CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 80 (2021) 105686

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Unusual presentation in Haiti of a recurrent giant cell tumor of bone affecting the distal radius: A case report

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ARTICLE INFO

Article history:
Received 26 December 2020
Received in revised form 16 February 2021
Accepted 18 February 2021
Available online 22 February 2021

Keywords:
Giant cell tumor of bone
Haiti
Global health
Radius
Centralization
Case report

ABSTRACT

INTRODUCTION: Giant cell tumor of bone (GCTB) is a benign-aggressive tumor that has a high-rate of recurrence with curettage resection alone. Patients with GCTB in underserved regions of the world can have progression of the tumor with significant disability due to a lack of specialty care. We present a case of an en bloc resection of an aggressive, recurrent GCTB of the radius with excellent function and no evidence of tumor recurrence two years after surgery.

PRESENTATION OF CASE: A 22-year-old right-hand dominant female in Haiti developed an aggressive recurrence of a giant cell tumor of bone (GCTB) of the distal radius. Treatment consisted of en bloc resection of the distal radius with the proximal row of the carpus and centralization of the ulna. At two-year follow-up, the patient maintained good functional capacity with no clinical or radiological evidence of recurrence.

DISCUSSION: GCTB can cause significant destruction of the bone and articular surface if not treated adequately. Treatment options should be considered carefully in underserved regions of the world based on the resources available. This case exemplifies that complex limb-salvage surgery is possible when coordination of care between international and local surgeons is provided with an emphasis on continuity of care post-operatively.

CONCLUSION: En bloc resection with centralization of the ulna remains a viable technique to address aggressive GCTB of the distal radius and can be appropriate in resource-limited settings.

1. Introduction

Cooper and Travers first described giant cell tumor of bone (GCTB) in 1818 [1]. GCTB represents 4–5% of all bone tumors, and 20% of all benign bone tumors [2,3]. It is a benign aggressive tumor most common in young adults between 20 and 40 years of age and more prevalent in females [4]. GCTB typically arises in the metaphysis and extends into the epiphysis [5].

Patients present with symptoms such as pain, swelling, joint effusion and limited range of motion (ROM). Local recurrence is strongly increased by curettage alone or with soft tissue extension, which is present in 20–25% of all GCTB [6]. Treatment options include en bloc resection, curettage with physical and chemical adjuvants, and denosumab [6,7].

We present a case of a recurrent Campanacci Grade II GCTB of the right distal radius in a right-hand dominant female patient from Haiti [8]. The patient was initially evaluated at Hôpital Bernard Mevs in Port au Prince and her surgery was performed at the Hôpital Adventiste d'Haïti in Carrefour. The purpose of this case is to demonstrate that complex limb-salvage surgery is possible in underserved regions of the world when care is coordinated between an international and local team. The importance of adequate follow-up and post-operative care with continued com-

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https://doi.org/10.1016/j.ijscr.2021.105686

Fig. 1. A-C Clinical images of the patient after recurrence of the tumor in pronation (**A**), neutral (**B**) and supination (**C**). Multiple areas of the skin had breakdown, but no tumor was exposed. The volar and ulnar skin was spared.

munication between the teams is essential. This work has been reported in line with the SCARE criteria [9].

2. Presentation of case

A 22-year-old female presented with a recurrent right wrist mass treated surgically three years prior to presentation. She had undergone a curettage resection without adjuvant and placement of iliac crest autograft for a GCTB of the distal radius. The mass had recurred over a period of 2.5 years and was 18 × 25 cm at the time of presentation. The mass involved the majority of the wrist and only the volar and ulnar aspects were spared. There was some superficial skin breakdown, but no tumor was exposed (Fig. 1). The mass was firm, non-mobile and minimally tender. She had full active finger ROM and thumb flexion, but lacked extensor pollicis longus (EPL), abductor pollicis longus (APL) and extensor pollicis brevis (EPB) function (Video 1). Her wrist dorsiflexion was 20 degrees and her palmar flexion was 40 degrees. Her supination was 55 degrees and pronation was 80 degrees. Radial and ulnar deviation was 10 and 15 degrees, respectively. She lacked sensation over her superficial radial nerve, but otherwise had full sensation. She had 2+ radial and ulnar pulses and a normal Allen's test.

Radiographs demonstrated an expansile, lytic lesion of the distal radius with internal trabeculations with a thin cortical rim. There was encasement of the proximal carpus with dislocation of the distal ulna (Fig. 2). Although an MRI was unable to be obtained in Haiti, a CT scan was available which demonstrated that the volar structures were free, whereas the EPL, APL and EPB went directly into the tumor. The extensor tendons to the fingers were traversed through the tumor but were felt to be uninvolved based on the patient's clinical exam. A chest CT was negative for metastatic disease. The patients care was then coordinated with an international team. A biopsy was obtained two weeks after presentation under direction of an orthopaedic oncologist. The specimen was evaluated in the United States and was consistent with recurrent GCTB. One month after the biopsy, an international surgical team traveled to Haiti to perform the definitive surgery.

Limb-salvage surgery was performed by a fellowship-trained orthopaedic oncologist (LZ) and hand surgeon (JD) with over thirty years of combined surgical experience. They both had made multiple trips to Haiti to perform surgery and had previously coordinated post-operative care with the local orthopaedic surgeons. A local Anesthesiologist used a propofol drip to provide general anesthesia and the patient was given 1 g of cefazolin pre-operatively. An en bloc resection of the distal radius with the proximal row of the carpus was performed. The EPL, APL, EPB, flexor carpi radialis, extensor carpi radialis longus and brevis and superficial radial nerve were

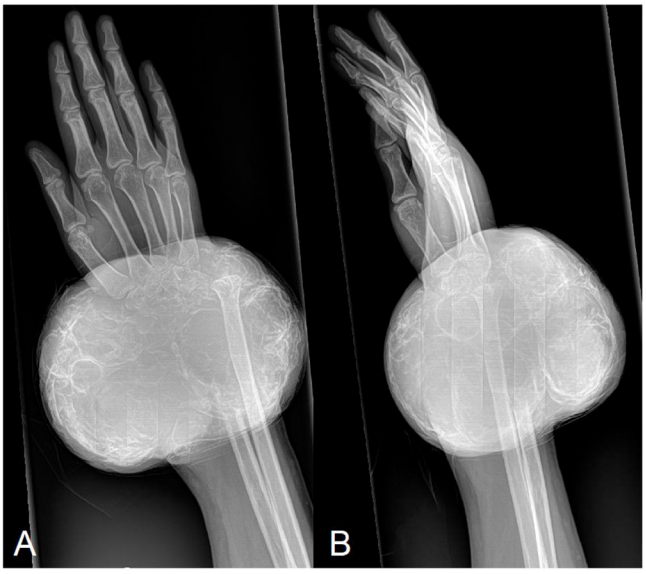

Fig. 2. A-B Initial anteroposterior (**A**) and lateral (**B**) radiographs demonstrating involvement of the distal radius with encasement of the proximal carpus and dislocation of the ulna. The expansile, lytic lesion has multiple internal trabeculations with a thin cortical rim.

resected proximal to the tumor due to tumor involvement. The tumor had grown around the extensor tendons, which were freely mobile and not involved. The tendons were cut distal to the tumor and were easily pulled through the tunnel that had formed for later repair. The volar structures were free of tumor and were preserved, and the tumor was able to be resected en bloc (Fig. 3A). A centralization of the ulna was then performed with fusion to the capitate and third metacarpal using an Acumed (Hillsboro, OR) wrist fusion plate (Fig. 3B). The extensor indicis proprius was transferred to the EPL to restore thumb extension. The remaining tendons were redundant due to being stretched out by the tumor. The flexor tendons were cut, and both the flexor and extensor tendons were shortened and repaired side-to-side using a Krakow suture technique with appropriate tension (Fig. 3C).

The skin was closed with a tension-free repair and a bulky, sterile dressing and volar wrist splint was placed (Fig. 3D). Histopathologic examination in the United States confirmed a GCTB without evidence of malignant transformation and negative margins (Fig. 4).

Passive ROM of all digits was initiated immediately after surgery. At 3 weeks gentle active and active-assisted ROM was allowed and formal rehabilitation therapy started at 2 months. The patient was monitored for recurrence and metastatic spread every three months for the first two years with a physical examination and radiographs of both the forearm and chest. She will then be followed every six months until the fifth year.

At 3 months, the patient lacked 5 degrees of extension of the fingers. She had full flexion of the proximal and distal interphalangeal joints of the fingers but lacked flexion at the thumb interphalangeal joint. She had approximately 5 degrees of flexion at the metacarpophalangeal joints and was able to grip and pinch (Fig. 5A and B). She had 45 degrees of functional supination and 80 degrees of functional pronation by utilizing her shoulder. There was early evidence of consolidation of the fusion and her weight bearing was progressed to as tolerated (Fig. 5C). Two years after surgery the patient had no evidence of recurrent or metastatic disease, was pain free, and had regained full extension of her fingers and maintained her flexion (Video 2). The patient feels her life is back to normal and she is planning on attending school to become a nurse.

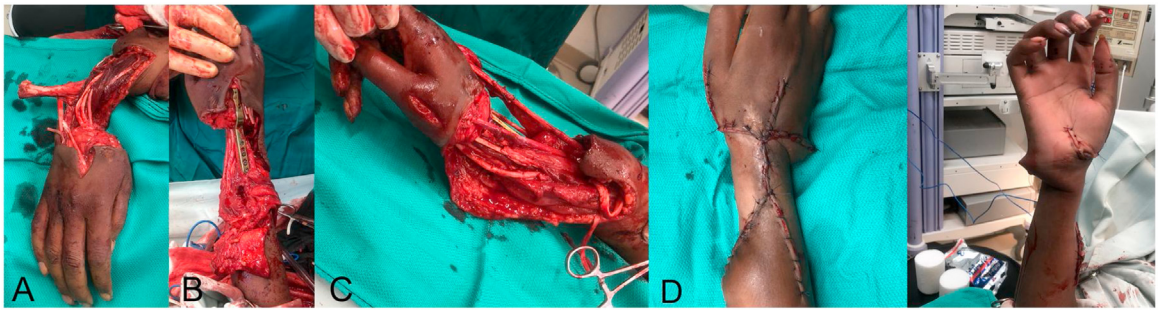

Fig. 3. A-D Intraoperative photos after resection of the tumor but prior to reconstruction (**A**), after centralization of the ulna with placement of a wrist fusion plate (**B**), after reconstruction of the tendons (**C**) and after wound closure (**D**).

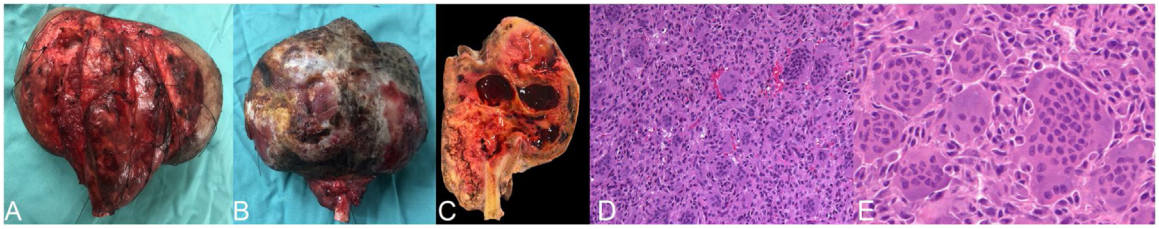

Fig. 4. A-D and 4-E After resection of the tumor, radial and volar (**A**) and dorsal and ulnar (**B**) aspects of the resected specimen are noted with no evidence of exposed tumor. A sagittal view of the tumor during pathologic examination (**C**) demonstrates cystic areas inside the tumor. The cut radius is identified at the bottom of the photos. Low-power (**D**) and high-power (**E**) histopathologic examination of the resected specimen demonstrates multiple multi-nucleated osteoclast-like giant cells in a background of mononuclear cells.

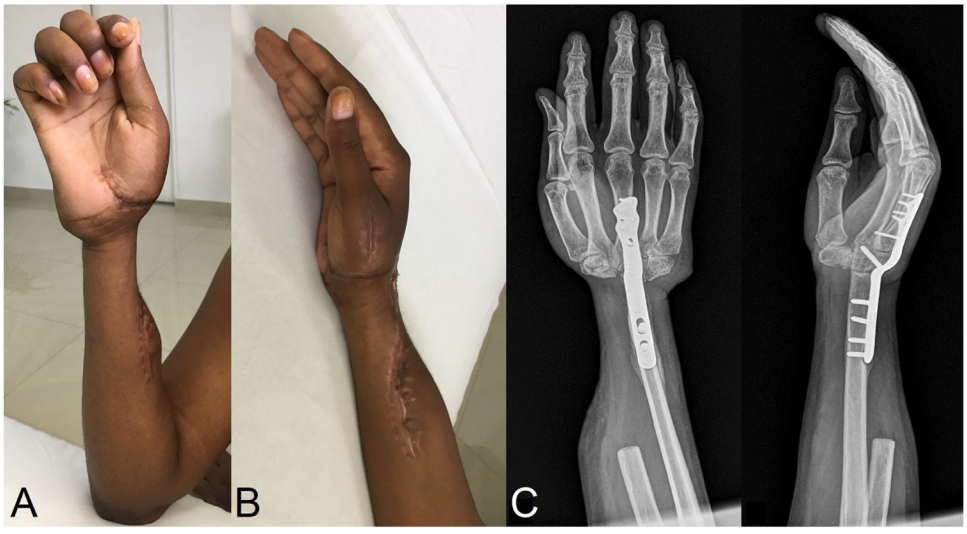

Fig. 5. A-C Three months after surgery the patient had full flexion of her proximal and distal interphalangeal joints of her fingers with the ability to pinch (**A**), and near full extension of her fingers (**B**). Anteroposterior and lateral (**C**) radiographs demonstrate no evidence of recurrent tumor or hardware failure and early consolidation of the fusion.

3. Discussion

GCTB involving the distal radius in underserved regions requires a focused medical decision to ensure the lowest possibility of recurrence and to restore as much function as possible. Surgical techniques include intralesional curettage with use of adjuvants, en bloc resection, and use of denosumab [7,10]. Reconstruction options for an en bloc resection include a vascularized or non-vascularized fibula, iliac crest autograft, and intercalary or osteoarticular allograft [11]. Allograft is currently unavailable in Haiti and use of autograft would increase blood loss and surgical time in this resource-limited environment. Therefore, a translocation or centralization of the ulna would be optimal.

Translocation of the distal ulna into areas of a radial defect was described by Hey-Groves in 1922 for fractures of the distal radius with a severe defect [12]. In 1934, Greenwood [13] and Watson-Jones [14,15] described severing the distal ulna and transferring the ulna shaft to the radius, creating a one-bone forearm. More recently and specifically for recurrent Campanacci Grade III GCTB, Seradge [16] in 1982, and Puri et al. [17] in 2010 described a standard technique for wide resection of the distal radius with translocation of the ulna onto the remaining radius. Bhan et al. [18] and Puri et al. [17] have observed the risk of non-union of the ulnoradial interface with translocation.

Centralization of the ulna to the carpus was first described by Sayre [19] in 1893 and modified by Lidge [20] in 1969. This proce-

dure was initially indicated for Type 3 or Type 4 radial longitudinal deficiencies. More recently Singh et al. [21] described centralization of the ulna for use in Grade III GCTB with good functional results.

In this case, a centralization of the ulna was chosen for reconstruction. The main advantages to this procedure include high rates of union and obviating the risk of non-union at a second site. Additionally, this procedure avoided further dissection and a longer procedure which would be necessary for both an ulnar translocation and autograft reconstruction [12,14,15,22,23]. The main disadvantage is loss of pronation and supination at the wrist by virtue of the ulno-humeral joint lacking rotation. At two years, the patient is able to compensate for her loss of motion through her shoulder and has a good functional result.

4. Conclusion

This case supports other reports that centralization of the ulna is an effective method of reconstruction after en bloc resection of the distal radius due to GCTB. When resources are limited, treatment options should be carefully considered in order to optimize outcomes. With adequate coordination and bidirectional collaboration, complex limb-salvage surgery can be successfully conducted in low- and middle-income countries like Haiti. The importance of continued collaboration and communication with the local team is essential in order to manage complications and provide proper follow-up.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Ethical approval

This case report is exempt from ethical approval at our institution.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Registration of research studies

1. Name of the registry: Research Registry
2. Unique identifying number or registration ID: researchregistry6403
3. Hyperlink to your specific registration (must be publicly accessible and will be checked): https://www.researchregistry.com/browse-the-registry#home/registrationdetails/5fe7e690fe46e3001b6cce62/

Guarantor

Ron Israelski, M.D.

Provenance and peer review

Not commissioned, externally peer-reviewed.

CRediT authorship contribution statement

Ronald Israelski: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing - original draft, Writing - review & editing. **Ted Nnamno Obi:** Supervision, Validation, Visualization, Writing - original draft, Writing - review & editing. **Christian Alexander Pean:** Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing - original draft, Writing - review & editing. **Jean Wildric Hippolyte:** Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing - original draft, Writing - review & editing. **John Durham:** Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing - original draft, Writing - review & editing. **Lee M. Zuckerman:** Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing - original draft, Writing - review & editing.

Declaration of Competing Interest

The authors state that they have no conflicts of interest for this report.

Acknowledgments

“It takes a village” is an appropriate quote when approaching such a case in a resource-limited environment. Without such collaborative efforts, this patient would likely go on to amputation which highlights the importance of humanitarian organizations such as Northern Arizona Volunteer Corp (NAVMC), Orthopedic Relief Services International (ORSI) and our host Hôpital Bernard Mevs in Port au Prince, Haiti, and Adventist Health International (AHI). Thank you to Dr. Andrew Rosenberg for reviewing the pathology and Dr. Scott Nelson for arranging care at the Hôpital Adventiste d’Haïti.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at https://doi.org/10.1016/j.ijscr.2021.105686.

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