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Toe salvage procedure for the recurrent chondromyxoid fibroma

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CASE STUDY

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ABSTRACT

The treatment options for recurrent chondromyxoid fibroma of the toe range from total amputation to salvaging a functional toe. There is no globally accepted treatment protocol available for this tumour because of its rarer incidence and lack of population based data. Here we suggest performing a staged approach, which involves en block resection initially and maintenance of metatarsophalangeal space by using a kirshner wire with the bone cement. If there is no sign of malignancy in the histopathology, we recommend performing interposition arthroplasty at the metatarsophalangeal joint with the tricortical iliac crest graft. The kirshner wire should be kept which incorporates the iliac graft and the soft tissue, which is being interposed at the metatarsal head. This will cause pseudoarthrosis and also decreases the chances of having chronic pain. We believe that this staged approach which leads to toe salvage is the best suitable treatment option for the recurrent chondromyxoid fibroma. This will prevent amputation of the toe and will give cosmetic success to the patient.

Key Words
Chondromyxoid fibroma, curettage, pseudarthrosis, salvage treatment

Implications for Practice:

1. What is known about this subject?
The recurrence is a known complication of CMF of the toe after its primary treatment like curettage and bone grafting.

2. What new information is offered in this case study?
If there is no feature of malignancy, interposition arthroplasty can be performed with the tricortical iliac crest graft at the metatarsophalangeal joint. This causes pseudoarthrosis and will prevent the chronic pain. We strongly recommend a staged toe salvage procedure for the recurrent benign CMF.

3. What are the implications for research, policy, or practice?

- Provides clinical features of chondromyxoid fibroma
- Salvage Procedure
- Staged surgical approach
- X ray findings
- Microscopic features of chondromyxoid fibroma
- One year follow up

Background

Jaffe and Lichtenstein[1] described the chondromyxoid fibroma (CMF) for the first time in the literature. CMF is an extremely rare tumour, which accounts for only about 1 per cent of all bone tumors.[2,3] Its reported incidence in the toe is only 5 per cent, as it commonly affects the other lower extremity bones.[2] It is commonly reported in the upper end of tibia and fibula or at the lower end of femur.[4] The foot represents the second most common location for this

tumour after the knee joint.[5] The signs and symptoms of this tumour include chronic pain, swelling and restriction of mobility. Sometimes this tumour ends up in a pathological fracture.[6] The initial treatment of CMF includes curettage and bone grafting. Unfortunately, the commonest complication of this tumour being its recurrence inspite of the curettage.[6–8] There are several causes, which are involved in its recurrence after the initial treatment. The recurrence is because of incomplete removal of tumour cells, higher mitotic division and aggressive behaviour. However the malignant transformation is extremely rare. The en block excision and amputations are considered as the recommended options to manage its recurrence.[2,8] In the present case of recurrent CMF, the amputation of the toe was avoided and it was salvaged by performing the staged surgical approach.

Case details

A 32-year-old male patient, who is a farmer by occupation, visited our hospital with history of pain and swelling in his second toe of left foot. This was present for the past 12 months. The patient is not a known smoker or tobacco chewer. He has no weight loss and loss of appetite. He is moderately built and nourished. In the general physical examination, there were no signs suggestive of anaemia, peripheral oedema and lymphadenopathy. The systemic examinations were normal. The patient says that he had history of pain in the foot, two and half years ago, which was insidious in onset and gradually progressed. After about 2 months of this pain, he observed a swelling around the left second toe. He had surgery for the same at a local hospital and they performed an open biopsy. They also did the curettage of the lesion and bone grafting. The histopathological examination report was suggestive of features of CMF. Following this, after about a year, patient developed pain again and the swelling was reappeared in the second toe, which was of smaller in size initially and gradually progressed in the size. For this recurrence of the symptoms, patient took a decision to visit to our institution.

On local examination, an oval shaped swelling in the second toe of the left foot measuring 30mm×30mm was present. This extended longitudinally from 2nd metatarso phalangeal joint to the middle of the toe and there was also extension horizontally into the adjacent web space pushing the third toe (Figure 1A). The skin was shiny with no erythema and engorged veins. The patient was admitted and preoperative investigations were done in the form of complete blood picture. This was not showing any abnormality with respect to infective conditions like osteomyelitis and dactylitis. The patient was sent for radiological examination, which revealed an eccentric, expansile, and lytic, multi loculated lesion (Figures 1B and 1C), which was arising from the proximal phalanx of second toe.

The MRI was showing well-defined expansile eccentric heterogenously enhancing lesion with signal characteristics as described involving the shaft and base of proximal phalanx of the left 2nd toe with extensions and relations as described. The features were suggestive of chondroid lesion, to consider possibilities of Chondromyxoid fibroid and Enchondroma.

The CT scan was suggestive of well defined, expansile lytic lesion involving the proximal phalanx of the second toe with narrow zone of transition. The lesion shows few internal septations and thin sclerotic margin. These were suggestive of enchondroma or chondromyxoid fibroma. There were no matrix formation and periosteal reaction. The soft tissue swelling was seen involving the second toe.

Finally it was planned for the En block excision of the tumour and bone grafting. Our intention was to save the toe and avoid doing the amputation.

The toe salvage surgery was performed in stages:

Stage 1: As a first stage, the dorsal approach was performed over the second toe, the incision extended between the metatarsophalangeal joint up to the distal interphalangeal joint. The extensor tendon of the second toe was exposed. It was observed that the tumour tissue was pushing the tendon, in an eccentrical manner. The total excision of tumour was performed (Figure 2A). The metatarsophalangeal space was maintained by using a kirschner wire, which was impregnated with the bone cement (Figure 2B). It was observed that the tumour was well encapsulated, leaving a vacant space at the proximal phalanx. The tumour was sent for histopathological examination (Figure 2C). The incision was closed by using the non-absorbable sutures. The patient had uneventful intra and post-operative periods. The operated limb was rested in a below knee plaster slab, which was extending up to the tip of the toe. During the 12th postoperative day, the sutures were removed. The below knee plaster slab immobilization was continued and the patient was advised non weight bearing crutch walk over the left lower limb for a period of about 3 weeks.

The histopathological examination revealed that the tumour tissue was weighing 20 grams, measuring 35×25×25 mms and it was greyish white on cut section. The microscopy revealed 'hypocellular lobules' along with the spindle to stellate cells (Figure 3). There was presence of chondromyxoid matrix with intervening cellular bands of fibrous tissue. The osteoclasts like giant cells were also

observed (Figure 3). There was no histological evidence of malignancy.

Stage 2: Since the histopathological examination had ruled out the malignant transformation, the definitive treatment of bone grafting and soft tissue interposition at the metatarsophalangeal joint was planned.

The patient was reviewed after 3 weeks following the first surgery and stage 2 was undertaken. The site was re-approached through the same dorsal incision. The metatarsophalangeal space was reached, the kirshner wire along with the cementation was removed and the required graft size was measured. A tricortical iliac crest graft was taken from the same side, after confirming the size required to fill the space of proximal phalanx. The graft was inter-positioned along with a soft tissue at the metatarso phalangeal joint. This was performed to develop pseudoarthrosis, which will avoid the chronic pain. The graft was positioned and retained by placing the kirshner's wire, which was passed through the distal phalanx, running across the metatarsophalangeal joint. The skin was closed by using the non-absorbable sutures. The sutures were removed after the 12th postoperative day and the patient was discharged from the hospital. A boot cast was given to protect the graft and advised to have non weight bearing crutch walk for 6 weeks.

At the 6th week postoperative visit, the transfixing kirshner wire was removed. The patient was trained partial to full weight bearing walk for about another 6 weeks. The patient was reviewed again at the 3rd month, he was found to be comfortable and the wound was healed. The patient was periodically reviewed again at the 6th month and 1 year. At one year follow up, the patient is symptomless except some occasional pain after walking for a long distance. However he does not require analgesics to get rid of this pain.

Discussion

CMF is a rare benign tumour, which originates from the cartilage cells. Its peak incidence occurs in the second or third decade of life.[9] However it has a wide range of age distribution.[10,11] It carries higher predilection for men than women.[12] In our case, it is interesting to note that the patient is male and 30 years of age. This agrees with the age and gender predilection of this tumour as per the available literature. This tumour commonly affects the metaphyseal region of long bones. In a short long bone like as in a phalanx, the metaphysis is towards the proximal end. The differential diagnosis of this tumour includes aneurysmal bone cyst, chondroblastoma and chondrosarcoma. This tumour may mimic like pseudotumors like osteomyelitis of the phalanx, tuberculous and leprous dactylitis.[9] However, the histopathological observation will be the accurate investigation and can easily diagnose this rare tumour. The most common complication of CMF is the recurrence and its incidence after the primary curettage and bone grafting accounts for about 15–20 per cent.[6–8] O'Connor et al.[13] reported 4 cases of CMF of the forefoot, of which 3 cases have recurred. One of their cases recurred after a very long 19 years. In the present case, the recurrence had occurred one year after the first surgery which was performed at a local hospital. This is in consistence with most of the previous reported cases of CMF. Even though the malignant transformation is rare, the histopathological examination is recommended for the CMF of toe[8] The radiological features of CMF include expansile, eccentric, lytic and radiolucent lesion with the sclerotic margin.[4] The present case had similar features in the radiological examination.

The chance of recurrence of this tumor is high if the surgical procedure involves only curettage. Bhamra et al.[14] reported that the curettage along with cementing, can decrease the prevalence of recurrence. According to them, cementing will provide the biomechanical stability and also helps in destroying the residual tumour cells. In the present case, the Kirschner wire was impregnated with the bone cement. The bone cement was aimed at the micro tumour tissue destruction. For the recurrent CMF, the recommended treatment is the radical surgical treatment, like block resection or amputation.[8,14] Sharma et al.[4] reported that the CMF of foot should be initially treated with curettage. They also opined that the patient needs follow ups on regular postoperative check-ups. However the recommended treatment for the recurrent CMF can only be an amputation.[4]

There is a lot of debate exists about the treatment options for the recurrent CMF. There appears to be no universally accepted treatment for the recurrent CMF. This is because rarity of this tumour and scarcity of the population based studies.[14] In the present case at our institution, we aimed to preserve the toe and not to go for an amputation. However the en bloc excision with autologous bone grafting will add to the prolonged morbidity to the patient. But in view of preventing the recurrence, it is worthwhile attempting the same. We adopted a staged approach of en block resection, cementation and bone grafting. This was followed by soft tissue interposition arthroplasty of the metatarsophalangeal joint. This pseudoarthrosis is important to prevent the chronic pain which patient develops over a period of time, because he is a farmer who works in the field. The definitive

surgical toe salvage procedure was performed after confirming the histopathological report which was suggestive of the benign nature of the tumour.

On few occasions, CMFs may behave like an aggressive fashion after the surgery, particularly if they are found at the axial skeleton. The malignant conversion is rare and it is difficult to distinguish from a misdiagnosed chondrosarcoma. The death due to CMF is unlikely and the radiation treatment should be avoided because of its side effects like postradiation sarcoma.

It was reported that the chemical cauterization after the curettage of the tumour by using agents like phenol, methyl methacrylate and liquid nitrogen have no advantage over preventing the recurrence (Figures 4-6).

Conclusion

The recurrence of CMF of the toe is a known complication after its primary treatment like curettage and bone grafting. We suggest performing a staged approach which initially involves the en block resection and maintenance of metatarsophalangeal space by using the kirshner wire with bone cementing. The tumour should be sent for histopathological examination to confirm its benign nature. If there is no malignancy features, we recommend the procedure of interpositional arthroplasty by using tricortical iliac crest graft at the metatarsophalangeal joint, which will develop the pseudoarthrosis. This pseudoarthrosis will prevent the chronic pain, which may develop over a period of time. We strongly recommend a staged toe salvage procedure for the recurrent benign CMF.

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PEER REVIEW

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CONFLICTS OF INTEREST

The authors declare that they have no competing interests.

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PATIENT CONSENT

The authors, *Krishnaprasad PR, Acharya K, Pandey V, Madi S, Murlimanju BV,* declare that:

1. They have obtained written, informed consent for the publication of the details relating to the patient(s) in this report.
2. All possible steps have been taken to safeguard the identity of the patient(s).
3. This submission is compliant with the requirements of local research ethics committees.

Figure 1: The swelling extended longitudinally from the 2nd metatarso phalangeal joint to the middle of the toe of left foot. It also extended horizontally to the adjacent web spaces, pushing the third toe (1A). The X ray films of the patient, which exhibited eccentric, expansile, lytic and multi loculated lesion (1B & 1C)

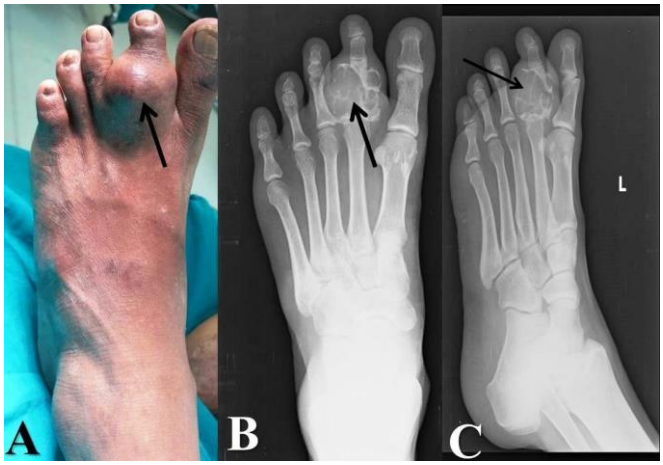

Figure 2: The total excision of tumour was performed by the dorsal approach (2A). The metatarsophalangeal space was maintenance by kirschner wire which was impregnated with bone cement (2B). The tumour was observed to be well encapsulated and it was weighing about 20 grams (2C)

Figure 3: The histopathological examination (H & E stain) showed osteoclast like giant cells. There was chondromyxoid matrix with intervening cellular bands of fibrous tissue. Hypocellular lobules were seen along with the spindle to stellate cells

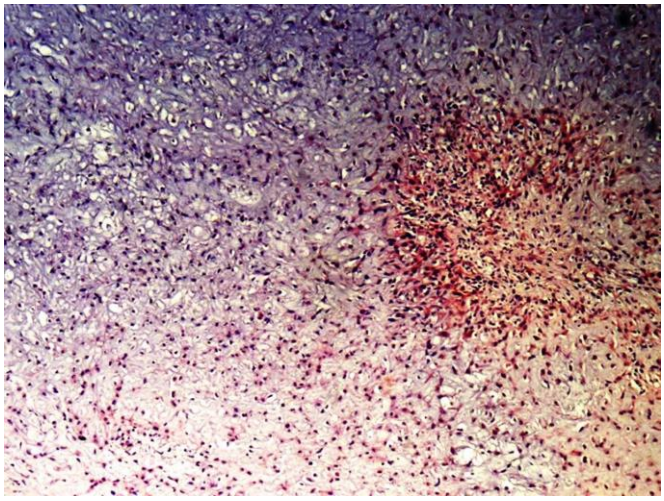

Figure 4: The X rays pictures showing iliac crest autograft interpositioned in the second metatarsophalangeal space with interpositional arthroplasty as a second stage surgical procedure

Figure 5: The x ray pictures post second stage 6 weeks

Figure 6: The CT (6A & 6B) and MRI images (6C & 6D) of the lesion