Original Article

Chondromyxoid Fibroma of Pelvis, Surgical Management of 8 Cases

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Abstract

Background: Chondromyxoid fibroma is a rare benign primary bone tumor of cartilaginous origin, which most commonly involves the metaphyseal bone of proximal tibia and distal femur. The purpose of the study is to report our experience with diagnosis and surgical management of Chondromyxoid fibroma in the pelvic region.

Methods: Eight consecutive patients with a final diagnosis of pelvic Chondromyxoid fibroma were treated from 2001 to 2010. We considered the presentations and outcome for surgical complications and local recurrence after extended curettage and allogenic corticocancellous bone grafting.

Results: Three patients were female and five were male. The median follow up period was 72 (30–126) months. The mean age of cases was 31.9 (20–41) years. Five patients had left side involvement and in the remaining three, the right side was involved. Four involved periacetabulum, two involved the ilium and the remaining two cases were ischiopubic. The mean Musculoskeletal Tumor Society Score was 94.1%. The major complications were recurrence in one case and herniation after pubic rami resection in another case.

Conclusion: Chondromyxoid fibroma should be distinguished from chondrosarcoma. Management recommendation includes extensive curettage and corticocancellous bone grafting. We also advocate use of fibular strut allograft for reconstruction of pubic rami after its resection to prevent hernia in cases with pubic rami involvement.

Level of evidence- IV

Keywords: Chondromyxoid fibroma, complication, fibular graft, pelvic tumor

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Introduction

hondromyxoid fibroma (CMF) is a rare benign primary bone tumor composed of immature myxoid mesenchymal and cartilaginous tissue. It comprises less than 1% of all tumoral and tumoral-like lesions of the bone.1 Although it has been reported in almost all bones, review of the literature reveals that CMF most commonly involves the metaphyseal area of long bones of lower extremities, especially proximal tibia and distal femur. However, this rare tumor has also been reported in small tubular bones of the feet, the pelvic flat bones and even in the rare locations like calcaneus.^{2,3} The proposed treatment for CMF is mainly curettage with or without bone grafting or cementing which can result in 20%–25% recurrence rate.⁴ Younger age, curettage without bone grafting and presence of pleomorphic nuclei and predominant myxoid tissue on pathology are reported to be associated with higher rates of local recurrences.5 To the best of our knowledge, there is not any case series in term of presentation and surgical results of CMF in a particular area of bone such as pelvis. We report eight cases of CMFs along with their presentations, treatment options, local recurrence and complications.

Materials and Methods

Institutional Review Board of our center approved the study. The

study was conducted at Shafa Yahyaeian hospital, Tehran, Iran. We studied medical records from 2001 to 2010 in our hospital and identified 10 patients with the primary diagnosis of CMF of the pelvic bone. After re-examination of pathologic slides, two cases were excluded because their final diagnosis was chondrosarcoma. The primary diagnosis of CMF in these excluded cases had been made by core needle biopsy. In histologic examination, the classic CMF has spindle-shaped cells arranged in lobules in a chondroid and myxoid matrix, with osteoclast-like giant cells (Figure 1).

We reviewed physical examination, laboratory studies, chest and pelvic radiographs, pelvic CT scan and MRI of all our patients. Six of our cases had open incisional biopsy and the remaining two cases had CT guided core needle biopsy. We classified the location of the lesion based on the system proposed by Enneking WF and Dunham WK.⁶ Based on this classification, Zone 1 denotes iliac bone involvement, Zone 2 refers to periacetabular area, and finally zone 3 marks the involvement of the pubis and ischium bones.⁶

In two of our cases (Cases 1&2) with iliac bone involvement (zone: 1), we performed resection of the lesion including the biopsy tract and 5 mm of safe margin by oscillating saw. These patients did not need pelvic reconstruction because the pelvic ring was intact.

In cases with periacetabular involvement (cases 3, 4, 5 and 6; zone 2), a large window was opened over the softest part of the lesion. After curettage of the tumor, at least 5 mm of the wall of the cavity was removed by high speed dental burring except for the cartilage of acetabulum. The cavity was filled and impacted with cancellous allograft. For cases no.4 and 5 with the extension of the tumor from zone 2 to the ischial bone, we used fibular allograft to

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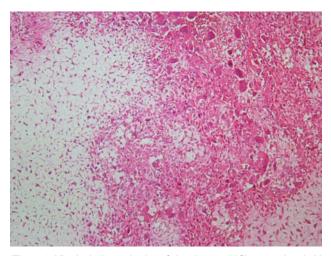


Figure 1. Histological examination of chondromyxoid fibroma : chondroid and myxoid matrix, with osteoclast-like giant cells (H&E, 40^*)



Figure 2. Anteroposterior radiograph of the pelvis of a 35-year-old man, 2 years after surgery showing well incorporated cancellous and fibular allograft (Case 4).



Figure 3. The 3-year post-operative pelvic radiography showing stable pelvic ring (Case 7).

augment the ischium (Figure 2). In case 6 with periacetabular and pubic rami involvement, in addition to extensive curettage of periacetabulum and filling of the void space with cancellous allograft, reconstruction with fibular strut allograft was done after resection of involved pubic rami. The allograft was fresh frozen and taken from the bone bank of our hospital. In patients with periacetabular involvement, we limited weight bearing for the involved side until union and consolidation were seen in plain radiographs.

For case 7, with the involvement of the superior ramus of left pubis, resection of superior ramus was done without grafting. This case was complicated by pelvic internal organs hernia through the window, which was created by ramus resection (Figure 3). The patient declined to undergo surgical intervention to solve the complication.

In case no 8 with the involvement of right superior pubic rami after resection, reconstruction was done with fibular strut allograft to prevent pelvic internal organs herniation. The patients were visited every three months for clinical and radiographic evaluation in the first year after surgery and every six months afterwards. Clinical examination included assessment of the hip range of motion, the gait and pain using a visual analogue scale (0, no pain;

368 Archives of Iranian Medicine, Volume 18, Number 6, June 2015

10, intolerable pain). Anteroposterior radiograph of pelvis was obtained and reviewed for signs of tumor recurrence, incorporation of the graft and sign of hip degenerative joint disease. At the latest follow-up, all patients were assessed using Musculoskeletal Tumor Society Score (MSTS).

According to the MSTS system, numerical values (0 to 5) are given to each of the six categories of pain, function, emotional acceptance, supports, walking and gait. The total score for the system between 0 and 30 is given to each patient with 0 indicating poor and 30 indicating good functional results.⁷

Results

Of eight cases with definite diagnosis of CMF, three patients were female and the remaining five patients were male. The mean age of cases was 31.9 (20–41) years (Table 1). Five patients had left side involvement and in the remaining three, the right side was involved. Pain was the primary chief complaint in all our cases. All laboratory tests in our patients were in normal range. The appearance of the tumor in plain pelvic radiography was a lytic lesion of variable size and well defined by a narrow rim of sclerosis.

Case	Sex	Age	Location	Treatment	Hernia	Recurrence	Follow up (Months)	MSTS score
1	М	30	Rt ilium (zone I)	Resection	Ν	Ν	120	28 (93.3%)
2	М	36	Lt ilium (zone I)	Resection	Ν	Ν	114	27 (90%)
3	М	34	Lt periacetabulum (zone II)	Curettage & cancellous allograft	Ν	Ν	96	30 (100%)
4	F	35	Lt periacetabulum to ischium (zone II)	Curettage & corticocancellous allograft	Ν	Ν	36	30 (100%)
5	F	31	Lt periacetabulum to ischium (zone II)	Curettage & corticocancellous allograft	Ν	Ν	30	30 (100%)
6	F	28	Rt periacetabulum to pubis (zone II)	Curettage & corticocancellous allograft	Ν	Y	42	25 (83%)
7	М	20	Lt sup rami of pubis (zone III)	Resection	Y	Ν	126	26 (86%)
8	М	41	Rt superior ramus of pubis (zone III)	Resection & fibular strut graft	Ν	Ν	48	30 (100%)
Rt=Right Lt=Left Y=Yes N=No MSTS =Musculoskeletal Tumor Society								

Table 1. Demographic data, complications and treatment results of patients with pelvic chondromyxoid fibroma.



Figure 4. Anteroposterior radiograph of the pelvis of a 41-year-old man, 18 months after surgery showing union of the fibular allograft (Case 8).

Expansion of the bone was seen in some areas. We used CT scan to determine bone destruction and calcification. CT scans of the involved areas revealed lytic lesions and all the lesions showed an area of cortical disruption with soft tissue component and all had a partial rim of bone over the lesion. Fine popcorn calcification was seen in two cases. MRI could precisely show the dimensions and extension of the tumor.

Whole body bone scans of all patients indicated increased uptake in involved areas of the pelvic bone.

The lesions were staged according to the system of the Musculoskeletal Tumor Society.⁷ Stage 3 refers to aggressive, benign lesions. With the use of these criteria, all patients had a stage-3 lesion. The mean follow up for cases was 72 (30–126) months (Table 1). MSTS score was 28.25.

One of our patients (Case 7) had a complication and developed direct hernia of the lower abdomen through the defect that resulted from the pubic rami resection. This patient refused any further treatment for this complication. To avoid this complication, for future patients with pubic rami involvement or periacetabular tumor with extension of the CMF to the pubic rami, we used curettage of the periacetabulum, resection of the pubic rami and bone grafting with augmentation by fibular strut allograft. In these patients, the operation ended in solid union without herniation (Figure 4).

The major complications in our cases were herniation in case 7 and also a recurrence in case 6 that was treated with curettage and bone grafting (Table 1).

Discussion

Jaffe and Lichtenstei first introduced CMF in 1948.⁸ Since then, it has been documented in almost all bones. CMF appears most frequently in the 2nd and 3rd decades of life with a male predilection.⁹ The majority of our patients were in the fourth decade of their life or older and with regard to age distribution, our cases were older than what was described in literature for CMF. The mean age of our patients was 31.8 years. There was also a male predominance in our cases. The history of trauma was negative and there were no pathologic fractures in our patients.

When it comes to the location of our cases, periacetabular area (zone 2) was the predominantly involved site in our patients; iliac wing and superior ramus were in the second most frequent sites of involvement.

In terms of symptoms of CMF, chronic pain is generally the main manifestation.¹⁰ This was true for our cases, where the patients' chief complaints were chronic pain.

CMF is a benign tumor and there have been no reports of metastases.⁸ All of our patients were in stage 3 and locally destructive. The common treatment options for CMF are curettage, curettage with cementing, curettage with bone grafting, and en bloc excision. The lowest chance for recurrence has been reported with resection and en block excision.⁴ This was compatible with our findings. We performed en block excision in cases with involvement of zones 1 and 3 and we did not observe any local recurrence. We had one local recurrence in one of our cases with the involvement of zone 2, which was treated with extensive curettage and bone grafting. We cannot compare recurrence rate of our series with reported results in the literature due to the small number of our patients.⁴

Internal pelvic organs herniation, which had occurred in one of our patients, is a unique complication. This occurred in a patient, who underwent pubic rami resection without reconstruction of bony defect. In the following case with pubic rami resection, we used fibular strut graft to reconstruct the defect. The complication did not occur in this patient.

Other tumor and tumor like lesions in this age group, which should be considered as differential diagnoses for CMF, include aneurysmal bone cyst (ABC), giant cell tumor (GCT), chondroblastoma, fibrous dysplasia, chondrosarcoma and osteosarcoma. These tumors and tumor like lesions can be recognized histologically.⁴

In pelvis, chondrosarcoma is most commonly mistaken for CMF. Microscopically, large nuclei are present in about one third of CMF cases. On the other hand, chondrosarcoma may have prominently myxoid areas. In the two patients who were excluded from this study, the primary diagnosis of CMF was made by core

needle biopsy but the final diagnosis was chondrosarcoma. This misdiagnosis has been reported previously in the literature.⁸

CMFs of the pelvis are aggressive lesions and should be distinguished from chondrosarcoma, especially when core needle biopsy has been used for tissue diagnosis. Management recommendations include extensive curettage and corticocancellous bone grafting. We also advocate use of fibular strut allograft for reconstruction of pubic rami after its resection to prevent hernia in cases with pubic rami involvement.

Conflict of interest: None

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