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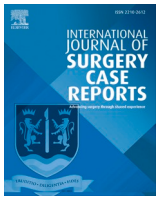

Case report

Tell-tale of a primary and recurrent giant cell tumour of distal ulna: A report of two cases

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ABSTRACT

Introduction: Wrist functionality is severely hampered by giant cell tumours (GCT) of the distal ulna, which require careful surgical treatment. A thorough preoperative evaluation and precise tumour grading are essential for developing a treatment plan that maximises functional results while maintaining oncological control.
Case presentation: We present 2 cases of distal end ulna giant cell tumour (GCT). The first case involves a primary GCT in a young male, treated with marginal excision and extensor carpi ulnaris tenodesis, with no recurrence over three years. The second case describes a recurrent GCT in a middle-aged male, initially treated with curettage and bone cement in primary stage, followed by a successful marginal excision after recurrence.
Discussion: Distal ulna giant cell tumours (GCTs) are uncommon, violent lesions that have a significant chance of recurring, particularly in Campanacci grade III instances. Stabilisation procedures such as ECU tendon reconstruction address post-resection issues such as discomfort, limited forearm rotation, and grip weakness, although en bloc resection is favoured to minimise recurrence.
Conclusion: Both cases highlight the challenges and considerations in managing distal ulna GCTs, emphasizing that en-bloc resection may reduce recurrence rates compared to curettage in grade III Campanacci GCTs.

1. Introduction

The definition of a giant cell tumour (GCT) of bone is an intramedullary bone tumour that is composed of mononucleated cells and multinucleated giant cells that resemble osteoclasts and have an unpredictable and variable propensity for growth. GCTs are distinctive in their age and location [1]. Roughly 5 % of primary bone tumours are GCTs. Despite being regarded as benign bone tumours, GCTs have a high recurrence rate [2,3]. In 90 % of GCT cases, the epiphyseal position is as expected. A tumour frequently touches the cartilage or even the articular subchondral bone. Seldom are the joint and/or its capsule penetrated. When skeletally immature patients experience GCT, it happens seldom, and the lesion is usually located in the metaphysis [4,5]. The distal femur, proximal tibia, distal radius, and sacral are the most frequently occurring sites, in decreasing order. Giant cell tumours are extremely uncommon to occur in the distal ulna, with a reported incidence of 0.45–3.2 % [6,7]. Clinical presentations that are typical include discomfort and swelling in the affected soft tissues, as well as mechanical challenges and pathological fractures brought on by osteolytic bone lesions. There are proponents of a variety of surgical procedures, from broad excision to intralesional curettage. The purpose of treatment is to remove the tumour as much as possible while maintaining limb function and preventing both local and distant metastases. In the past ten years, a number of adjuvant techniques that go beyond basic curettage have been documented in the orthopaedic literature to improve local control and stop recurrences. For restoring limb function in distal ulna, there have been numerous methods described, for example, flexor carpi ulnaris tenodesis, extensor carpi ulnaris tenodesis, lasso tenodesis with palmaris longus graft, and pronator quadratus interposition stump. In this report of two cases, we present a primary GCT and a recurrent GCT of distal ulna and discuss about the management.

Our work has been reported in line with the SCARE Guidelines 2023 criteria [8].

2. Case one

A male patient in his early 20s presented to our tertiary specialist unit with swelling and pain in left distal forearm for the past 5 months. He had history of swelling over the distal forearm characterised by a gradual progression and associated pain.

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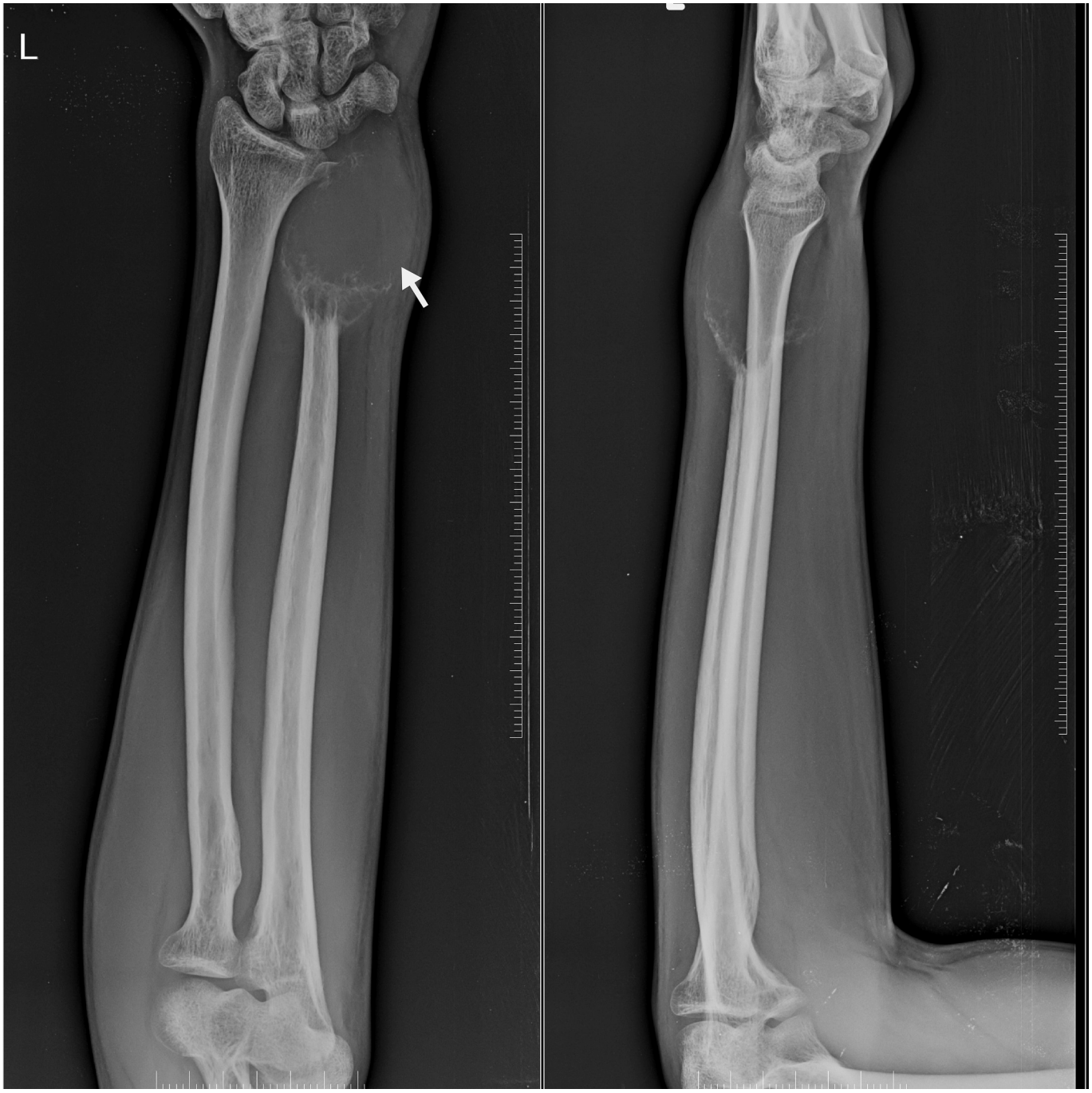

Fig. 1. Preoperative radiograph of wrist with forearm showing Subarticular lytic expansile lesion with a "soap bubble appearance" (arrow) affecting the distal ulna in the epiphysis and metaphysis.

On examination, there was an ovular swelling measuring 8 × 5 × 5 cm over the left distal forearm. The swelling displayed a firm consistency with irregular surfaces and was tender on palpation. The overlying skin exhibited no swelling, sinus or ulcer. Distal neurovascular structures were found to be intact. He underwent radiological investigations indicated a GCT of the distal ulna and the diagnosis subsequently confirmed by histopathological examination of core needle biopsy tissue.

2.1. Investigations

The plain radiographs revealed an eccentric, expansive, epiphysial lytic lesion affecting the distal third of the ulna (Fig. 1). Magnetic resonance imaging (MRI) showed heterogenous space occupying lesion in distal end of ulna with destruction of epimetaphyseal region appearing intermediate to hypointense in T1 weighted images while intermediate to hyperintense in T2 weighted and fat suppressed images without involvement of muscles, extensor and flexor tendons and major neurovascular structures (Fig. 2). Metastatic workup was done and no distant metastasis was found. Core needle biopsy showed scattered multinucleated giant cells and mononuclear cells. The mononuclear cells are plump, oval to spindled, have vesicular chromatin and conspicuous nucleoli (Fig. 3).

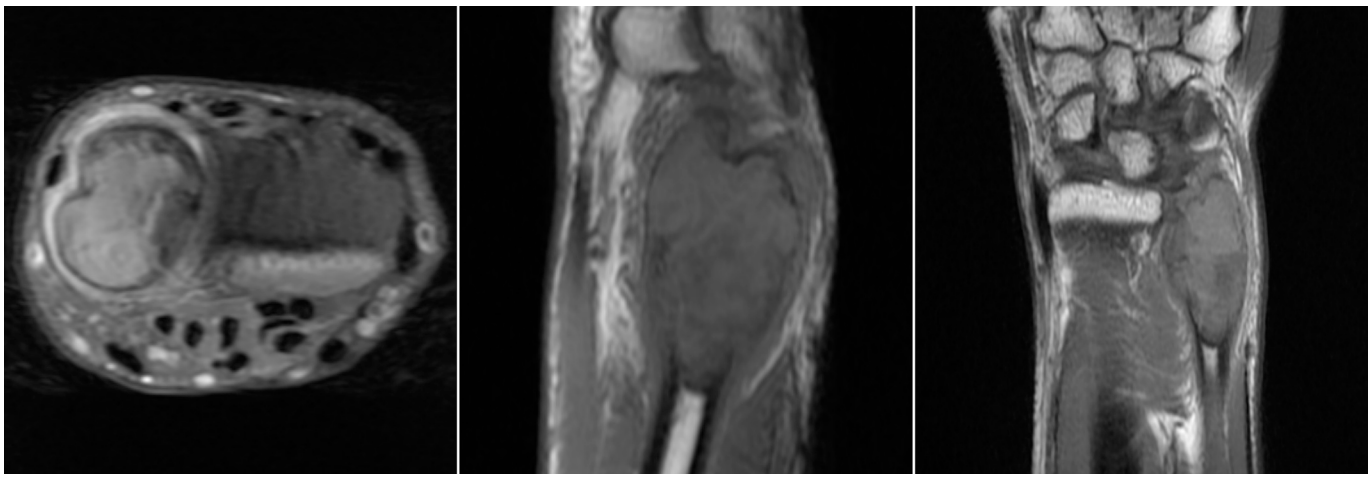

Fig. 2. Magnetic resonant images showing proton density (PD) fat suppressed (FS) axial section, T1 weighted sagittal and T1 weighted coronal section of distal ulna (from left to right).

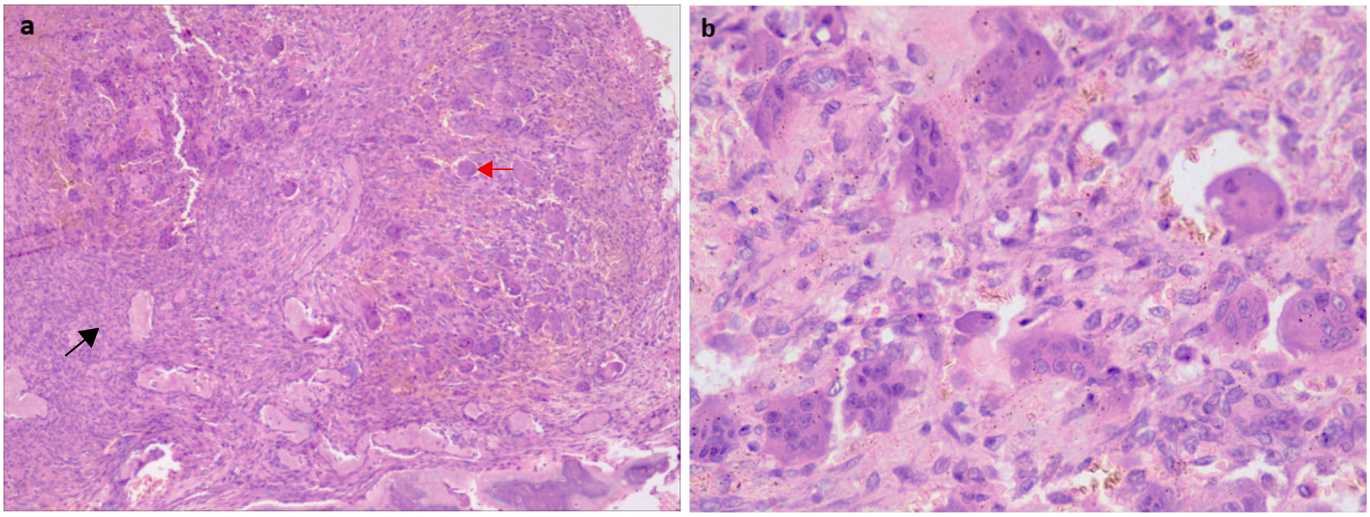

Fig. 3. Histopathological section showing scattered osteoclast-like multinucleated giant cells (red arrow) and mononuclear stromal cells (black arrow) (a) scanner view (H&E ×40) (b) high power view (H&E ×400). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

2.2. Treatment

Following the histopathological confirmation of the diagnosis a comprehensive plan was devised for the en-bloc resection of the tumour. The surgical approach was conducted through dorsomedial approach on the ulna. On exploration, the tumour was measuring approximately 7 × 4.8 × 3 cms in size, displaying an irregular surface with areas of haemorrhage, and exhibiting a soft to firm consistency. The patient underwent marginal excision and extensor carpi ulnaris tenodesis. Post marginal excision, extensor carpi ulnaris (ECU) tendon was dissected from base of 5th metacarpal up-to ulna osteotomy site and split in half. Dissected half of the tendon passed through a drill hole at ulna osteotomy stump with forearm in full supination and was sutured back to itself and remaining half of intact ECU tendon. Intraoperatively, negative margins were confirmed with frozen section histopathological examination.

The postoperative period transpired without any complications. Postoperative radiograph of left wrist (Fig. 4). Histopathological examination of excised tissues confirmed the presence of GCT with neoplastic mononuclear stromal cells and marked nuclear pleomorphism with scattered osteoclast-like giant cells.

2.3. Outcome

The patient was discharged on the 5th day post-surgery, and during the follow-ups, incision exhibited satisfactory healing. Patient resumed routine daily activities within 2 months post-surgery. Patient attained full range of motion at wrist with unrestricted supination and pronation. Plain radiographs were conducted during the follow-up visits, revealing no signs of recurrence. Completed 3 years of follow up (Fig. 5). Patient wrist function score was calculated as per Disabilities of the Arm, Shoulder and Hand (DASH) score and was 0.8 at 3-year follow-up.

3. Case two

A male patient in his mid-40s presented to our tertiary specialist unit with recurrent swelling and pain in left distal forearm for the past 4 months. He was an elsewhere operated case of GCT of distal ulna 2 years ago. Initially, he had a history of swelling over the distal forearm 2 years ago, characterised by a gradual progression and an absence of associated pain. Seeking medical attention elsewhere, radiological investigations indicated a GCT of the distal ulna and the diagnosis subsequently confirmed by histopathological examination of core needle biopsy

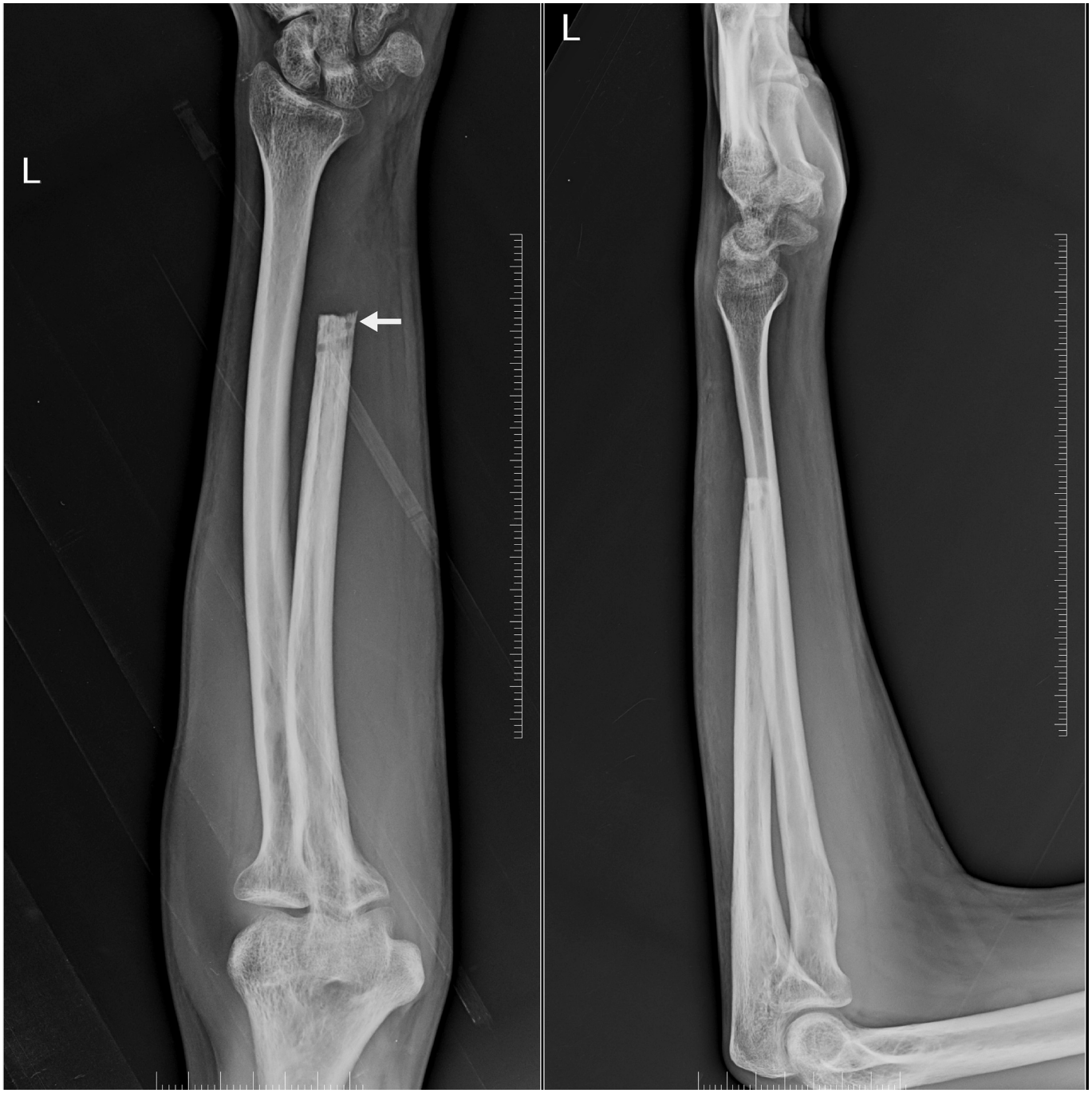

Fig. 4. Postoperative radiograph after excision of the lesion in anteroposterior and lateral view of forearm with wrist. Extensor carpi ulnaris tenodesis labelled as arrow.

tissue. He underwent extended curettage and bone cement placement. Following surgery, the patient resumed his routine activities and remained asymptomatic for the subsequent year. However, after 2 years, he observed swelling in the same region where the surgical excision had taken place earlier. Accompanied by pain, he sought consultation at our hospital for further evaluation. On examination, there was an ovular swelling measuring 10x5x5 cm over the left distal forearm. The swelling displayed a firm consistency with irregular surfaces and was tender on palpation. The scar from the previous surgery appeared healthy, and the overlying skin exhibited no swelling, sinus or ulcer. Distal neurovascular structures were found to be intact.

3.1. Investigations

Plain radiographs of the recurrent lesion, taken on the patient's presentation to our hospital, revealed the curetted end of the ulna with an irregular matrix-forming growth extending towards the wrist joint with bone cement in-situ (Fig. 6). MRIs showed heterogenous space occupying lesion in distal end of ulna with destruction of epi-metaphyseal region appearing intermediate to hyperintense in T2 weighted with hypointense bone cement on T2 without involving significant neurovascular structures (Fig. 7). Metastatic workup was done and no distant metastasis was found. Core needle biopsy showed scattered multinucleated giant cells and mononuclear cells, findings consistent with GCT.

Fig. 5. Follow up clinical images of functional activity of the limb.

3.2. Treatment

Following the histopathological confirmation of the diagnosis. The surgical approach was conducted through the previous dorsomedial scar on the ulna. On exploration, the tumour was measuring approximately 7x4x3 cm in size, displaying an irregular surface with areas of haemorrhage, and exhibiting a soft to firm consistency. The patient underwent marginal excision and extensor carpi ulnaris tenodesis (Fig. 8). Post marginal excision, extensor carpi ulnaris (ECU) tendon was dissected from base of 5th metacarpal up-to ulna osteotomy site and split in half. Dissected half of the tendon passed through a drill hole at ulna osteotomy stump with forearm in full supination and was sutured back to itself and remaining half of intact ECU tendon. The patient tolerated the surgery well, and the postoperative period transpired without any complications (Fig. 9). Histopathological examination of excised tissues confirmed the presence of many evenly distributed and uniformly scattered multinucleated osteoclastic giant cells with interspersed numerous mononuclear stromal cells.

3.3. Outcome

The patient was discharged on the 5th day post-surgery, and during the follow-up appointments, the incision exhibited good healing. Patient resumed routine daily activities within 3 months post-surgery with full range of motion at wrist with unrestricted supination and pronation. Plain radiographs were conducted during follow ups, revealing no signs of recurrence up-to 1 year (Fig. 10). Patient wrist function score was calculated as per DASH score and was 1.7 at 1 year follow-up.

4. Discussion

GCTs are typically benign neoplasms that originate in the epi-metaphyseal region of skeletally mature patients. They account for approximately 5 % of all primary bone tumours [2]. Research indicates that the majority of cases in Asia are in men, with a gender ratio of 1.27 to 1.77 [9–12].

The most frequently affected sites include the distal femur, proximal tibia and distal radius. According to available reports, the incidence of GCTs in the distal end of the ulna is estimated to range between 0.45 % and 3 % of all GCTs [6,7].

Three grades were established for GCT radiology by Campanacci et al. [1]. Tumours having a well-marginated rim of mature bone are classified as grade I. Grade II cancers lack a distinct radiopaque rim, but their growth is permeative and unrestricted by the production of reactive bone, while grade III tumours have permeative growth and an extended rim produced by the combination of the cortex and reactive bone. Because the tumour in this instance lacks clear borders and extends into soft tissue, both the cases classified as grade III GCT.

Distal ulna GCT is an uncommon condition for which there is no accepted therapeutic method [13]. High risk of failures after wide resection due to the ulnar stump was displaced to dorsal (winging) and converge towards the radius, reduced grip strength and tortional strength [14,15]. This inevitably leads to handgrip weakness, persistent pain, and limitation of forearm rotation [13,16]. Stabilisation using the Extensor Carpi Ulnaris (ECU) tendon after ulnar resection was originally described by Goldner & Hayes in 1979 [16,17]. Reconstruction with ECU tenodesis might be more stable in cases of shorter resections where longer ulnar stumps are still present. As soft tissue reconstruction necessitates providing ulnar stabilisation for better functional outcomes,

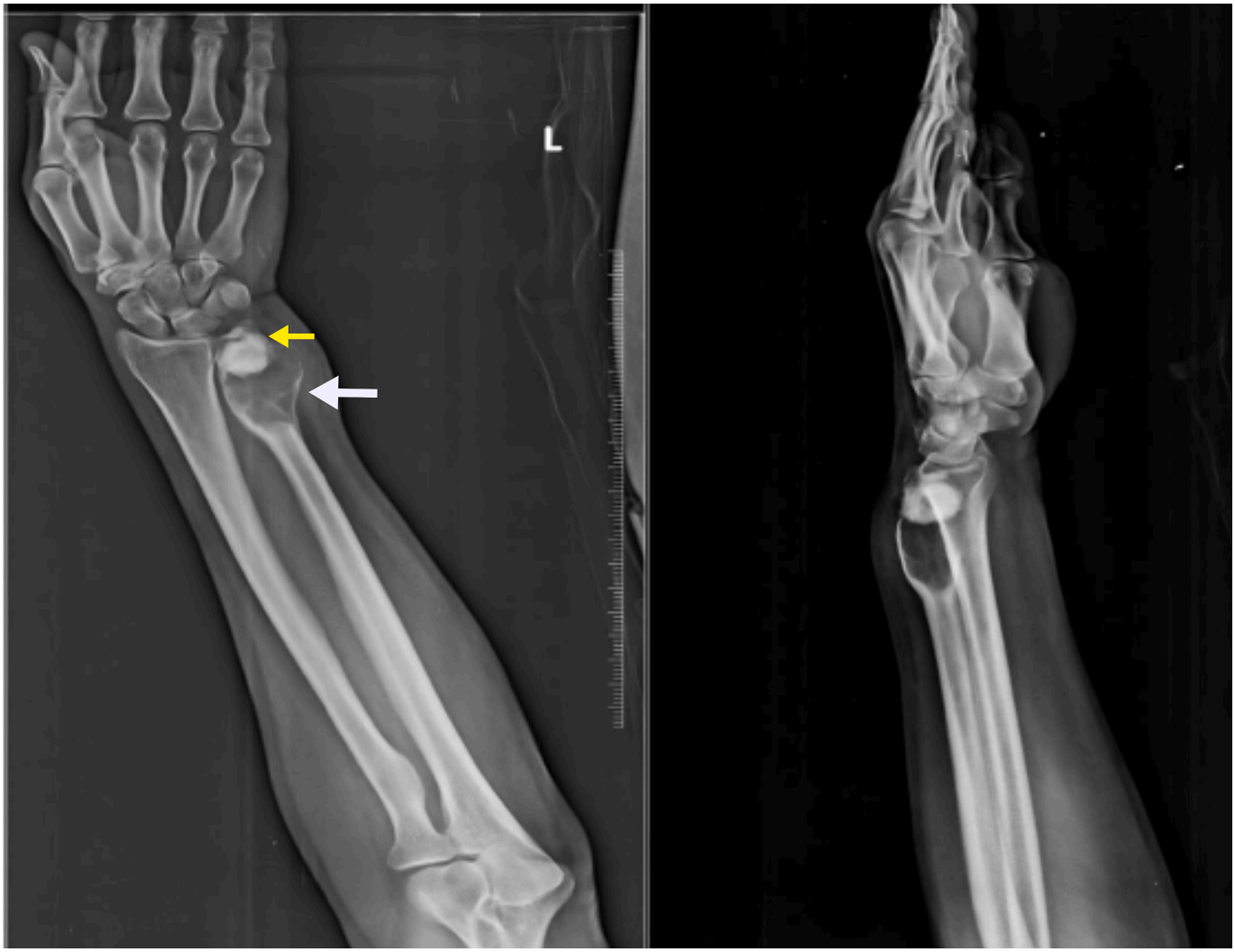

Fig. 6. Preoperative radiograph of wrist with forearm showing lytic, expansile lesion (white arrow) in distal ulna with bone cement in-situ (yellow arrow) anteroposterior and lateral projection. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

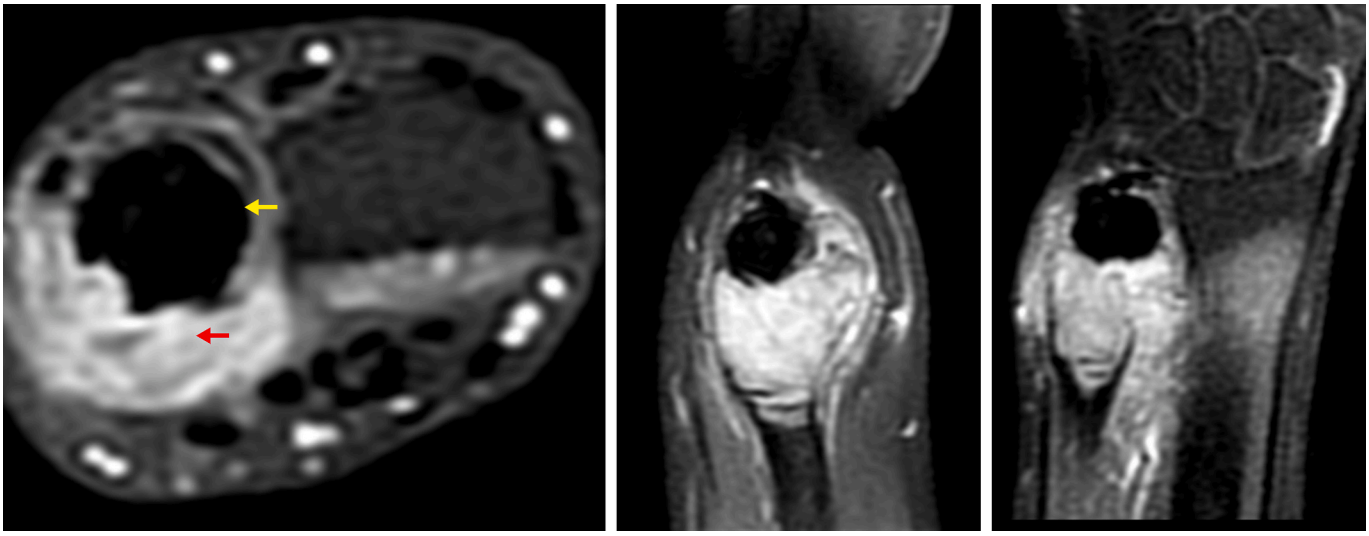

Fig. 7. MRI showing T1 weighted axial section showing lesion (red arrow) in distal ulna and bone cement (yellow arrow), T1 weighted sagittal section and T1 weighted coronal section (from left to right). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

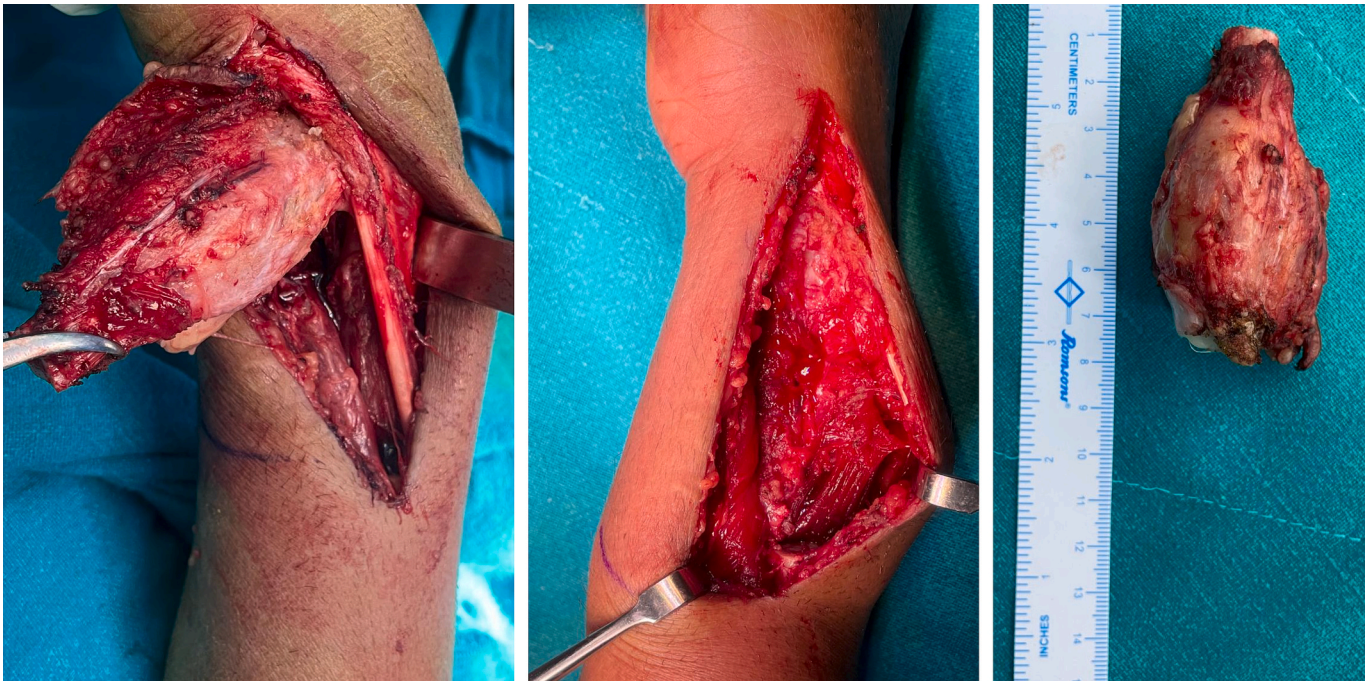

Fig. 8. Intraoperative clinical photographs showing complete excision of giant cell tumour of distal ulna appearing as lobulated mass.

many of the biomechanical studies do not show any clear advantage [18,19].

For Campanacci grade III distal ulnar GCTB, there is no proven treatment; however, en bloc resection has historically been the empirically chosen method to prevent tumour recurrence. In the past 10 years of published studies of distal ulna GCT [20–30], 20 cases of grade III Campanacci were studied, of which 19 underwent resection and 1 underwent curettage. There was recurrence documented in 3 cases out of 19 resection cases (15.7 %), and 100 % recurrence in the curettage case.

Looking at the recurrence following curettage in grade III Campanacci, this suggests that this is not a good modality. The recurrence in resection patients was on the lower side.

5. Patient perspective

- Case one: When I first noticed the pain and swelling, I was worried but never imagined it could be a tumour. Learning the diagnosis was overwhelming, but the doctors explained everything clearly, which helped ease my fears. Though the treatment was challenging, I always felt supported by my medical team. After surgery, I was relieved to hear the tumour was completely removed. Recovery has been tough, but with the care from my doctors and physiotherapy, I'm gradually regaining my strength. I'm thankful for the regular follow-ups showing no signs of recurrence, and I feel hopeful and optimistic about the future.
- Case two: I was devastated when the pain and swelling returned after my initial surgery, and learning it had turned into cancer was terrifying. But the doctors here explained everything clearly, and though the treatment were tough, I felt supported. After surgery, I was relieved to hear the tumour was gone. Recovery wasn't easy, but with my doctors' care and physiotherapy, I'm getting stronger. I'm grateful for the regular check-ups showing no recurrence, and overall, I'm satisfied and hopeful for the future.

6. Take home message

- Although they are uncommon, distal ulna giant cell tumours (GCTs) have a high recurrence rate, particularly when curettage is the only treatment used.
- Effective care of distal ulna GCTs requires a multidisciplinary strategy that includes precise diagnosis by radiographic and histological investigation as well as a comprehensive metastatic workup.
- After resection, it is essential to stabilise the ulnar stump with methods like extensor carpi ulnaris tenodesis in order to preserve hand and forearm function and avoid issues like ulnar stump winging.
- En bloc resection is typically chosen over curettage for grade III Campanacci GCTs of the distal ulna because of its reduced recurrence rate.

CRediT authorship contribution statement

Prabodh Kantiwal: Project administration, Manuscript review and editing, operating surgeon (First author)

Aakash Kumar Choudhary: Manuscript original draft preparation, figure editing (Corresponding author)

Sumit Banerjee: Manuscript proof-reading, Supervision (Third author)

Abhay Elhence: Manuscript proof-reading, Supervision (Fourth author)

Informed consent

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

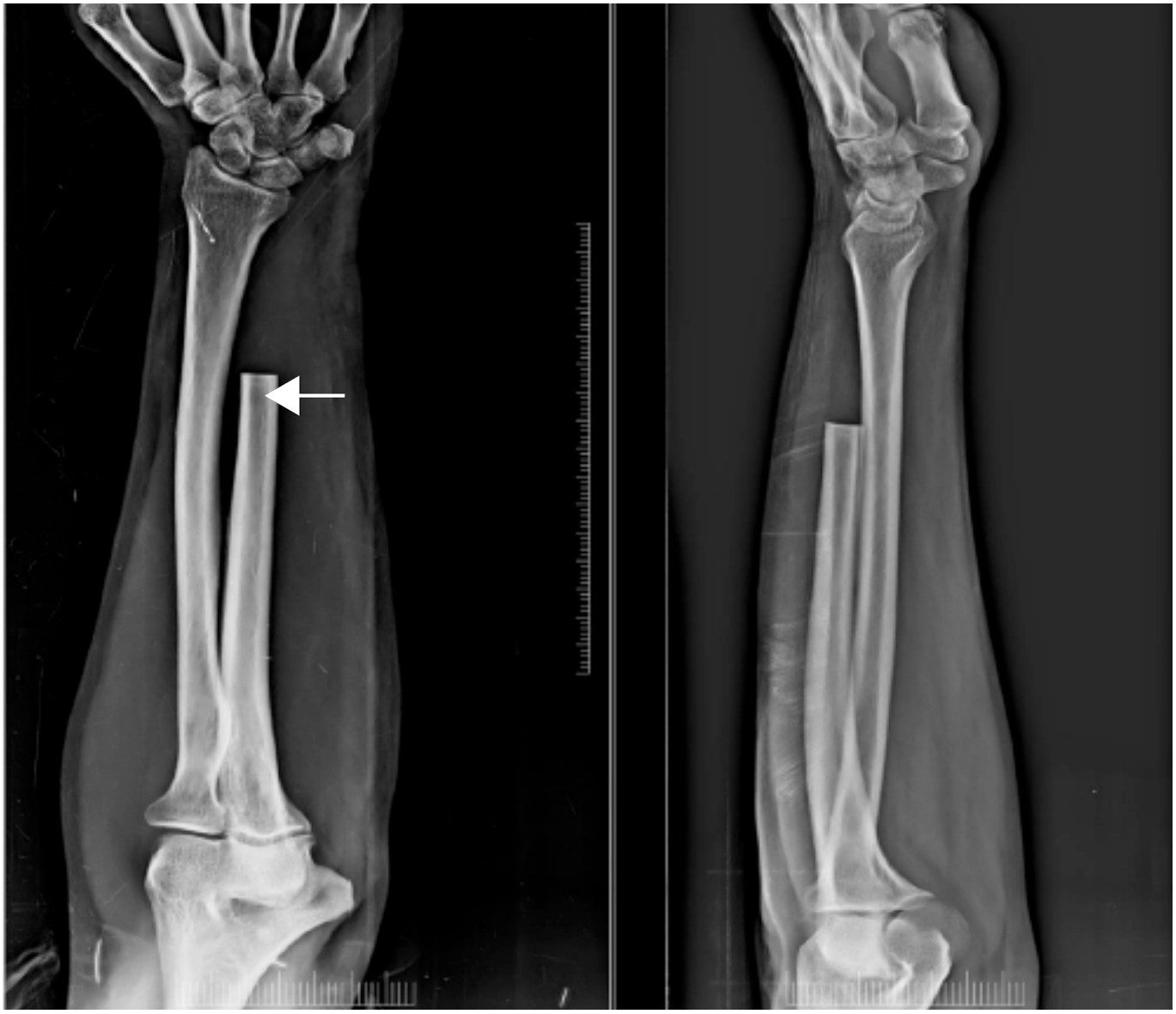

Fig. 9. Postoperative radiograph of forearm with wrist in anteroposterior and lateral projection. Extensor carpi ulnaris tenodesis labelled as arrow.

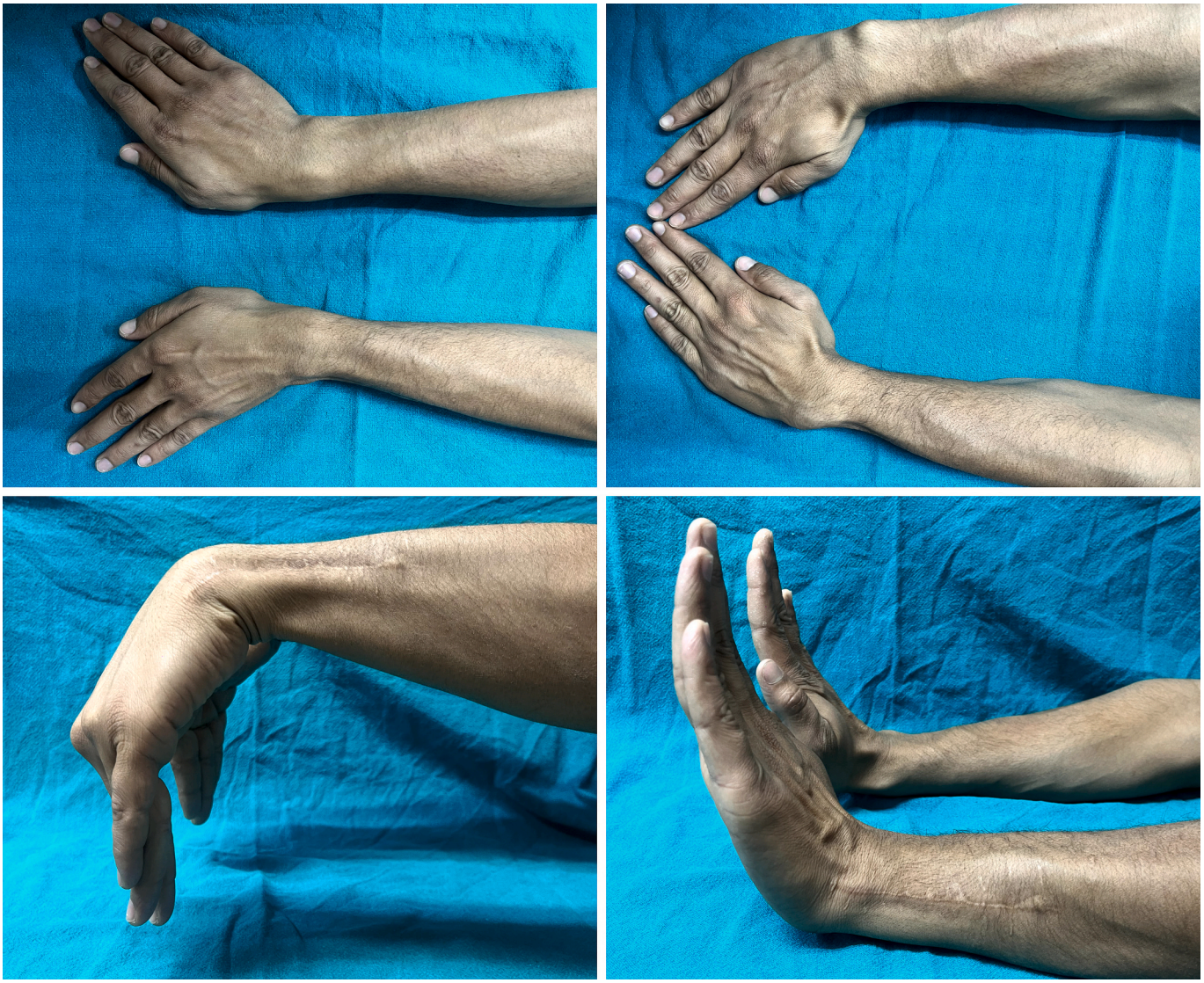

Fig. 10. Follow up clinical images of functional activity of the limb.

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.

Ethical approval

This study did not require ethical approval as it is a case report. However, informed consent has been obtained and can be provided on request.

Guarantor

(First author) Prabodh Kantiwal, Associate Professor, MBBS, MS (Orthopaedics), Department of Orthopaedics, All India Institute of Medical Sciences (AIIMS), Marudhar Industrial Area, 2nd phase, M.I.A. 1st phase, Basni, Jodhpur, Rajasthan, 342005, E-mail address: prabodhkantiwal@yahoo.com

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Declaration of competing interest

None. Authors declare that they have no conflict of interest.

References

[1] M. Campanacci, N. Baldini, S. Boriani, A. Sudanese, Giant-cell tumor of bone, JBJS 69 (1) (1987 Jan 1) 106–114.
[2] ECKARDT JJ, GROGAN TJ. Giant cell tumor of bone. Clin. Orthop. Relat. Res. (1976–2007). 1986 Mar 1;204:45–58.
[3] P.J. McGrath, Giant-cell tumour of bone: an analysis of fifty-two cases, The Journal of Bone & Joint Surgery British Volume. 54 (2) (1972 May 1) 216–229.
[4] J.C. Hoeffel, M.A. Galloy, Y. Grignon, P. Chastagner, J. Floquet, L. Mainard, R. Kadiri, Giant cell tumor of bone in children and adolescents, Rev. Rhum. Engl. Ed. 63 (9) (1996 Oct 1) 618–623.
[5] H.N. Shih, R.W. Hsu, F.H. Sim, Excision curettage and allografting of giant cell tumor, World J. Surg. 22 (5) (1998 May) 432–437.
[6] K.A. Raskin, J.H. Schwab, H.J. Mankin, D.S. Springfield, F.J. Hornicek, Giant cell tumor of bone, JAAOS-Journal of the American Academy of Orthopaedic Surgeons. 21 (2) (2013 Feb 1) 118–126.
[7] D. Vanni, A. Pantalone, E. Andreoli, P. Caldora, V. Salini, Giant cell tumor of the distal ulna: a case report, J Med Case Reports 6 (2012 Dec) 1–5.

[8] C. Sohrabi, G. Mathew, N. Maria, A. Kerwan, T. Franchi, R.A. Agha, The SCARE 2023 guideline: updating consensus surgical CAse REport (SCARE) guidelines, Int J Surg Lond Engl. 109 (5) (2023) 1136.
[9] R. Gupta, V. Seethalakshmi, N.A. Jambhekar, S. Prabhudesai, N. Merchant, A. Puri, M. Agarwal, Clinicopathologic profile of 470 giant cell tumors of bone from a cancer hospital in western India, Ann. Diagn. Pathol. 12 (4) (2008 Aug 1) 239–248.
[10] T. Yanagawa, H. Watanabe, T. Shinozaki, K. Takagishi, Curettage of benign bone tumors without grafts gives sufficient bone strength: a case-series of 78 patients, Acta Orthop. 80 (1) (2009 Jan 1) 9–13.
[11] C.R. Reddy, P.S. Rao, K. Rajakumari, Giant-cell tumors of bone in South India, JBJS 56 (3) (1974 Apr 1) 617–619.
[12] T. Marugame, K. Katanoda, T. Matsuda, Y. Hirabayashi, K.I. Kamo, W. Ajiki, T. Sobue, The Japan cancer surveillance report: incidence of childhood, bone, penis and testis cancers, Jpn. J. Clin. Oncol. 37 (4) (2007 Apr 1) 319–323.
[13] J. Kumar, V. Vijayachandran, E. Dinesh, T. Manickam, Giant cell tumour of the distal ulna: a rare presentation, Malaysian Orthopaedic Journal. 5 (2) (2011) 44–46.
[14] E.J. Bieber, R.L. Linscheid, J.H. Dobyns, R.D. Beckenbaugh, Failed distal ulna resections, J. Hand Surg. Am. 13 (2) (1988 Mar 1) 193–200.
[15] J. Field, R.J. Majkowski, I.J. Leslie, Poor results of Darrach's procedure after wrist injuries, The Journal of Bone & Joint Surgery British Volume. 75 (1) (1993 Jan 1) 53–57.
[16] M. Singh, S. Sharma, C. Peshin, I.H. Wani, A. Tikoo, S.K. Gupta, D. Singh, Wide resection and stabilization of ulnar stump by extensor carpi ulnaris for giant cell tumor of distal ulna: two case reports, Cases J. 2 (2009 Dec) 1–6.
[17] E.H. Kayias, G.I. Drosos, G.A. Anagnostopoulou, Resection of the distal ulna for tumours and stabilisation of the stump. A case report and literature review, Acta Orthop. Belg. 1;72(4):484 (2006 Aug).
[18] M.S. Petersen, B.D. Adams, Biomechanical evaluation of distal radioulnar reconstructions, J. Hand Surg. Am. 18 (2) (1993 Mar 1) 328–334.
[19] M. Sauerbier, R. Berger, M. Fujita, M. Hahn, Radioulnar convergence after distal ulnar resection mechanical performance of two commonly used soft tissue stabilizing procedures, Acta Orthop. Scand. 74 (4) (2003 Jan 1) 420–428.
[20] K. Jamshidi, M. Bahrabadi, A. Bagherifard, M. Mohamadpour, Surgical treatment outcome of giant cell tumor of distal ulna: En bloc resection vs. curettage and bone graft. Medical journal of the Islamic Republic of, Iran 32 (2018) 44.
[21] I.D. Papanastassiou, O.D. Savvidou, G.D. Chloros, P.D. Megaloikonomos, V. A. Kontogeorgakos, P.J. Papagelopoulos, Extensor carpi ulnaris tenodesis versus no stabilization after wide resection of distal ulna giant cell tumors, Hand 14 (2) (2019 Mar) 242–248.
[22] D. Von Borstel, A Taguibao R, A Strle N, E Burns J. Giant cell tumor of the bone: aggressive case initially treated with denosumab and intralesional surgery. Skeletal radiology. 46 (2017 Apr) 571–578.
[23] S.Y. Park, M.H. Lee, J.S. Lee, J.S. Song, H.W. Chung, Ossified soft tissue recurrence of giant cell tumor of the bone: four case reports with follow-up radiographs, CT, ultrasound, and MR images, Skeletal Radiol. 43 (2014 Oct) 1457–1463.
[24] K.B. Ferguson, M. Jane, A. Mahendra, Giant cell tumour of the distal ulna: is reconstruction required after excision of the distal third of the ulna? Journal of Hand Surgery (European Volume). 41 (7) (2016 Sep) 771–772.
[25] I. Gracia, I.R. Proubasta, L. Trullols, A. Peiró, E. Moya, S. Cortés, O. Buezo, J. Majó, Distal radioulnar joint prosthesis for the treatment of giant cell tumor of the distal ulna: a case report and literature review, Strategies in trauma and limb reconstruction. 6 (2011 Aug) 103–106.
[26] N.F. Jones, D.J. Graham, Radical resection of a recurrent giant cell tumor of the distal ulna and immediate reconstruction with a distal radio-ulnar joint implant arthroplasty, Hand 15(5):NP68-72 (2020 Sep).
[27] D. Kotrych, A. Żyluk, I. Walaszek, Reconstruction of the distal radioulnar joint with a prosthesis after excision of a recurrent giant-cell tumour of the distal ulna, Journal of Hand Surgery (European Volume). 36 (5) (2011 Jun) 428–430.
[28] D. Kotrych, A. Żyluk, I. Walaszek, Reconstruction of the distal radioulnar joint with a prosthesis after excision of a recurrent giant-cell tumour of the distal ulna, Journal of Hand Surgery (European Volume). 36 (5) (2011 Jun) 428–430.
[29] A. Thiounn, E. Guerre, C. Szymanski, C. Maynou, C. Fontaine, Two cases of distal ulna giant cell tumors treated by extensive resection, prosthetic reconstruction and stabilization using the brachioradialis tendon, Hand surgery and rehabilitation. 36 (6) (2017 Dec 1) 419–422.
[30] A. Sanchez-Pareja, F. Larousserie, S. Boudabbous, J.Y. Beaulieu, N. Mach, E. Saiji, A.L. Rougemont, Giant cell tumor of bone with pseudosarcomatous changes leading to premature denosumab therapy interruption: a case report with review of the literature, Int. J. Surg. Pathol. 24 (4) (2016 Jun) 366–372.