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[1]. 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 border was distinct and the cortex was intact. The surfaces of the joints were smooth. Subsequently, the proximal phalanx, interphalangeal joint, and metacarpophalangeal joint of left thumb were resected. Iliac bone measuring 3.0 x 1.0 x 1.0 cm was grafted. The interphalangeal joint was held in place longitudinally and cross-fixed using two Kirschner wires. The stop point of the adductor pollicis was reconstructed at the base of the proximal phalanx. The hallux abductor brevis stop was sutured to the radial side of the metacarpophalangeal capsule (Fig. 2). Histological analysis revealed heterogeneous fusiform or fibroblast-like cells with occasional mitotic figures. Some fusiform cells were filled with lipid without definite atypia. There were scattered multinucleated giant cells (Fig. 2). No clinical or radiographic evidence for a recurrent lesion was found at 17-month follow-up (Fig. 3). The palm-to-palm test of the thumb was negative (Fig. 3). Pinch strength was measured using the Biometrics E-LINK system (Biometrics Limited, US). The pinch strength of the left thumb was slightly lower than normal (Table 1). Using the AMA impairment guidelines, the analysis suggest that the left upper limb lost 12% of its function after surgery.

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 2. 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%), carpel 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%).

Table 1
Pinch strength analysis

Left					**Unit: kgs**	**Right**				
try1	try2	try3	Avge	CV%	Position	try1	try2	try3	Avge	CV%
5.0	5.1	4.8	5.0	3.1	Key	8.3	7.6	8.0	8.0	4.4
4.2	3.7	3.9	3.9	6.4	Three jaw	7.7	7.5	7.6	7.6	1.3
2.8	2.6	2.8	2.7	4.2	Tip to tip	5.6	5.1	4.9	5.2	6.9
	index	mid	ring	small	Thumb to	index	mid	ring	small	
	2.4	1.4	1.1	0.4		4.4	2.6	2.3	1.3	

Table 2
Giant cell lesions of small bones. Systematic review of the literature from 1983 to 2020. NR = Not Reported

Year	Authors	Number of patients	Age (years)/Gender	Site	Symptom	Treatment	Recurrence	Treatment after recurrence	Follow-up (months)
1983	Glass et al.[2]	3	27/Male	Metacarpal, capitate lunate	NR	Curettage	No	-	-
			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.[3]	1	24/Male	Metacarpal, capitate	Swelling, pathologic fracture	Curettage and grafting	No	-	13
1985	Merkow et al.[4]	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.[5]	1	13/Male	Phalanx	Swelling	Curettage and grafting	Yes	Resection and reconstruction	12
1989	Robinson et al.[6]	1	17/Male	Calcaneus	Pain, pathological fracture	Curettage and grafting	No	-	24
1989	Dwyer et al.[7]	1	32/Female	Phalanx	Swelling, pain	Curettage and grafting	No	-	17
1994	Panico et al.[8]	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.[9]	1	67/Female	Metacarpal	Pain, swelling	Amputation	No	-	36
1998	Bertoni et al.[10]	1	52/Male	Phalanx	Pain	Curettage and grafting	No	-	24
1998	Arenson et al.[11]	1	19/Female	Metatarsal	Pain, swelling	Resection and reconstruction	No	-	23
1999	Ugwonali et al.[12]	1	4.5/Female	Metacarpal	Pathological fracture	Curettage and grafting	Yes	Resection and reconstruction	36
2000	Forouhar et al.[13]	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.[14]	1	14/Female	Metatarsal	Pain	Curettage and grafting	No	-	36
2003	Macdonald et al.[15]	1	25/Male	Metacarpal	Pain, swelling	Resection and reconstruction	No	-	7

Year	Authors	Number of patients	Age (years)/Gender	Site	Symptom	Treatment	Recurrence	Treatment after recurrence	Follow-up (months)
2007	Yoshida et al.[16]	2	7/Female	Metacarpal	Pain	Curettage and grafting	Yes	Curettage, phenol and ethanol sterilizing, and grafting	84
			23/Male	Metacarpal	Pain	Curettage, phenol and ethanol sterilizing, and grafting	No	-	36
2008	Saghieh et al.[17]	1	13/Female	Metacarpal	Pain, swelling	Resection and reconstruction	No	-	13
2008	Cook et al.[18]	1	26/Female	Phalanx	Pain	Resection	No	-	24
2011	Perkins et al.[19]	1	16/Male	Phalanx	Pain	Curettage and grafting	No	-	8
2012	Monacelli et al.[20]	1	16/Male	Phalanges	NR	Resection and reconstruction	No	-	6
2016	Huan et al.[21]	1	21/Female	Phalanx	Pain, swelling	Resection and reconstruction	No	-	8
2017	Telisselis et al.[22]	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

Table 3
Collation and analysis of data in Table 2

	Number of lesions	Recurrence number	Rate of recurrence	Treatment after recurrence	Final number of treatment
Curettage	25	9	36.0%	Curettage (3) Resection (5) Amputation (1)	19
Resection	10	0	0	0	15
Amputation	2	0	0	0	3
Total	37	9	24.3%	Curettage (3) Resection (5) Amputation (1)	37

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[1]. 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[23]. 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[2]. It occurs particularly in the phalanx and metacarpal bones[2]. 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[1]. 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[1]. On genetic analysis, rearrangement of chromosomes 8 and 22 were found in a patient with GCRG of the jaw[24]. 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[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.[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

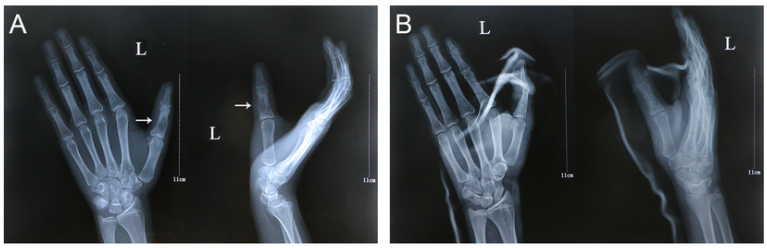

Figure 1

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.

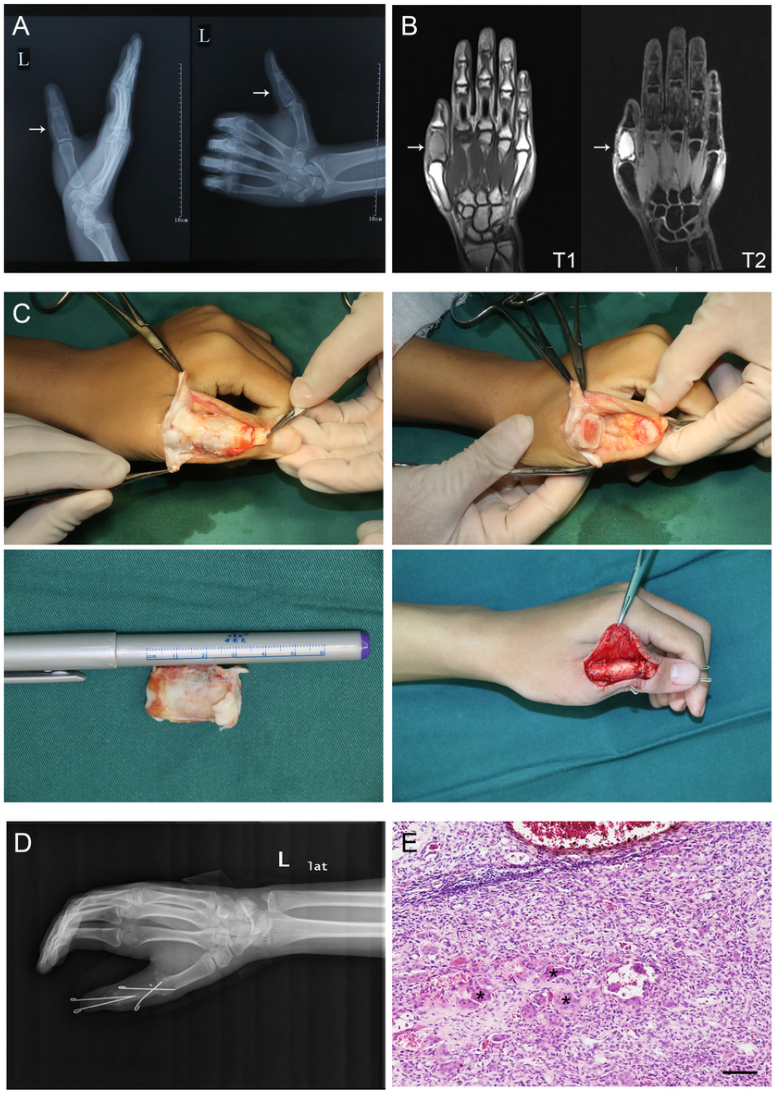

Figure 2

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

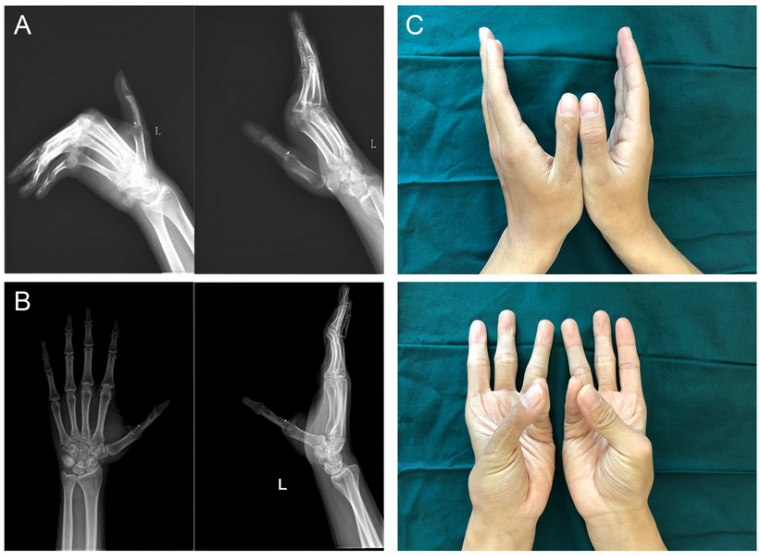

Figure 3

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.