Resection and Reconstruction is the Procedure of Choice for Giant Cell Lesions of Small Bones: A Case Report and Literature Review

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Case report

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Abstract

Background: Giant cell lesion of small bones (GCLSB), also known as giant cell reparative granuloma, is a rare tumor-like condition occurring in the small bones of the hands and feet. GCLSB lacks specific clinical, radiological, and histological manifestations. There are no standardized protocols for treatment.

Case presentation: Here, we report a 16-year-old male with recurrent GCLSB in the proximal phalanx of the left thumb. The lesion was successfully resected with bone grafting.

Conclusions: We summarized the characteristics of 33 reported cases of GCLSB from 1983 to date, including gender, age, lesion sites, recurrence, and treatment. We conclude that resection and reconstruction with curettage is the treatment of choice.

Background

Giant cell lesion of small bones (GCLSB) is an infrequent tumor-like condition occurring in the hands and feet. Also known as giant cell reparative granuloma (GCRG), GCLSB is defined by the World Health Organization as a lesion consisting of fibrous tissue with hemorrhage, hemosiderin deposits, irregularly distributed giant cells, and reactive bone formation. Because the features of GCLSB lack specificity, diagnosis requires a combination of clinical examination, imaging, and pathology. At present, there is no standardized treatment for GCLSB, primarily because there are so few reports. We present a case of recurrent GCLSB of the phalanx treated with surgical resection. We discussed the clinicopathological features of the disease and its treatment and provided a review of the literature.

Case Presentation

In August 2017, a 16-year-old male presented with swelling and pain of the left thumb. A lump the size of a fingernail with pain on palpation was found on the radial side of the left thumb. There was no previous history of trauma. He went to Central Hospital of Siping city in September 2017. X-ray revealed a lytic expansile lesion of the proximal phalanx of the left thumb (Fig. 1). The patient was treated with curettage, 99.5% ethanol sterilization, and grafting in September 2017. Postoperative pathology identified giant cell reparative granuloma.

In July 2018, the patient presented with recurring swelling and pain of the left thumb. As the lump grew progressively, the patient came to our hospital in September 2018. Laboratory examinations (complete blood count, prothrombin time, partial thromboplastin time, and serum level of calcium) were within normal ranges. Magnetic resonance imaging (MRI) revealed a distended lesion in the proximal phalanx of the left thumb that presented with high signal intensity in in both enhanced T1-weighted and enhanced T2-weighted images (Fig. 2). The patient was treated with curettage, 99.5% ethanol sterilization, and grafting in September 2018. Postoperative pathology identified giant cell reparative granuloma.

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Literature review

We performed a Medline literature search to identify cases of GCLSB. English and non-English-language papers were searched in PubMed using a combination of terms: ((((((metacarpal) OR (carpal bones)) OR (phalanx)) OR (metatarsal bones)) OR (tarsal bones)) OR (hand)) OR (foot)) AND (giant cell reparative granuloma)) OR (giant cell lesion of small bones). The search was carried out using the literature from 1983 to the present. The data available are summarized in Table 1. We were able to find 33 patients (include this study) with 37 lesions. The 33 patients included 16 men and 17 women with a mean disease duration of 22.3 years (range 4.5 to 67 years). Most patients were in their second decade (48.5%) followed by the third (21.2%), first (8%), fourth (9.1%) and fifth decades (9.1%). The lesion occurred most often in a phalanx (17 lesions, 45.9%), followed by metacarpal (11 lesions, 33.3%), metatarsal (five lesions, 15.1%), carpal bones (three lesions, 9.1%), and calcaneus (one lesion, 3%). Pain (19 cases), swelling (12 cases), and pathological fracture (four cases) were the symptoms mentioned most often. Among the 38 lesion sites, 26 were treated with curettage, ten were treated with resection and two were treated with amputation. Recurrence (in nine lesions) occurred only after curettage that proceeded a second surgery. Five of the nine lesions were treated with resection, three were treated with curettage and one was treated with amputation (Table 2). Regardless of the operation frequency, 19 lesions (51.3%) were finally cured with curettage, 16 were cured with resection (40.5%), and three were cured with amputation (8.1%).
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| try3 | 4.8          |           | 8.0           |      |
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| Position | Key       |           | 6.4           |      |
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| try2 | 3.7          |           | 7.5           |      |
| try3 | 3.9          |           | 7.6           |      |
| Avge | 3.9          |           | 7.6           |      |
| CV%  | 4.4          |           | 1.3           |      |
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| Index | 7.7          |           | 5.1           |      |
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| Small | 7.6          |           | 6.9           |      |
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|      |              |           | 2.6           |      |
|      |              |           | 2.7           |      |
| Year | Authors          | Number of patients | Age (years)/Gender | Site                 | Symptom                          | Treatment                          | Recurrence | Treatment after recurrence | Follow-up (months) |
|------|------------------|--------------------|--------------------|----------------------|----------------------------------|-----------------------------------|------------|---------------------------|------------------|
| 1983 | Glass et al.     | 2                  | 27/Male            | Metacarpal, capitate, lunate | NR                              | Curettage                         | No         | -                         | 26               |
|      |                  | 26/Female          |                    | Phalanx              | Painless                         | Curettage and grafting            | Yes        | Resection and reconstruction | 48               |
|      |                  | 13/Male            |                    | Phalanx              | Swelling                         | Curettage and grafting            | Yes        | Amputation                | 121              |
| 1985 | Caskey et al.    | 1                  | 24/Male            | Metacarpal, capitate | Swelling, pathologic fracture    | Curettage and grafting            | No         | -                         | 13               |
| 1985 | Merkow et al.    | 3                  | 46/Female          | Phalanx              | Pain, swelling                   | Curettage and grafting            | No         | -                         | 14               |
|      |                  | 16/Female          |                    | Metacarpal            | Pain, swelling                   | Curettage and grafting            | Yes        | Curettage and grafting    | 24               |
|      |                  | 14/Male            |                    | Phalanx              | Pathologic fracture              | Curettage and grafting            | No         | -                         | 24               |
| 1987 | Wenner et al.    | 1                  | 13/Male            | Phalanx              | Swelling                         | Curettage and grafting            | Yes        | Resection and reconstruction | 12               |
| 1989 | Robinson et al.  | 1                  | 17/Male            | Calcaneus            | Pain, pathological fracture      | Curettage and grafting            | No         | -                         | 24               |
| 1989 | Dwyer et al.     | 1                  | 32/Female          | Phalanx              | Swelling, pain                   | Curettage and grafting            | No         | -                         | 17               |
| 1994 | Panico et al.    | 5                  | 31/Female          | Metatarsal           | NR                               | Curettage and grafting            | No         | -                         | 32               |
|      |                  | 17/Male            |                    | Metatarsal           | NR                               | Curettage and grafting            | No         | -                         | 33               |
|      |                  | 16/Male            |                    | Phalanx              | NR                               | Curettage and grafting            | Yes        | Curettage                 | 27               |
|      |                  | 34/Female          |                    | Phalanx              | NR                               | Resection                         | No         | -                         | 30               |
|      |                  | 41/Male            |                    | Metacarpal           | NR                               | Resection                         | No         | -                         | 65               |
| 1997 | Giza et al.      | 1                  | 67/Female          | Metacarpal           | Pain, swelling                   | Amputation                        | No         | -                         | 36               |
| 1998 | Bertoni et al.   | 1                  | 52/Male            | Phalanx              | Pain                             | Curettage and grafting            | No         | -                         | 24               |
| 1998 | Arenson et al.   | 1                  | 19/Female          | Metatarsal           | Pain, swelling                   | Resection and reconstruction       | No         | -                         | 23               |
| 1999 | Ugwonali et al.  | 1                  | 4.5/Female         | Metacarpal           | Pathological fracture            | Curettage and grafting            | Yes        | Resection and reconstruction | 36               |
| 2000 | Forouhar et al.  | 3                  | 15/Female          | Metatarsal           | Pain                             | Amputation                        | No         | -                         | 204              |
|      |                  | 8/Female           |                    | Phalanx              | Pain                             | Curettage and grafting            | No         | -                         | 24               |
|      |                  | 12/Female          |                    | Phalanx              | Pain                             | Curettage and grafting            | Yes        | Resection and reconstruction | 48               |
| 2003 | Gouin et al.     | 1                  | 14/Female          | Metatarsal           | Pain                             | Curettage and grafting            | No         | -                         | 36               |
| 2003 | Macdonald et al. | 1                  | 25/Male            | Metacarpal           | Pain, swelling                   | Resection and reconstruction       | No         | -                         | 7                |
Table 3: Collation and analysis of data in Table 2

| Year  | Authors                        | Number of patients | Age (years)/Gender | Site          | Symptom       | Treatment                                      | Recurrence | Treatment after recurrence | Follow-up (months) |
|-------|--------------------------------|--------------------|--------------------|---------------|---------------|------------------------------------------------|------------|---------------------------|--------------------|
| 2007  | Yoshida et al.                  | 2                  | 7/Female           | Metacarpal    | Pain          | Curettage and grafting                          | Yes        | Curettage, phenol and ethanol sterilizing, and grafting | 84                 |
|       |                                |                    |                    |               |               |                                                |            | -                         | 36                 |
| 2008  | Saghieh et al.                  | 1                  | 13/Female          | Metacarpal    | Pain, swelling| Resection and reconstruction                    | No         | -                         | 13                 |
| 2008  | Cook et al.                     | 1                  | 26/Female          | Phalanx       | Pain          | Resection                                       | No         | -                         | 24                 |
| 2011  | Perkins et al.                  | 1                  | 16/Male            | Phalanx       | Pain          | Curettage and grafting                          | No         | -                         | 8                  |
| 2012  | Monacelli et al.                | 1                  | 16/Male            | Phalanges     | NR            | Resection and reconstruction                    | No         | -                         | 6                  |
| 2016  | Huan et al.                     | 1                  | 21/Female          | Phalanx       | Pain, swelling| Resection and reconstruction                    | No         | -                         | 8                  |
| 2017  | Telisselis et al.               | 1                  | 16/Male            | Metacarpal    | NR            | Resection and reconstruction                    | No         | -                         | 24                 |
| 2020  | Present study                   | 1                  | 16/Male            | Phalanx       | Pain, swelling| Curettage and grafting                          | Yes        | Resection and reconstruction | 17                 |

Discussion

GCLSB is a very rare tumor-like lesion that occurs in hands and feet; it was newly defined in the WHO classification of tumors of soft tissue and bone in 2013. Previously, it was known as giant cell reparative granuloma. In 1953, Jaffe first reported GCRG as a macrophage-rich bone lesion occurring in jaw bones; he emphasized that it was clinically and histologically different from giant cell tumors. Later, it was found not only in the jaw bones, but also in bones throughout the body. In 1983, GCRG was reported in metacarpal, capitate, lunate bones for the first time. It occurs particularly in the phalanx and metacarpal bones. It tends to occur in adolescents in their second decades. Patients initially experience pain and swelling in the lesion site, and pathologic fractures may develop as the disease progresses. Radiographically, there is an expanded osteolytic lesion in the metaphysis or diaphysis, with thinning of the cortical bone, but no destruction and no periosteal response. Histopathological examination shows spindle-shaped fibroblast hyperplasia with hemorrhage, hemosiderin deposits, irregularly distributed osteoclast-like giant cells and reactive bone formation. Osteoclast-like giant cells are smaller and have fewer nuclei than giant cell tumor of bone. On genetic analysis, rearrangement of chromosomes 8 and 22 were found in a patient with GCRG of the jaw. Nevertheless, it remains to be determined whether this rearrangement is prevalent in GCLSB. Because GCLSB is not rare, both radiologically and histologically, it is necessary to strictly follow the principle of clinical-image-pathology to make appropriate treatment decisions.
To date, there have been no guidelines for treatment because of the low level of morbidity. According to our data in Table 2, surgeons tend to choose curettage and grafting. This results in minimal damage to function; however, it also carries the risk of recurrence. Phenol and ethanol were used by Yoshida et al. to reduce the recurrence rate\textsuperscript{16}. In our case, ethanol did not prevent recurrence. The analysis shows that 36% of lesions recurred after curettage; however, none of resected lesions recurred. Only 51.3% of lesions were finally cured by curettage, while 43.2% of the lesions were cured by resection, and 5.4% of the lesions were cured by amputation (Table 3). Amputation is rarely chosen by surgeons. As it is an extreme form of surgery which lead to totally loss of function.

Resection and reconstruction is the treatment of choice. It not only cures the lesion without a second operation, but it also tends not to compromise function. Macdonald et al. reconstructed the lesion site with iliac crest bone grafting and fascial arthroplasty. His patient’s grip strength and motion at metacarpophalangeal joint was preserved\textsuperscript{15}. For the young patient described in this report, the lesion occurred in the shaft of the proximal phalanx of the thumb. To prevent recurrence, we had to remove the proximal phalanx, interphalangeal joint, and metacarpophalangeal joint, and conduct joint fusion with bone grafting. Postoperative follow-up revealed slightly lower pinching power of the thumb. If we had tried to preserve the metaphysis and remove the diaphysis of phalanx with grafting during the first operation, the proximal phalanx, interphalangeal joint, and metacarpophalangeal joint could be retained, and the function of thumb might be better preserved.

**Conclusion**

In summary, we report a case of a 16-year-old man with recurrent GCLSB that was successfully resected with bone grafting. We conclude that resection and reconstruction with curettage is the treatment of choice.

**Abbreviations**

GCLSB, giant cell lesion of small bones; GCRG, giant cell reparative granuloma; MRI, magnetic resonance imaging

**Declarations**

**Ethics approval and consent to participate**

Not applicable.

**Consent for publication**

Informed consent was obtained from the patient.

**Availability of data and materials**

All data generated or analyzed during this study are included in this published article.

**Competing interests**

The authors have no potential conflicts of interest to disclose.

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**Authors’ contributions**

Jiannan Li wrote the initial draft. Zhan Zhang and Guangzhi Wu were the surgeon. Weizhong Zhang performed the pathological examination. All authors read and approved the final manuscript.

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**Figures**
Figure 1
Pre- and postoperative images in 2017.07. (A) Lytic expansile lesion of the proximal phalanx of the left thumb on X-ray (white arrow). (B) The lesion was curetted.

Figure 2
Preoperative, intraoperative and postoperative images in 2018.08 (A) Recurrent lesion of the proximal phalanx of left thumb on X-ray (white arrow). (B) MRI revealing the recurrent lesion (white arrow). (C) The lesion was resected with grafting, interphalangeal joint arthrodesis. The stop point of the adductor pollicis was reconstructed. (D) Postoperative images on X-ray. (E) Pathological examination revealing peculiar features of the giant cell lesion of small bones. Multinucleated giant cells (asterisks) are clustered in the lesion. Bar=100μm
Figure 3

Post-surgical follow-up evaluation (A) X-ray revealing no local recurrence at 3-month follow-up. (B) X-ray revealing no local recurrence at 17-month follow-up. (C) The palm-to-palm test of the thumb was negative.