

An Unusual Presentation of Low-grade Chondrosarcoma of the First Metatarsal Bone requiring Extensive Resection: A Case Report

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ABSTRACT

Chondrosarcoma (CS) is one of the common malignant bone tumours arising from chondrocytes with variable degrees of malignancy. It is mainly noted in flat bones like the pelvis and scapula, and sometimes in long bones like the proximal femur. This case report describes a 40-year-old female who presented with pain over her right great toe for six months and swelling for four months. An open biopsy revealed low-grade CS. Subsequently, the patient underwent amputation of the first ray till the tarsometatarsal joint with meticulous dissection of the tumour mass. At eight years of follow-up, the patient had no recurrence and no functional disability. Therefore, in cases of low-grade CS, first ray amputation along with excision of the tumour mass helps avoid recurrence and metastasis, providing a satisfactory functional outcome.

Keywords: Low-grade sarcoma, Ray amputation, Recurrence

CASE REPORT

A 40-year-old female reported to the orthopaedic outpatient department with complaints of pain over her right great toe for six months and swelling for four months. The pain started intermittently and then progressed nocturnally and was also associated with swelling that had restricted her ability to walk. There was no history of swelling elsewhere and no symptoms suggestive of gout or any other infection, and she denied a history of any chronic medical illness in the past. On examination, there was localised swelling on the medial aspect of the dorsum of the right first metatarsal shaft. No evidence of acute inflammation was seen [Table/Fig-1]. The swelling was variable in consistency with tenderness on deep palpation. The margins were well-defined with preserved neurovascular status and intact toe movements. The X-ray revealed an expansile centric lytic lesion in the body of the first metatarsal not involving the tarsometatarsal joint [Table/Fig-2]. The lesion had a clear matrix and mottled calcification with a narrow zone of transition. There was

no evidence of cortical break, periosteal reaction, or pathological fracture. Further radiological investigation could not be carried out due to the poor financial status of the patient.

The patient underwent an open biopsy. Histopathological examination showed osseocartilaginous tissues with irregular lobules and high cellular fibrous tissue. Occasional lacunae containing low-density chondrocytes with deeply stained nuclei and binucleate and multinucleated cells were also seen, along with focal areas of infarction with mild cellular atypia. These findings were suggestive of low-grade CS. Other metastatic workups revealed no evidence of metastasis. CS is resistant to chemotherapy and radiotherapy. Subsequently, the patient underwent disarticulation of the first ray until the tarsometatarsal joint with meticulous en-bloc dissection of the tumour mass [Table/Fig-3,4]. The available peroneus longus and tibialis posterior tendinous portions were sutured together.



[Table/Fig-1]: Clinical image showing swollen dorsum of the right first metatarsal shaft area.

[Table/Fig-2]: Antero-posterior and oblique view X-ray of right foot showing expansile centric lytic lesion in the body of first metatarsal not involving the tarsometatarsal joint. (Images from left to right)

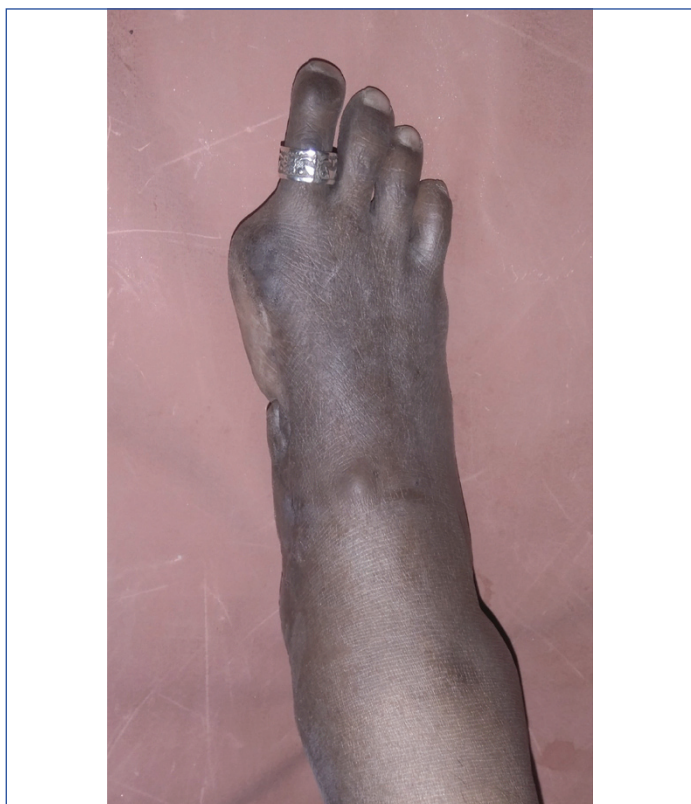


[Table/Fig-3]: Intraoperative image showing post En-bloc dissection and disarticulation of the first ray till tarsometatarsal joint.



[Table/Fig-4]: Resected gross specimen with tumour spread.

The patient was then followed-up serially for evidence of recurrence clinically and radiologically. At eight years of follow-up, the patient had no recurrence and no functional disability in her daily routine activities [Table/Fig-5].



[Table/Fig-5]: Eight years postoperative with no signs of recurrence.

DISCUSSION

The CS is one of the most common primary malignant bone neoplasms arising from chondrocytes. The condition can also be caused by pre-existing lesions such as Ollier's disease and Maffucci syndrome. Often noted in flat and long bones, this tumour is commonly seen in the age group of 40-70 years [1]. The reported incidence of all primary malignant bone tumours is 25% [2]. Hands and feet are uncommon sites for CS, whereas enchondromas are also common [3,4]. The prevalence of foot CS, as reported by Tsuda Y et al., is approximately 3.0% [5].

The patient presented with localised pain over the dorsum of the 1st metatarsal. This was the main distinguishing feature of CS compared to benign cartilaginous tumours; the pain increases with increasing tumour malignancy grade. Initially, the patient had mild intermittent pain, which became severe nocturnally due to the invasive growth of the tumour. The CS prognosis varies based on tumour grade. Histologically, it is divided into three grades: low, intermediate, and high [6]. Low-grade CS has an excellent prognosis with a 10-year survival rate of around 83%, unlike

intermediate and high-grade CS. As the tumour grade increases, cellular density increases, and cellular atypia and mitotic changes occur. Chondromyxoid fibromas mimic low-grade CS, with the main differentiating feature histologically being the presence of myxoid elements and the absence of soft-tissue involvement. In the present case, microscopic examination revealed low-density chondrocytes with deeply stained nuclei and occasional binucleate cell features, mild atypia, and no myxoid elements, suggesting low-grade CS.

Radiologically, CS presents as expansile central lytic lesions involving the medullary cavity with punctate intralesional calcifications [7]. These calcifications are typically described as a ring, an arc, or a popcorn calcification, better detected by Computed Tomography (CT) scan than X-ray [8]. Bone scans show high uptake from the lesion. Various treatment options have been described for low-grade CS, with surgical resection remaining the treatment of choice. Resection can be local, radical, or extensive. Local resection includes intralesional curettage and marginal resection [9]. Radical resection entails removing the tumour until the margins are microscopically negative.

Kask G et al., conducted a multicentric study on 810 patients with CS of the metatarsal bone. They showed that local recurrence occurs following intralesional curettage in low-grade CS [10] and suggested that extensive resection can reduce the chance of local recurrence [11]. In the present case, the patient underwent extensive resection of the tumour due to its proximity to the joint, despite it being a low-grade CS. Local resection is performed when the tumour is away from the joint without soft-tissue involvement. Low-grade CS is a locally aggressive tumour that rarely metastasises, with a reported risk of metastasis of 6% [12]. The patient underwent 1st ray amputation (extensive resection), and an eight-year postsurgery follow-up showed no local recurrence or metastasis. Despite this observation, no direct relationship between patient survival and local recurrence is mentioned in the literature [10].

CONCLUSION(S)

The CS is a malignant bone tumour arising from chondrocytes with variable malignant potential. Intralesional curettage is the most commonly performed surgery in low-grade tumours. Extensive resection of the tumour mass helps to avoid recurrence and metastasis, with no limitation of the patient's daily activities and good functional outcomes. Hence, extensive resection can be considered a treatment plan for low-grade CS of the metatarsals.

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