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Case Report

Approach to the Treatment of Expansile Metacarpal Aneurysmal Bone Cysts: A Report of Two Cases

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The two cases presented demonstrate the management of aneurysmal bone cysts of the metacarpal, which destroyed the normal bone architecture. Treatment of both cases included wide resection and metacarpal reconstruction with an intercalary fibular allograft. Denosumab use contrasts these two cases and is helpful in reestablishment of a cortical rim for fixation in the absence of a 1-cm margin proximally or distally to preserve the native carpometacarpal and metacarpophalangeal joints. Surgical resection and allograft reconstruction is a viable treatment for expansile metacarpal aneurysmal bone cysts, and neoadjuvant denosumab has utility in creating an ossified margin for fixation.

Aneurysmal bone cysts (ABCs) are benign, locally destructive skeletal tumors that are most commonly found around the knee, with a peak incidence in the first two decades of life.[1] Aneurysmal bone cysts account for approximately 1% to 6% of all primary osseous tumors.[2] These expansile lesions produce blood-filled cavities lined by fibroblasts, trabecular bone, and giant cells.[3] Recent work has demonstrated the role of the ubiquitin-specific protease *USP6 (Tre2)* gene in a subsect of primary ABCs. However, secondary ABCs can occur in conjunction with benign and malignant pathologies, such as chondroblastoma, telangiectatic osteosarcoma, and giant cell tumor of bone.[3]

Primary goals of the treatment of ABCs are to eradicate the lesion, minimize recurrence, and preserve function, with the mainstay of therapy consisting of curettage with possible bone graft supplementation.[1] To minimize recurrence through margin expansion, adjuvant treatments have evolved with argon beam coagulation, cryotherapy, and intralesional phenol.[4] Additionally, neoadjuvant denosumab may have added utility when aggressive resection may dramatically increase local morbidity, such as with ABCs in the metacarpals. Aneurysmal bone cysts in the hand skeleton account for <5% of all ABCs, with most available literature to guide treatment consisting of case reports.[5] Given the rarity of these tumors and the often complete involvement of the bone, optimal treatment methodology is controversial.

This case report, which adheres to the guidelines for case reports (CARE), describes the surgical technique and outcomes of management of a pediatric metacarpal ABC with and without the use of neoadjuvant denosumab. Both patients/guardians provided consent for the collection of personal health data and submission for publication.

Case Reports

Case 1

Patient A is a 12-year-old right handed otherwise healthy girl who presented to another facility with an enlarging, tender right hand mass for 1 year. Examination revealed a prominent 3 × 4 cm dorsal mass with an associated rotation of the ring finger and proximal interphalangeal (PIP) joint extension lag. Initial computed tomography and magnetic resonance imaging demonstrated an expansile lesion in the fourth metacarpal with extreme cortical thinning and fluid-fluid levels (Figs. 1–3). Prior to referral, the patient had an incisional biopsy that demonstrated an ABC with evidence of giant cell tumor of bone components.

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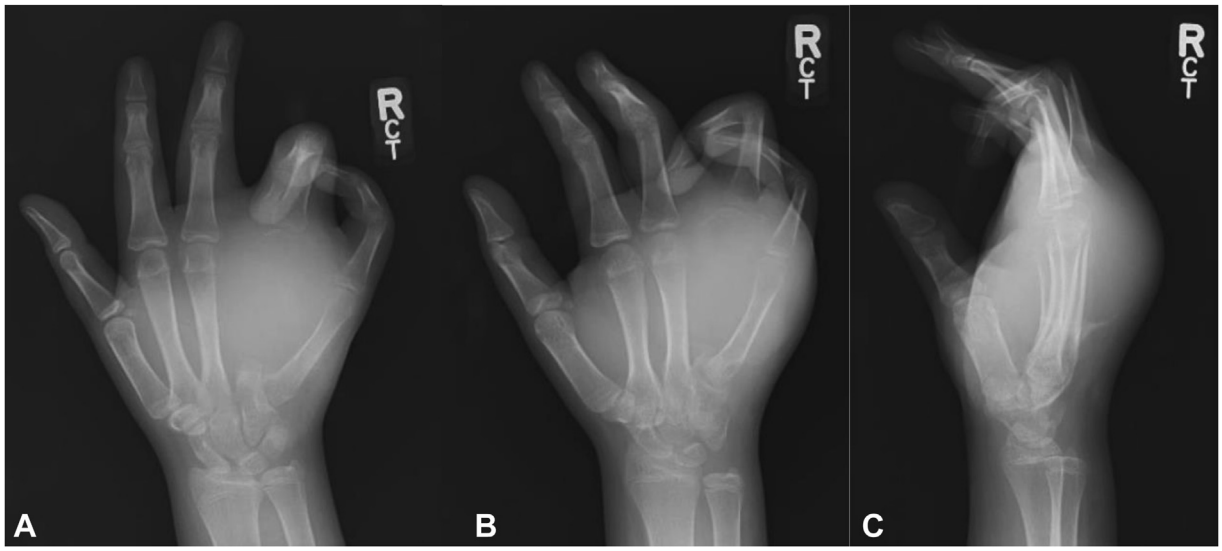

Figure 1. Initial posteroanterior, oblique, and lateral radiographs demonstrating expansile lytic lesion with effacement of the cortical margin.

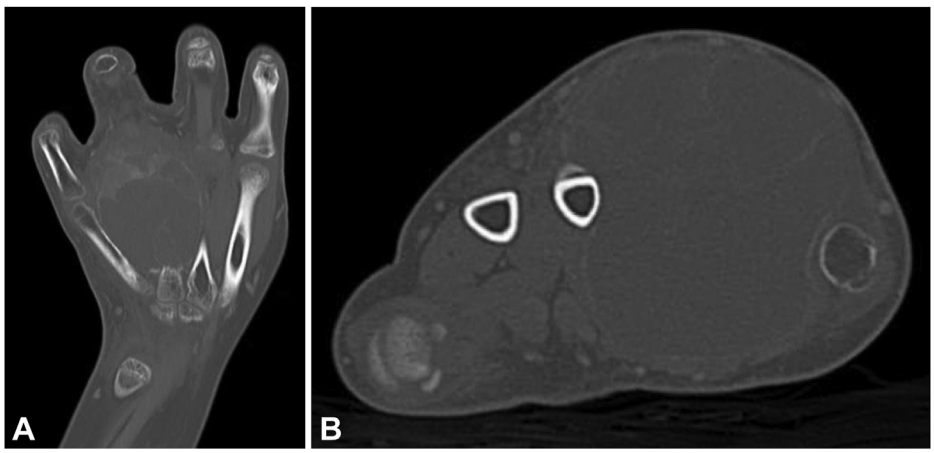

Figure 2. Coronal and axial cuts of a computed tomography scan demonstrating an expansile lesion encompassing the entirety of the fourth metacarpal with near-complete erosion of the cortical margins.

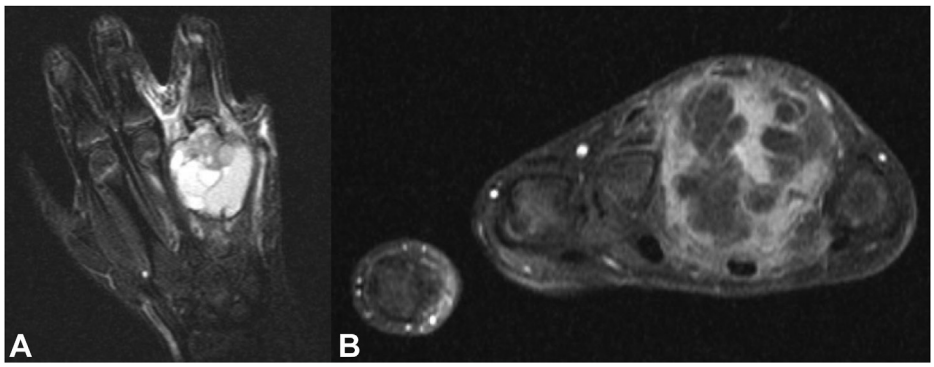

Figure 3. **A** Coronal sections of a short tau inversion recovery sequence depicting a fourth metacarpal lesion with fluid-fluid levels. **B** Axial sections of a T1 fat-suppressed sequence with evidence of an expansile heterogeneous mass with mixed hyper- and hypointense signals.

After biopsy analysis, the patient was referred to our institution for further evaluation and management. Given the lack of cortical margin and concomitant giant cell tumor of bone components, the patient was indicated for neoadjuvant denosumab to decrease the overall tumor volume and calcify the lesion to facilitate resection. The patient received 120 mg denosumab subcutaneously every 4

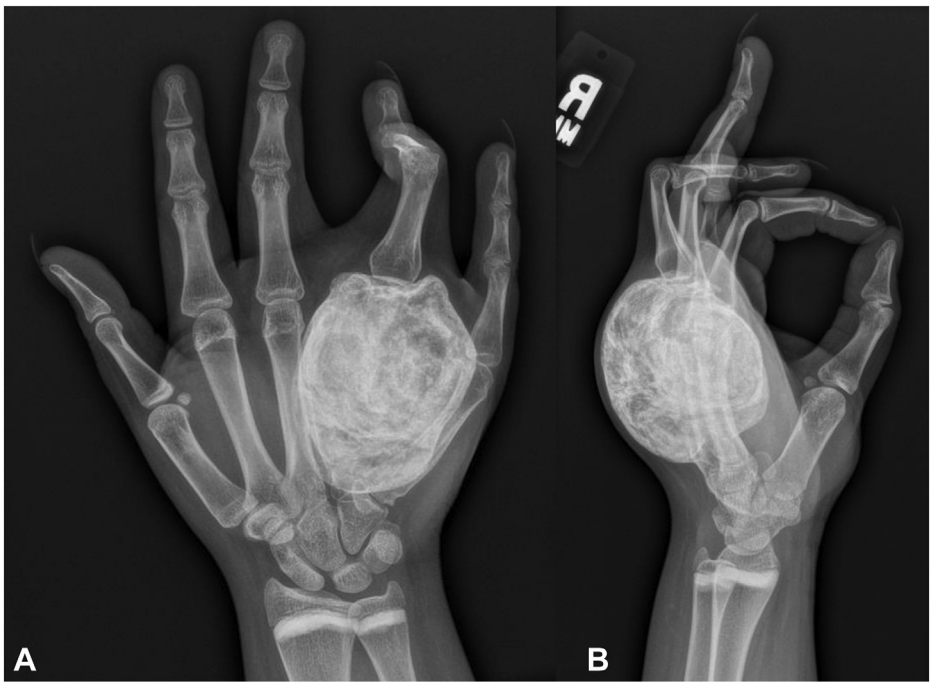

Figure 4. Posteroanterior and lateral radiographs after 12 months of denosumab treatment with interval development of lesional calcifications and defined border measuring 6.3 × 5.4 × 5.6 cm.

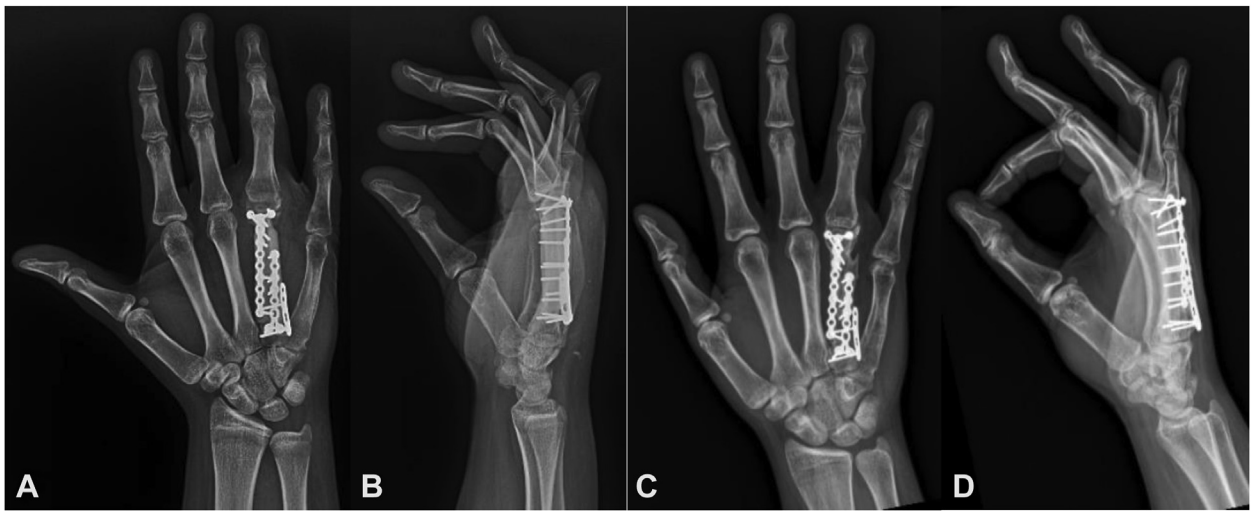

Figure 5. A, B Patient A's immediate posteroanterior and lateral postoperative radiographs demonstrating resection of the calcified mass with fibular strut reconstruction. **C, D** The 3.5-year postoperative radiographs with stable hardware and congruent ring finger MCP joint with evidence of volar subluxation of the ring finger metacarpal head.

weeks for 12 months, which resulted in considerable lesional calcification with a well-defined border that was more amenable to resection and reconstruction (Fig. 4).

Surgical technique

A dorsal, longitudinal incision centered over the mass was used for the approach. After dissection and tenolysis of the extensor digitorum communis tendon, the fourth metacarpal was exposed. Subperiosteal dissection was carried out circumferentially; however, completion of a subperiosteal volar dissection was not possible. As such, the mass was divided and removed piecemeal, leaving 12 mm of the calcified rim proximally and distally for fixation. After removal, the remaining calcified mass was contoured to resemble the metacarpal head and base. A 3-mm round bur was used to achieve margin expansion and create a medullary canal in the proximal and distal segments for placement of the fibular allograft. Two 1.5 mm-locking T-plates were used proximally and distally to secure the allograft with an additional 1.5-mm straight locking plate along the dorsal-ulnar surface. A closed manipulation was then performed to address the ring finger PIP flexion contracture, which yielded full passive extension of the joint. Due to the chronic mass effect of the tumor, the extensor digitorum communis tendon to the ring finger was noted to be excessively lax. This was addressed by shortening the tendon with imbrication sutures to approximate the length of the bone. At the end of the

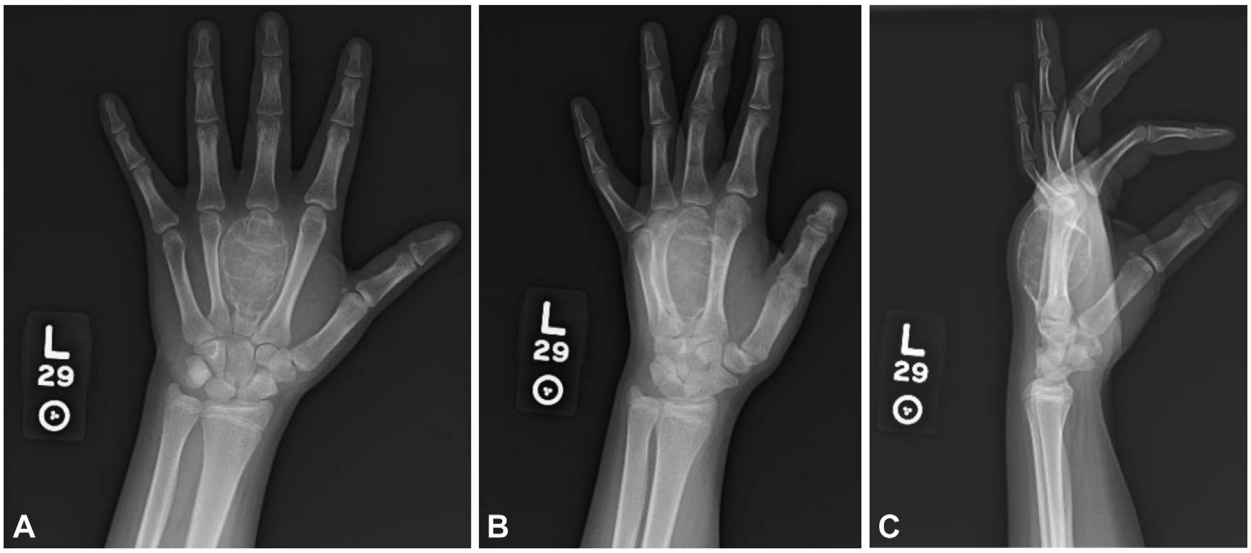

Figure 6. Presenting posteroanterior, oblique, and lateral radiograph of the left hand demonstrating a lytic third metacarpal lesion with cortical expansion and thinning measuring 4.5 × 2.9 × 3.3 cm.

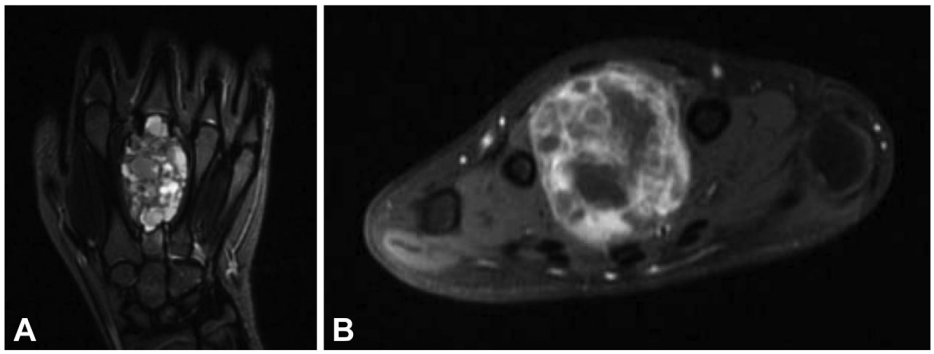

Figure 7. **A** Coronal sections of a short tau inversion recovery sequence depicting a third metacarpal lesion with fluid-fluid levels. **B** Axial sections of a T1 fat-suppressed sequence demonstrating the tumor with mixed hyper- and hypointense signals.

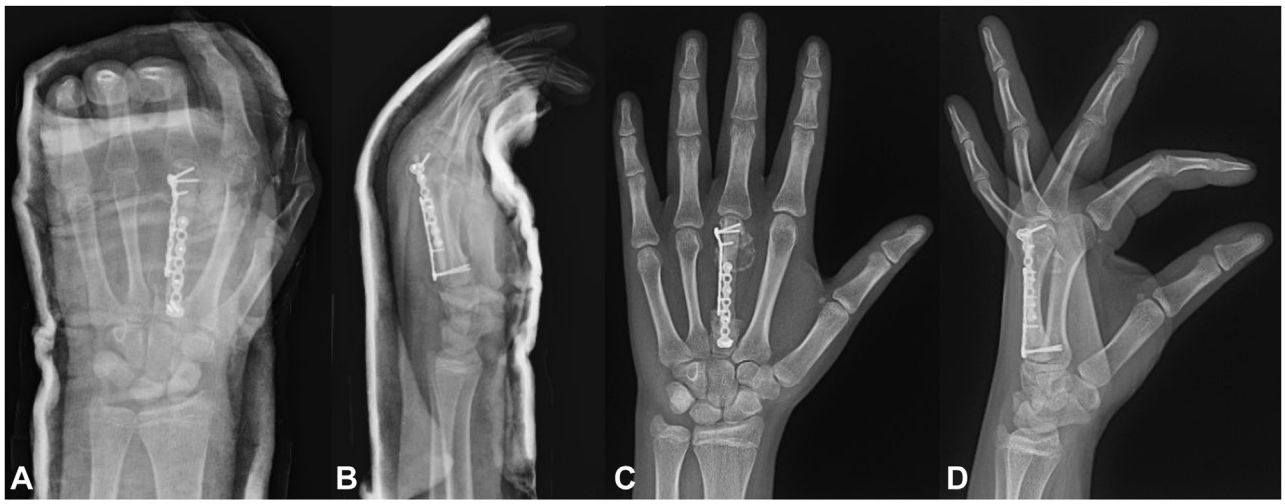

Figure 8. **A, B** Patient B's immediate posteroanterior and lateral postoperative radiographs demonstrating resection of the mass with fibular strut reconstruction. **C, D** The 3-month postoperative radiographs with stable hardware and congruent middle finger MCP and carpometacarpal joints.

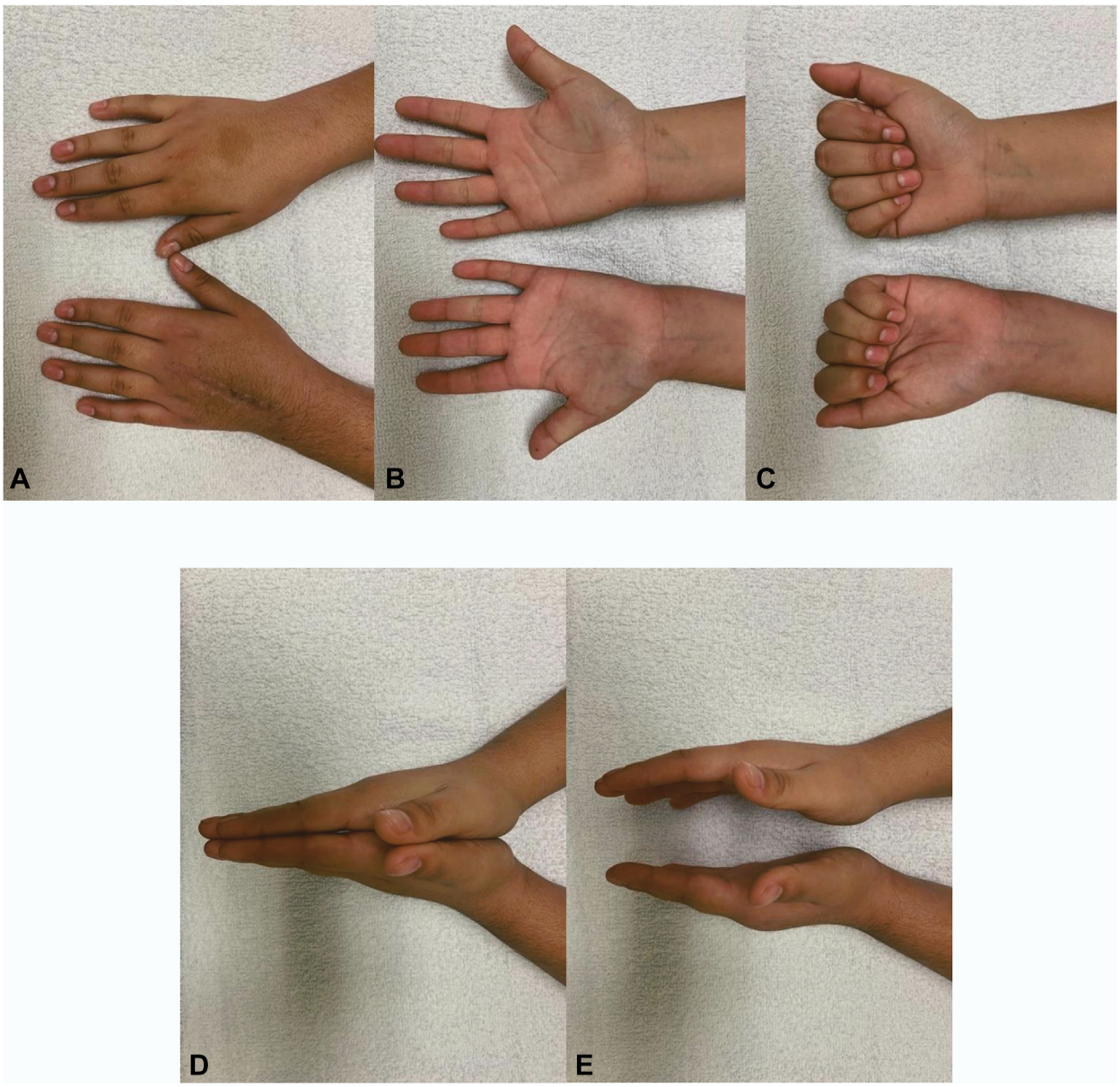

Figure 9. Patient B's 3-month postoperative clinical photographs. Patient B has regained full extension without lag and can make a composite fist without evidence of scissoring or malrotation.

procedure, all fingers were noted to have full passive flexion and extension with a normal resting cascade.

Postoperative course

After surgery, the patient was placed in a short arm splint for 3 weeks. After the splint was removed, the patient was noted to be pain free and began working with hand therapy on active and passive range of motion exercises. At 29 months after surgery, the surgical digit was noted to have motion at the distal interphalangeal and PIP joints from 0° to 90°. However, the metacarpophalangeal (MCP) joint had a limited range of motion from 0° to 30°, which was independent of the wrist position that was likely attributable to limited extensor excursion from tendon adhesions and capsular contracture. Latest radiographic follow-up at 41 months after surgery demonstrated intact hardware with volar subluxation of the MCP joint (Fig. 5).

Case 2

Patient B is a 12-year-old right handed otherwise healthy female student who initially presented with an enlarging left dorsal hand mass for 5 months, with associated pain at night. Examination revealed a 2.5 × 3—cm firm, tender dorsal hand mass centered over the third metacarpal. All extensor and flexor tendons were intact; however, the patient had marked limitation in extension of her index, middle, ring, and little fingers due to obstruction from the mass and limited middle finger flexion.

Dedicated radiographs demonstrated an expansile, lytic lesion of the third metacarpal with associated cortical thinning (Fig. 6). Magnetic resonance imaging demonstrated a cystic lesion with fluid-fluid levels suggestive of an ABC (Fig. 7). Given the radiographic and magnetic resonance imaging evidence of ABC, the decision was made to proceed with open biopsy, tumor resection, and allograft reconstruction.

Surgical technique

Dissection was completed in a similar manner as described above. Once benign pathology was confirmed with an intraoperative frozen section, the incision was extended, and a subperiosteal dissection was carried out circumferentially. The mass was incised proximally and distally, leaving 1 cm of bone stock for the carpometacarpal and MP joints, respectively. The remaining shell of bone was curetted, and a 3-mm round bur was used to create a trough for the fibular allograft and perform margin expansion. The fibular allograft was contoured to the appropriate size and secured proximally and distally in a similar manner described above.

Postoperative course

After surgery, patient B was placed in a short arm ulnar gutter splint for 1 week. At that time, the patient began working on finger range of motion with hand therapy, focusing on MCP extension. At the most recent follow-up 6 months after surgery, the patient had no significant pain with stable radiographic appearance of the hardware (Fig. 8). The patient was able to make a composite fist with 10° of middle finger MCP extension lag, which improved to 5° with the PIP joint held in extension (Fig. 9). The patient was able to complete all activities of daily living without limitation. A relative motion splint was prescribed to aid with MCP extension.

Discussion

Metacarpal ABCs remain a rare, vexing challenge for hand and oncology surgeons. Treatment must balance the need for tumor eradication with maintenance of local architecture to preserve function. Given the high rate of recurrence after curettage and grafting, as well as the extreme deformity of the bone that can be caused by such expansile lesions, there has been recent interest in en-bloc resection to decrease local recurrence rates.[6] When resection of metacarpal ABCs has been employed, there have been no reported recurrences.[5,7] Additionally, outcomes from these studies have shown consistent improvement in pain and function, which was corroborated in this case report.[5,7] Both patients had complete resolution in their pain with improvement in their function, despite mild extensor lag, without evidence of recurrence at the latest follow-up.

The role of adjuvant treatments in the management of ABCs is expanding because there is increased literature demonstrating decreased morbidity and local recurrence.[4,8,9] Application of these treatments is limited when dealing with the small metacarpals near neurovascular structures and minimal overlying soft tissue. Recent literature has demonstrated increased complications for metacarpal ABCs, such as wound breakdown, neurovascular injury, and complex regional pain syndrome.[3,5] However, use of these techniques is important in the achievement of margin expansion if there is concern for any residual tumor cells.

Denosumab functions in the receptor activator of NF-κB and receptor activator of the NF-κB ligand (RANKL) pathway through competitive inhibition of RANKL. There are reports of off-label denosumab use in the management of ABCs, given their increased NF-κB and RANKL expressions.[9] By inhibiting RANKL, denosumab alters the bone deposition–resorption cycle to allow for increased lesional ossification that creates an ossified rim without killing pathologic stromal cells.[10] As such, complete eradication of the tumor through resection versus curettage and margin expansion is critical if surgery is pursued. With patient A, denosumab therapy enabled bone preservation adjacent to the MCP and carpometacarpal joints for fixation, in conjunction with margin expansion, to allow for the preservation of these joints in the patient's reconstruction (Fig. 4).

Surgical resection and reconstruction with fibular strut allograft is a viable treatment option for expansile metacarpal ABCs to decrease local recurrence and improve function. Denosumab as a neoadjuvant treatment may allow for preservation of the carpometacarpal and MCP joints in resection of widely expansile lesions by creating an ossified rim for fixation. Nevertheless, this technique relies on appropriate adjuvant treatment and margin expansion to ensure successful oncologic treatment, as denosumab does not eradicate pathologic cells.

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