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CASE REPORT
KAZUISTIKA

Giant Cell Tumor of the Proximal Phalanx: Report of two Cases Treated by Two Different Methods and Review of the Literature

Obrovskobuněčný nádor proximálního článku: zpráva o dvou případech léčených dvěma různými metodami a přehled literatury

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SUMMARY

The hand is an extremely rare site for giant cell tumor (GCT). There are only a few reported cases of GCT including the hand, and even fewer reporting involvement of phalanges. GCTs in small bones are typically more aggressive and have higher local recurrence and rate of metastasis in younger patients compared to long bone involvement, so the treatment is more clinically challenging in the hand. In this study, we present the management of giant cell tumors of the proximal phalanxin two patients treated with two different method; ray resection and arthrodesis using an iliac crest graft.

Key words: giant cell tumor, phalanx, hand, recurrence.

INTRODUCTION

Giant cell tumor (GCT) of bone is a relatively common, benign but aggressive tumor and usually involves the metaphyseal-epiphyseal region of long bones, usually at the distal femur and proximal tibia (8). GCTs are typically seen in early adulthood with a peak incidence between 30 and 50 years (10). However, the hand is an extremely rare site for GCT. Only 0.6 % of all GCT are reported to occur in the phalanx (15). There are only a few reported cases that include the phalanges. Furthermore, GCT in small bones is typically more multicentric and more aggressive and has a higher rate of both local recurrence and metastasis in younger patients compared to GCTs involving the long bones (3, 4). Thus, the treatment is critical and more clinically challenging in the hand.

Surgical treatment of small bone GCT is still controversial because each method has advantages and disadvantages. The aims of treatment are to provide local control and prevention of metastasis, restore function and maintain good cosmoses.Curettage + adjuvant therapy + grafting is the suggested method in many publications but the recurrence rate was reported to be as high as 75% (14). En bloc resection, amputation and ray resection are the surgical treatment options that will reduce recurrence. Short-course neoadjuvant denosumab is also advised for advanced GCT, but increased recurrence rates remain of concern (16).The anatomical site and the extent of the tumor also affect the choice.

In this study, we present the management of GCT of proximal phalanx in two patients treated using two different methods. One was treated with ray resection and the other with arthrodesis using iliac crest graft.

CASE 1

A 19-year-old male patient was admitted to the emergency department with symptoms of pain and deformity of his left ring finger after a minor trauma. There were no symptoms prior to the trauma. He was subsequently diagnosed as having a fracture of the proximal phalanx of the fourth finger and treated with a splint for four weeks (Fig. 1a). After four weeks the splint was removed but the patient continued to suffer from pain and increased swelling. He attended another hospital, two weeks after splint removal, for roentgenographic assessment. The X-ray showed a lytic and expansible lesion, causing geographic destruction at the base of the proximal phalanx extending distally to mid-diaphysis (Fig. 1b). He was then referred to our oncology department for further investigation. On physical examination, there was a remarkable swelling at the proximal end of the left fourth finger. Magnetic resonance imaging (MRI) was ordered due to the possible differential diagnoses of brown tumor, aneurysmal bone cyst, GCT or Ewing sarcoma of the proximal phalanx.

Contrast enhanced MRI showed anintensely enhancing solid mass that invaded the phalanx and had destroyed the cortex, as well as extending to surrounding soft tissue and involved neurovascular structures (Fig. 1c–1d). There was also involvement of the fourth metacarpal head. His chest tomography was negative for pulmonary metastasis and tecnetium^{99} (Tc^{99m}) bone scintigraphy was also neg-

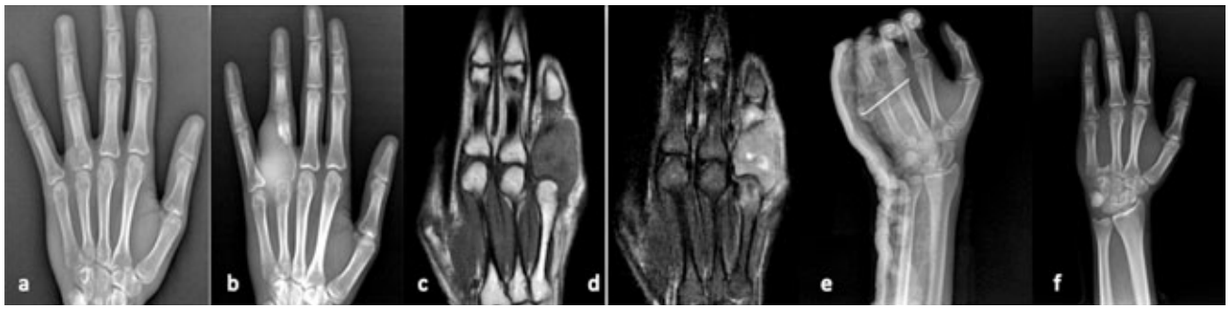

Fig. 1. X-ray and MRI views of the hand in Case 1: a – AP roentgenography of the hand at initial admission to the hospital; b – AP roentgenography of the hand six weeks after the first admission; c – T1 and d – T2 images of the lesionon MRI; e – post-operative AP roentgenography of the hand; f – AP roentgenography of the hand two months after surgery.

ative for other bone lesions. A core needle biopsy was performed after imaging work up, which was consistent with GCT of bone. Combining histopathological information with the imaging findings, the tumor was classified as Campanacci Stage 3. Taking into consideration the involvement of neurovascular structures and the metacarpal head, a ray amputation was performed. temporary fixation of the fifth metacarpal head to the third metacarpal head was performed with a K-wire, which was removed four weeks after the surgery (Fig. 1e–1f). The patient was well at follow-up two months post operatively with normal daily living activities using his left hand (Fig. 2). Functionality of the hand was evaluated by the Musculoskeletal Tumor Society (MSTS) Scoring System and the score was 26/30 (86%rating) (Table 1). No recurrence was observed after 2 years follow-up.

Macroscopically, the lesion was 4x4x3cm in size and pink-white colored. It had infiltrated the proximal phalanx, extending into the metacarpal head and surrounding soft tissue. The tumor had also infiltrated the digital arteries and nerves on both sides. Microscopically, the tumor consisted of diffuse, osteoclast-like, giant cells and spindle-shaped mononuclear cells that were set in single row stroma. The nuclei of the mononuclear cells were normochromatic and were similar to the nuclei of the osteoclast-like giant cells. Furthermore, the giant cells consisted of 10-30 nuclei that were mostly centrally located (Fig. 3).

Table 1. Musculoskeletal Tumor Society (MSTS) Scoring System for the upper extremity

Score	Pain	Functional activity	Hand positioning	Dexterity	Lifting ability (muscle power)	Emotional acceptance
5	No pain	No restriction (no disability)	Unlimited (can be elevated 180°)	No limitations	Normal load (muscle power 5)	Enthused
3	Modest/ Non-disabling	Recreational restriction (minor disability)	Not above shoulder or no pronation supination (can be elevated 90°)	Loss of fine movements (buttoning, writing, eating)	Minor load (muscle power 3)	Satisfied
1	Moderate/ Intermittently disabling	Partial occupational restriction (major disability)	Not above waist (can be elevated 30°)	Cannot pinch	Cannot overcome gravity (muscle power 2)	Accepts
0	Severe/ Continuously disabling	Total occupational restriction (complete disability)	None (0° elevation)	Cannot grasp	Cannot move (muscle power 0,1)	Dislikes

Fig. 2. Image of the hand two months after surgery in Case 1.

CASE 2

A 21-year-old female patient was admitted to an emergency department with symptoms of pain and deformity of her right middle finger after a minor trauma with a ball. She was initially diagnosed with a pathologic fracture of her proximal phalanx of the third finger of the right hand. However, a lytic lesion with geographic destruction at the base of the proximal phalanx was seen on X-ray (Fig. 4a). Subsequent contrast enhanced

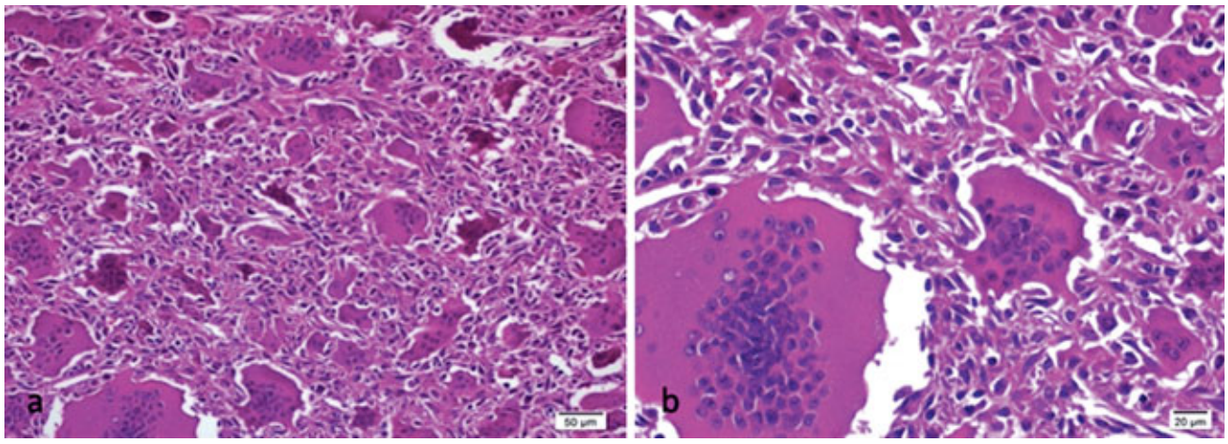

Fig. 3. Case 1: a – diffuse osteoclast-like, giant cells and spindle-shaped mononuclear cells that were set in single row stroma; b – giant cells with 10–30 nuclei.

MRI showed a T1 hypointense, T2 hyperintense solid mass without a soft tissue component. A core needle biopsy was performed that was consistent with GCT of bone. Her thorax tomography and Tc^{99m} bone scintigraphy were negative for metastasis. The tumor was classified as Campanacci Stage 2. Curettage and bone grafting was the choice of treatment and a Kirshner wire was used as a scaffold to maintain the length of the phalanx (Fig. 4b). Two weeks after the surgery, the wound was clean and the sutures were removed. She was next see none year after surgery because the patient missed the scheduled follow-ups because of the pandemic and was admitted to the clinic with a deformed and swollen third finger. On X-ray there was a large and lytic expansible mass due to local recurrence (Fig. 4c). Her thorax tomography and Tc^{99m} bone scintigraphy were again negative for metastasis. After radiologic assessment there were two options, wide resection of the phalanx and arthrodesis of the MCP and PIP joint using iliac crest graft or ray resection of the third finger. Taking into consideration the functional results of the MCP joint arthrodesis and the risk of recurrence and lung metastasis, the clinical decision was taken to perform ray resection. However, the patient refused amputation and underwent wide resection and arthrodesis. Minifragment plate and iliac crest autograft were used for the arthrodesis of the MCP and PIP joints (Fig. 4d). Satisfactory result had been achieved with good cosmesis and there is no recurrence 1 year after the revision surgery (Fig. 5). She scored 24/30 (80% rating) according to the MSTS Scoring System, similar to Patient 1 (see Table 2).

DISCUSSION

GCT of the bone is a benign but aggressive lesion with a propensity for local recurrence and metastasis to the lung. GCT of bone has characteristic radiolucent and geographic appearance with a narrow zone of transition, often without a sclerotic rim. Although GCT is

Fig. 4. Radiographs of Case 2: a – at first admission; b – after the curettage and grafting; c – as a result of recurrence, a large and lytic expansile mass was evident; d – after revision surgery.

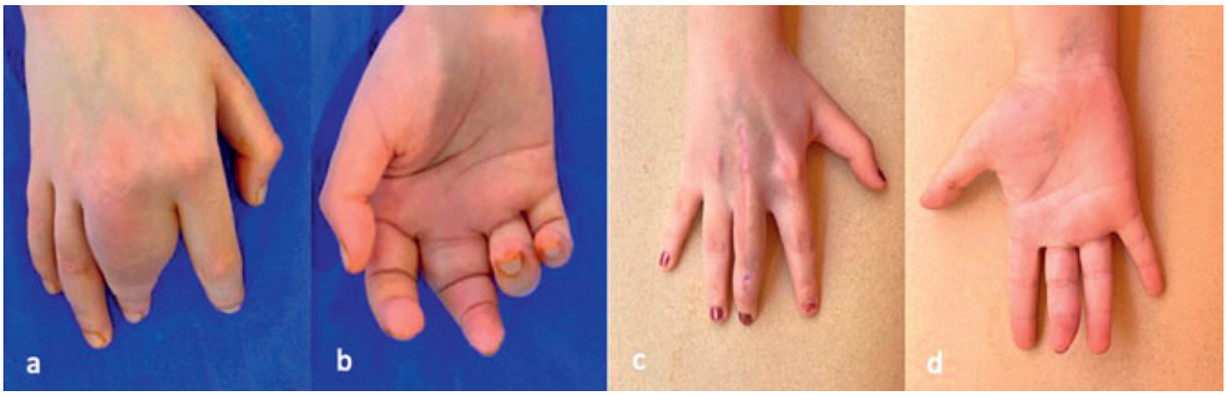

Fig. 5. Dorsal (a) and volar (b) images of the hand before and three months after (c, d) the surgery.

a benign lesion, cortical destruction with soft tissue extension may also occur and is easily detected with MRI.

GCT of the bones of the hand is extremely rare and GCT affecting the phalanges is extremely rare. In a GCT case series of 108 patients, Coley et al. reported only two(1.85%) cases arising from the phalanges (7), Goldenberg et al. reported 218 cases of GCT and only six(2.75%) arising from phalanges (17), while Campanacci et al. reported 327 GCT cases and only one (0.3%) arising from a phalanx (6). Besides being rare, GCT of the hand usually occurs in younger age groups. There is a reported mean patient age of 22 years, which is younger than conventional GCT (4). Furthermore, GCT of the hand has a more aggressive behavior pattern with a higher incidence of local recurrence after intralesional curettage comparing to GCT occurring in other bony locations (12).

There is no consensus concerning the management of GCT in the phalanx. Edward et al reported 13 cases in the hand, of which seven lesions occurred in the metacarpals, five in the phalanges and one in the scaphoid. Ten of these patients were treated initially with curettage, with or without bone grafting, and recurrence was noted in 8 of the10. The remaining three patients were initially treated either with wide excision or ray resection and recurrence was noted in one of three. Eight of the nine recurrent cases had secondary treatment. Three of them were treated by repeat curettage and a second recurrence was noted in all three patients. Excision, amputation or ray resection was done after initial recurrence in five patients and three of these also had recurrence (3). Patel et al. reported five cases in the phalanges and advocated that the high risk of recurrence was related to the difficulty to eradicate the tumor in the hand because of the complex arrangements of soft tissues in the hand and suggested amputation or ray resection if the cortical integrity was compromised (13). Depalmaet al. found 100% success rate when the tumor was completely excised and either the joint was fused or replaced by a bone graft (8).

However, lesions in the proximal phalanges should be considered as a different matter because the MCP, PIP or both may be involved. Cosmetic appearance and function both need to be taken into consideration. Ansari et al. reported a proximal phalanx GCT treated with silicone implant and fibula autograft after resection of the proximal part of the phalanx (2). Spiro et al. used osteochondral autograft from lateral femoral condyle and Kanaya et al. used vascularized metatarsophalangeal joint transfer for GCT of the proximal phalanx (9,15). All of these three case reports reported good functional results in the selected cases with only proximal joint involvement without soft tissue extension and neurovascular involvement.In contrast, Mohammad et al. reported two (0.6%) cases of GCT arising from phalanges in 350 GCT cases between 1985 and 2020 (1). Both of them were in the proximal phalanx in the first and the index fingers and both MCP and PIP joints were involved. They were treated by excision of the involved phalanx and arthrodesis with cement spacer. As the thumb is critical to normal hand function, arthrodesis was the preferred method rather than amputation in that case, and in the second case affecting the index finger, the patient did not accept amputation so arthrodesis was the preferred method. The authors advocated that arthrodesis may not be functional or cosmetically good but could be an alternative to amputation in certain situations.

CONCLUSIONS

GCT of the hand should be included in the differential diagnosis of the hand bone lesions. Phalangeal GCT le-

Table 2. Evaluation of the patients by Musculoskeletal Tumor Society Scoring System

	Pain	Functional activity	Hand Positioning	Dexterity	Lifting ability	Emotional acceptance
Patient 1	5	5	5	5	5	1
Patient 2	5	3	5	3	3	5

sions are extremely rare, the treatment is challenging because of the high recurrence rate and should be treated in specialized centers. Lesions in the phalanx, except the thumb, with soft tissue extension or involving both the distal and proximal articular surface, may be treated by ray amputation with good functional result and satisfactory cosmesis. Furthermore, although the arthrodesis may not be functional or cosmetically good, satisfactory results may be achieved if this method is the preference of the patient.

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