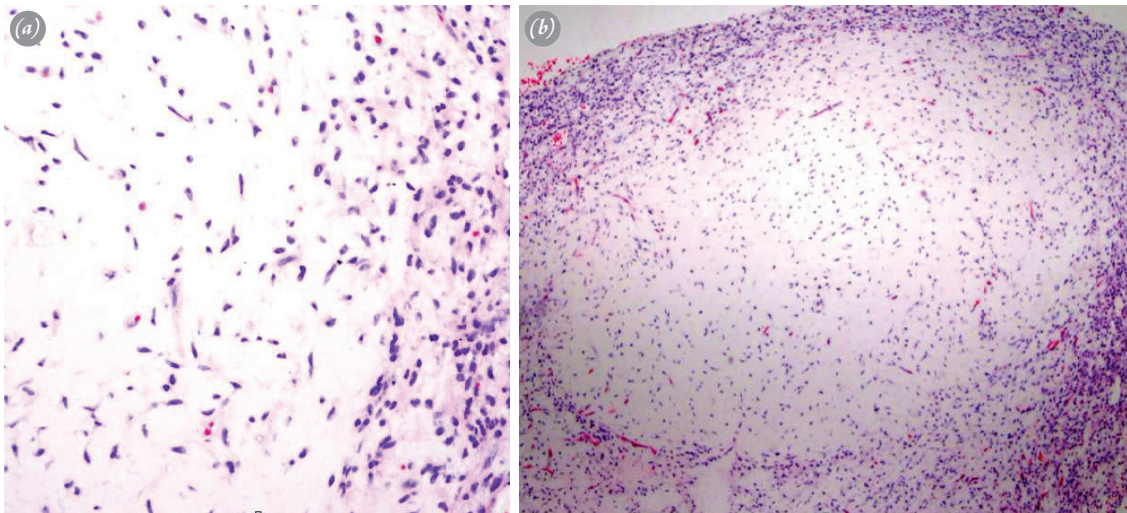


Fig. 3. (a) Pathologic examination showed a lesion with stellate or spindle-shaped cells in a myxoid background on 200x magnification. (b) Pathologic examination showed a lobular pattern with hypocellular centers and hypercellular peripheries on 100x magnification. [Color figures can be viewed in the online issue, which is available at www.aott.org.tr]

The patient was free of disease and experienced no pain at 3-month follow-up. Figure 4a shows radiograph of the pelvis at 2-year follow-up, and Figure 4b shows 6-year follow-up; no local recurrence has been noted.

Discussion

Geographic bone destruction with well-defined sclerotic margin and cortical expansion of the bone are characteristic findings of the site of origin.^[1,2,12] The disease preferentially occurs in young patients during the second or third decades of life.^[13] The classic site of involvement

is the metaphyseal region of long tubular bones, usually presenting in the second to third decades of life with nonspecific pain and swelling of the affected part.^[14] The most usual localization is the metaphysis of long tubular bones, especially at the tibia, and 50% are located in the region surrounding the knee.^[15] The pelvis and distal femur are other common locations.

Clinical presentation is usually chronic pain (85%), swelling (65%), restriction of motion, and, less frequently, pathological fracture.^[7,13] In our case presentation, a tender firm mass was noticed in deep palpation over the left groin area, and a limping and restriction of motion

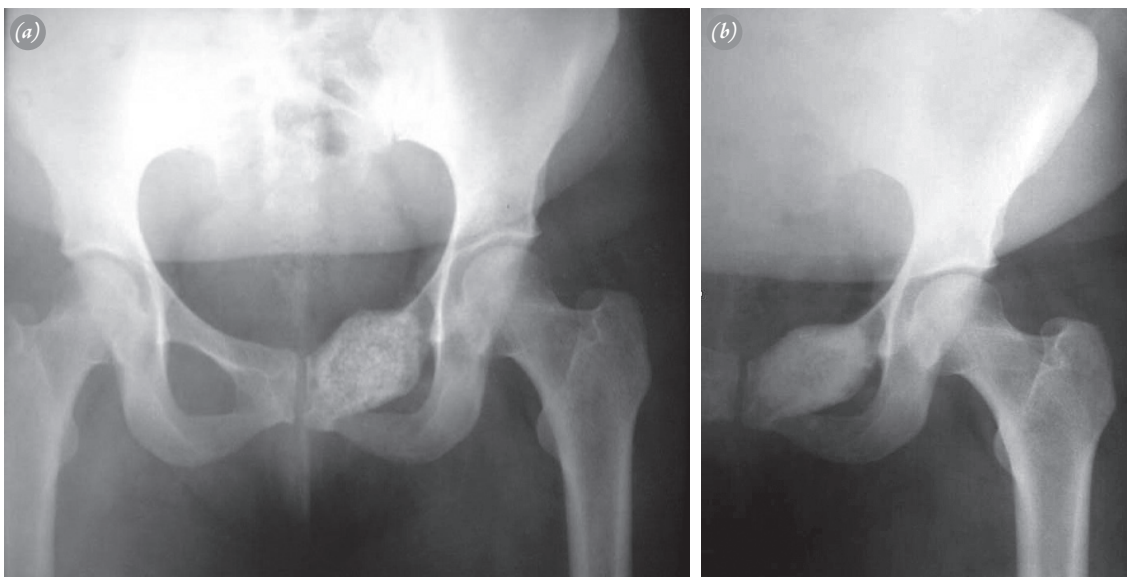


Fig. 4. (a) Anteroposterior radiograph of the pelvis at 2-year follow-up, with no recurrence. (b) Anteroposterior radiograph of the pelvis at 6-year follow-up with, no recurrence.

in the left hip joint, occasionally related to pain, were additional clinical presentation signs.

Radiologically, CMF appears as an eccentric well-defined single slightly expansile and lytic lesion with endosteal sclerosis.^[1,15] The fragmentation of the bone cortex is common, as it appeared in our case in the superior portion of the pubic bone, both in CT scans and direct roentgenograms. Periosteal reactions, when present, are thought to largely be the result of pathologic fracture, although this is not universally true.^[14,16] CT helps in defining cortical integrity and in confirming that there is no mineralization of the matrix, in contrast to other cartilage tumors. MRI shows decreased signal on T1-weighted images and increased signal on T2-weighted images.^[7,13]

Microscopically, CMF is usually well-demarcated, even in instances when it erodes the bone. The lesions are nodular, with myxoid lobules. Multinucleated giant cells, blood vessels, and collagenization are found at the lobular peripheries.^[1,13]

Differential diagnosis includes chondrosarcoma, chondroblastoma, nonossifying fibroma, enchondroma, unicameral bone cyst, giant cell tumor of bone, and aneurysmal bone cyst.^[8,17] Chondrosarcoma is the major item of differential diagnosis, as both entities have similar radiologic and pathological features. Cartilage cell-containing tumors of the pelvis are responsible for one-quarter of all primary bone lesions in this site. Benign tumors are uncommon but when present have classical radiological appearances similar to those described elsewhere. Only the rarest, CMF, may cause difficulty in diagnosis.^[18] Curettage alone is associated with a higher recurrence rate, but if performed thoroughly, curettage combined with bone grafting closely approaches the nearly zero recurrence rate of en bloc resection.^[19–22] Overall recurrence rate of 23% is reported by some authors in the literature, using curettage combined with bone grafting or application of PMMA.^[1,6,19] However, other authors report a much lower rate of 4% when curettage is combined with corticocancellous bone grafting.^[3,4,19,22] Thus, curettage combined with bone grafting seems to be an appreciable alternative treatment to resection. As CMF is well known for its tendency for regional expansion and usual recurrence after curettage of the lesion, we used high-speed burr and burned the walls of the cavity with electrocautery in order to reduce the risk of recurrence. Radiation therapy, because of its oncogenic potential, is contraindicated.

Even though it is reported that resection should be performed for large lesions or in anatomic sites such as the pelvis or the proximal fibula where such a procedure

does not increase the risk of fracture and reduce the likelihood of requiring a second operation, we preferred aggressive curettage and chip bone grafting in our case, as it is a safe, easy, and less morbid procedure for this anatomic site.^[9,23,24] In addition, good results with a combination of curettage and bone grafting encouraged us in this procedure.

CMF in pubic ramus is an exceptional anatomical localization, and, to our knowledge, this is the first case reported in the English literature.

Conflicts of Interest: No conflicts declared.

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