Palliative Approach in Advanced Pelvic Osteosarcoma: A Single Centre Experience of a Rare Disease

Ritesh Kumar, Rakesh Kapoor, Divya Khosla, Narendra Kumar, Pramod Kumar Singh, Mahendra Kumar[1], SC Sharma

Departments of Radiotherapy and Oncology, [1]Pathology, Postgraduate Institute of Medical Education and Research, Chandigarh, India

Address for correspondence: *Dr. Ritesh Kumar; E-mail: riteshkr9@gmail.com*

ABSTRACT

Background: Pelvic osteosarcoma is a rare and aggressive malignant neoplasm with poor outcomes. It represents only 5% of all osteosarcomas. The authors present our institute's experience in management and outcomes of five successive patients of pelvic osteosarcoma.

Materials and Methods: We retrospectively reviewed five patients of pelvis osteosarcoma treated in our institute from September 2008 to December 2010. Clinical characteristics and treatment (CCT) modality in form of surgery and chemotherapy were noted. Statistical analysis was done with regards to progression-free survival (PFS) using Kaplan-Meier survival analysis.

Results: The median age of the patients was 16.0 years. The median duration of symptoms was 9 months. One patient had lung metastases at presentation. All patients received systemic chemotherapy. One patient underwent surgery in the form of limb sparing approach. Three patients had partial response to treatment, one had complete response, and one had progressive disease. Median duration of PFS was 7 months only.

Conclusions: Pelvic osteosarcomas are rare neoplasms with aggressive growth patterns. Survival results are poor in view of advanced stage of presentation and difficult surgical approaches. The combined modality approach is needed to improve the results.

Key words: Chemotherapy, Osteosarcoma, Pelvic

INTRODUCTION

Pelvic osteosarcomas are rare malignant neoplasms comprising only 5% of all osteosarcomas.[1,2] Pelvic osteosarcomas have poor prognosis as they are often diagnosed in an advanced stage with a large tumor size.[3] The management of pelvic osteosarcoma is challenging because it is difficult to achieve a complete surgical excision that results in local recurrence and progression.[4] With the use of multiagent chemotherapy (CCT) and improvement in surgical techniques, the results are improving as compared to historical controls.[5,6] Although there are few reports on additional radiotherapy (RT) after tumor excision with inadequate margins, the effect of RT for local tumor control remains unknown.[7,8] We herein report our institutional experience of five successive patients of pelvic osteosarcomas being treated from September 2008 to December 2010.

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MATERIALS AND METHODS

Patient population and initial evaluation

We retrospectively reviewed the patients of pelvic osteosarcomas from September 2008 to December 2010 treated in our institute. The total number of patients were 5. We reviewed the records of these patients to extract the following information: Age, sex,

clinical symptoms, histology, radiology (CT/MRI), tumor extent, and extent of surgical resection, chemotherapy (regimen, number of cycles), toxicity, response, progression, metastases, and death. Metastatic workup for distant metastases included CECT of the chest, abdomen, and pelvis and bone scintigraphy. Laboratory studies included blood chemistry (electrolytes, liver, and kidney function tests), and a complete blood count was performed.

Pathological review and staging

Pathology reports were obtained for all patients and the tumors were classified according to the histopathological subtype. Staging was based on the surgical, radiological, and pathological criteria as per the TNM staging system.

Treatment

Surgery and CCT was used in the treatment. Systemic CCT was administered with a regimen comprising cisplatin and adriamycin 3 weekly for 3-6 cycles. Only one patient underwent primary surgery in form of local excision of the residual tumor after systemic CCT. The median number of CCT cycles was 6.

Follow up

The period between the first complaint and diagnosis was registered as symptom duration. Survival, recurrence, and progression information were collected through chart review, patient, or relative contact. Response evaluation was noted both clinically and radiologically and response evaluation criteria in solid tumors (RECIST) were applied.[9]

Statistical analysis

SPSS v 15 was used for statistical analysis. The Kaplan-Meier survival analysis was performed for analyzing progression-free survival (PFS).[10]

RESULTS

Patient characteristics

Patient characteristics are summarized in Table 1. Between September 2008 and December 2010, five patients of pelvic osteosarcoma were registered in our department. The median age of the patients was 16.0 years and ranges from 5 years to 38 years. The median duration of symptoms was 9 months. Pain in the pelvis was the most common symptom followed by the awareness of swelling and neuropathic symptoms. All the patients had locally advanced tumors with median size of 10 cm that ranged from 6 to 12 cm. On histopathological analysis, all patients had classical osteosarcoma histology. On metastatic workup, one patient had lung metastases.

Clinical outcomes

After treatment completion, patients were assessed for response both clinically and radiologically. One patient was asymptomatic, three patients had significant improvement in symptoms, and one patient had no improvement in symptoms. As per the RECIST criteria, one patient had complete response (CR), three patients had partial response (PR), and one patient had progressive disease (PD). Palliative RT to the local site was delivered to the four patients who did not undergo surgery. Symptomatic treatment in form of analgesics as per WHO ladder was prescribed. The median duration of follow-up was 8 months. Median duration of PFS was 7 months and mean duration of PFS was 8 months.

Treatment toxicity and compliance

There were no surgical complications in the one patient who underwent surgery in form of local excision. CCT toxicity was seen in three patients in form of grade 1-2 hematological toxicity.

DISCUSSION

Pelvic osteosarcoma is rare and consists only 5% of all osteosarcomas.[1,2] Pelvic osteosarcomas are regarded as a highly malignant tumor with frequent recurrence and poor survival.[4] Spread of the tumor is mostly by local extension but hematogeneous spread does occur, most commonly to the lungs and liver. They are usually present in locally advanced stages in view of vague initial symptoms. The optimal management of pelvic osteosarcomas remains unclear because of the rarity of the disease in adults. Treatment strategies reviewed in the literature include local surgical excision, systemic chemotherapy (CCT), and radiotherapy (RT).[6,11]

In general, the treatment of extremity osteosarcoma involves excision of the tumor with an adequate surgical margin after systemic CCT. Chemotherapeutic agents commonly used are cisplatin, adriamycin, methotrexate cyclophosphamide, and ifosfamide.[12,13] The regimen used in the patients in our series constituted cisplatin and adriamycin. However, in pelvic osteosarcomas, the tumor volumes changes little because of poor response to CCT. Surgery is also difficult and associated with marked morbidity. The surgical approaches involved ranges

Table 1: Patient characteristics, treatment details and outcome

Age	Sex	Site	Size	Metastases	Surgery	CCT cycles	Clinical response	Radiological response
16	M	Sacrum	10	Nil	No	3	Improved	PR
14	F	Ilium	12	Nil	No	6	Improved	PR
5	M	Ilium	6	Nil	No	4	No improvement	PD
35	M	Ilium	12	Lungs	No	6	Improved	PR
38	F	Ilium	10	Nil	Local excision	6	Asymptomatic	CR

CCT: Combination chemotherapy, PR: Partial response, CR: Complete response, PD: Progressive disease

from hemipelvectomy to limb sparing approaches.[14,15] The European Cooperative Osteosarcoma Study Group reported 67 patients of pelvic osteosarcomas with a median age of 20 years and range up to 63 years.[13] Of these, 50 underwent surgical resection, 38 limb-sparing and 12 hemipelvectomies. Fifty percent of patients who had resections had positive residual disease. The overall 5-year survival was only 27%. One of the patient underwent surgery in form of local excision (limb sparing approach) in our series.

In 1998, Kawai *et al*. reported a cohort of 40 patients with pelvic osteosarcoma with a median age of 25 years and range up to 76-year old.[6] The 5-year OS for the whole group was 34%, with a 10% 5-year survival for patients with unresectable tumors and 41% 5-year survival for patients with resections. Fuchs *et al*. reported the OS for patients with pelvic osteosarcoma treated during a 20-year period.[16] In their cohort, the mean age was 34 years with an upper limit of 66 years. They found that 13 of 30 patients did not have an adequate tumor-free margin and the 5-year OS was only 38%. The survival in our series was poor and all the patients in our series presented in locally advanced stages and one patient had lung metastases at presentation. Median duration of PFS was 7 months and mean duration of PFS was 8 months in our series.

Pelvic site itself confers poor prognosis in osteosarcoma.[17] Other poor prognostic factors reported in the literature are large tumor size, metastases at presentation, and no or intralesional surgery.[17-19] Alternate therapies, including the use of standard external beam or proton radiation therapy, could be considered for patients with tumors that are not completely resected or unresectable.[7,8] Hernberg *et al*. reported the role of chemoradiotherapy (CRT) in inoperable nonextremity osteosarcomas.[20] RT relieved symptoms efficiently and five of the seven patients were alive 5 years after treatment. With a lack of randomized trials assessing these modalities in patients with unresectable osteosarcoma, a definitive recommendation is still unclear.

Overall, pelvic osteosarcoma is a rare and aggressive neoplasm with poor prognosis. Improvements in surgical technique may lead to improvement in prognosis, although many patients still will not have tumors that can be resected. Improved surgical or medical therapy is needed, and patients with pelvic osteosarcoma may warrant alternate or experimental therapy. Palliative RT and supportive care remains an important component in the management of locally advanced pelvic osteosarcoma.

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