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Pathological fracture of the humerus due to hydatid disease: A rare osseous presentation and staged reconstructive management

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Abstract
Hydatid disease of bone is a rare manifestation of *Echinococcus granulosus* infection, with humeral involvement being exceptionally uncommon. We present the case of a 24-year-old male who developed a pathological fracture of the right humerus following trivial trauma. The fracture was initially stabilized elsewhere, but the patient later presented with persistent pain and purulent discharge. Imaging revealed osteolytic changes, and histopathology confirmed a hydatid cyst. The patient underwent staged surgical management, including debridement, implant removal, cyst excision, reconstruction with a non-vascularised fibular graft, and definitive fixation with a long locking plate. Albendazole therapy was given throughout the course. This case highlights the importance of considering hydatid disease in the differential diagnosis of unexplained lytic lesions or pathological fractures, particularly in endemic areas, and the role of staged reconstruction in achieving successful outcomes.

Keywords: Hydatid cyst, pathological fracture, external fixator, non-vascularised fibula bone grafting, albendazole

Introduction
Hydatid cyst is a parasitic infection resulting from the larval stage of the tapeworm Echinococcus, most commonly *Echinococcus granulosus* [1]. Common Sites: Liver - 60-70%, Lungs - 20-30%, Spleen, kidney, brain, muscle, heart and rarely in bone.
Bony hydatidosis, a rare form of echinococcosis, occurs when the larval cysts of *Echinococcus granulosus* invade bone tissue, leading to progressive, destructive, and infiltrative lesions that often resemble neoplastic or infective processes. It represents approximately 0.5% to 4% of all cases of hydatid disease. The spine is the most commonly affected site, involved in nearly 50% of skeletal cases, followed by the pelvis, femur, humerus, tibia, ribs, and skull [2, 3].
Hydatid involvement of the humerus typically manifests as a gradual onset of dull, localized pain in upper arm. As the lesion enlarges, patients may notice a painless or mildly tender swelling, often mistaken for a benign tumour or chronic infection. Limitation of shoulder motion can occur when the cyst is located in the proximal humerus due to mechanical obstruction or pain. If left untreated, the expanding cyst may lead to cortical erosion, leading the patient to pathological fractures. Systemic features like fever or malaise are usually absent, except in cases of secondary infection or rupture [4].

Case Study
A 24-year-old male sustained a fracture of the shaft of the right humerus following a trivial trauma. The fracture was initially managed surgically with stabilization using 3-4 titanium elastic nails (TENs) along the shaft of the humerus.
Approximately 3-4 months post-injury and surgery, the patient presented to our facility with complaints of persistent right arm pain and purulent discharge from the proximal arm region.
Radiographic evaluation revealed significant osteolysis of the humeral shaft with the implants in situ, suggestive of progressive bone destruction.

The patient was subsequently admitted, thoroughly investigated, and planned for surgical debridement and biopsy of the involved area.

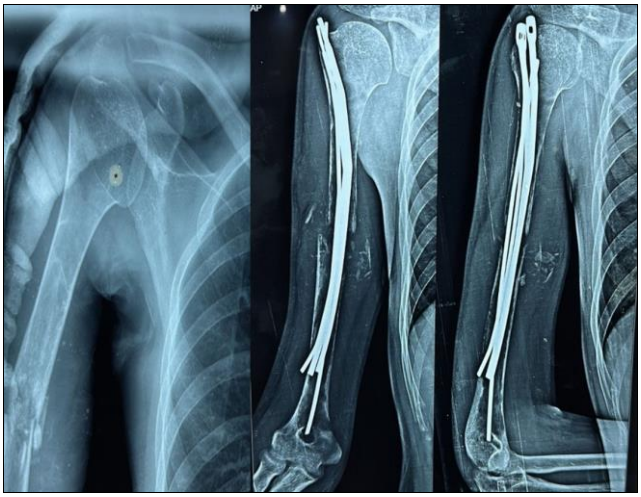

Fig 1: Preoperative fracture of shaft of humerus and fixation of fracture with Enders nails.

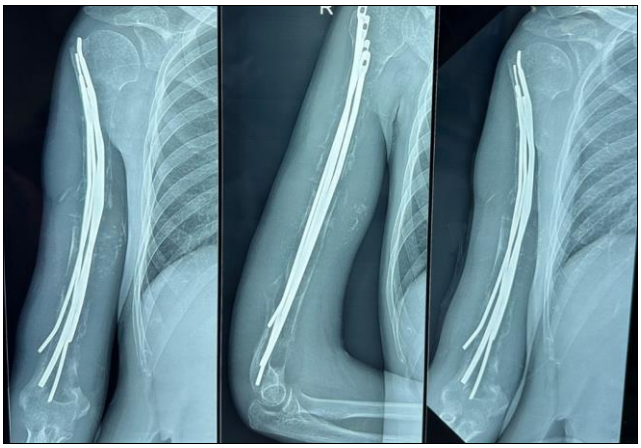

Fig 2: X-ray appearance of patient when he first presented to us after 3-4 months of injury and surgery done outside.

Management and Operative Findings

The patient was taken to the operating theatre for initial surgical intervention. Intraoperatively, thorough debridement of the affected area was performed. On exploration, a significant amount of purulent collection was noted within the humeral shaft, along with the presence of multiple translucent vesicles characteristic of hydatid cysts. Samples were obtained and sent for histopathological examination and microbiological culture and sensitivity.

Histopathology confirmed the diagnosis of osseous hydatid disease (*Echinococcus granulosus*). Post-operatively, the patient was managed with intravenous antibiotics, analgesics, and initiated on oral Albendazole therapy. He was discharged in stable condition with close follow-up.

Second Stage surgery - Debridement and Reconstruction

Approximately one month later, a second-stage surgery was planned and performed. The procedure involved:

- Extensive debridement of the residual cystic lesion and infected tissue,
- Removal of previously placed implants along with the involved segment of the humeral shaft,
- Complete excision of cysts and cyst wall,
- Distraction of the humeral ends using Titanium Elastic Nailing (TEN),
- Insertion of a cement spacer impregnated with antibiotics.
- Stabilization with a rail-type external fixator.

The patient tolerated the procedure well. The following intra-operative and post-operative clinical images illustrate the extent of the lesion, surgical steps, and fixation construct.

Fig 3: Intraoperative picture of debrided material showing some vesicle and cystic like consistency along with cyst wall extraction

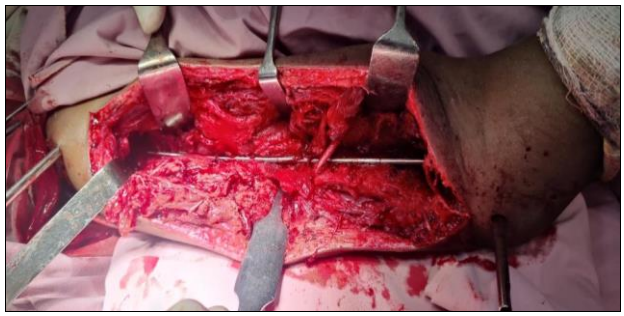

Fig 4: Picture showing placement of titanium elastic nail after shaft removal keeping two ends of humerus distracted.

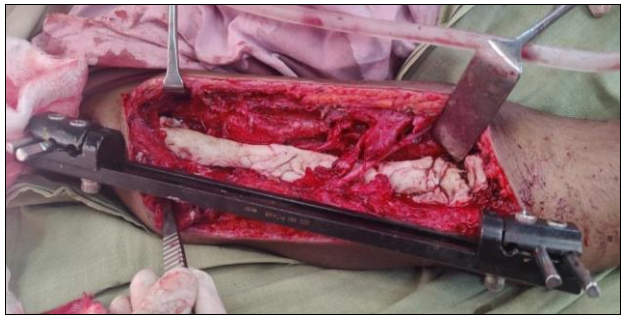

Fig 5: Showing coating of titanium elastic nail with bone cement containing antibiotics and stabilization with rail external fixator.

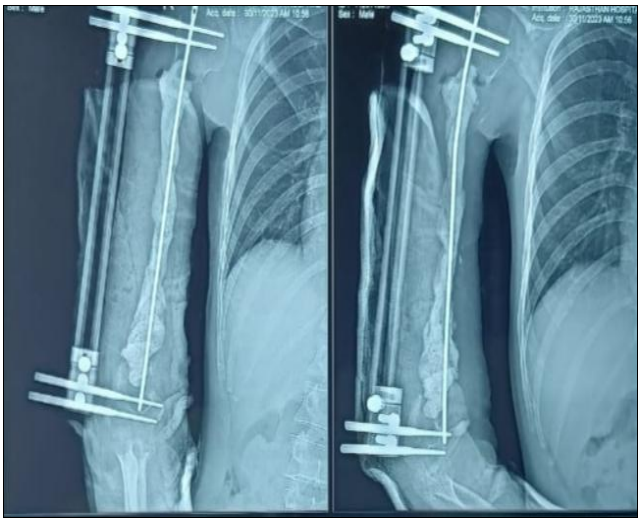

Fig 6: Immediate post-operative x-ray after second stage of surgery showing antibiotic impregnated cement special along with external fixation of the bone.

Postoperatively, the patient received intravenous antibiotics, analgesics, and anti-helminthic therapy in the form of oral Albendazole. Daily sterile wound dressings were carried out under supervision. The patient showed gradual improvement and was discharged after one week, with instructions for continued Albendazole therapy and regular outpatient wound care.

Third Stage surgery- Biological Reconstruction of Humeral Defect

At two months post-surgery, the patient was re-admitted for the planned third-stage reconstructive procedure. During this surgery:

- Thorough wound debridement was performed once again to ensure eradication of any residual infection.
- The previously inserted TEN nails and cement spacer were carefully removed.
- A non-vascularised fibular graft was harvested and inserted into the segmental bone defect between proximal and distal ends of humerus.
- The construct was stabilized using Titanium Elastic Nailing (TEN) through the fibular graft to maintain alignment and length.
- Additional autologous cancellous bone grafting was performed at both the proximal and distal interfaces to enhance biological healing and construct stability.
- A rail-type external fixator was reapplied to maintain the alignment and facilitate bone regeneration.

Fig 7: Showing harvested non-vascularised fibula auto graft.

Postoperative X-rays demonstrated proper placement of the fibular graft, satisfactory alignment, and stability of the fixation. The patient was monitored closely for graft incorporation, infection control, and limb function.

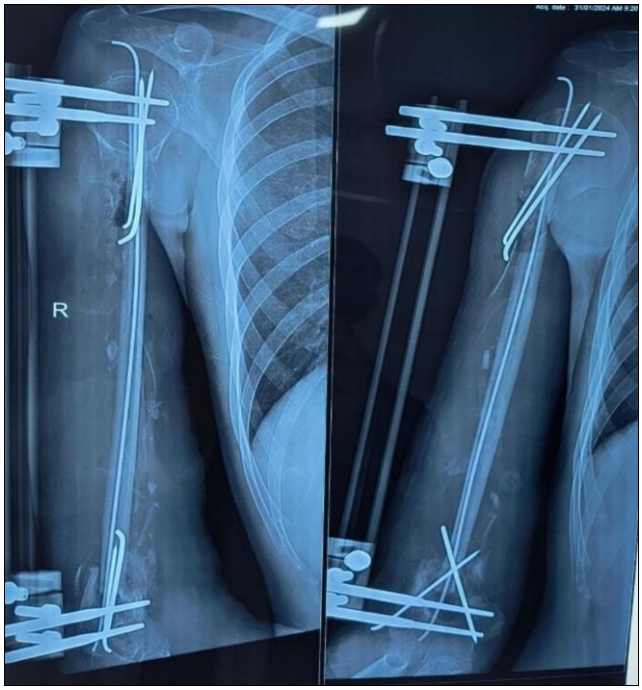

Fig 8: Immediate post-operative x-ray after 3rd stage of surgery showing fibula grafting along with autologous cancellous bone grafting at proximal and distal and fixation externally with rail fixator

Four months post-operative following third stage surgery

At 4 months post fibular grafting, X-ray shows early signs of union at selective graft-host junctions, more evident at the proximal end. However, significant portions of the graft remain unincorporated, with minimal callus formation distally. No signs of implant loosening or mechanical failure are seen.

Fig 9: Four months post-operative x-ray after 3rd stage of surgery showing some signs of union.

Six months after third stage of surgery

At 6 months post fibular grafting, X-ray shows progressive union with improved graft-host integration, particularly at the proximal and distal junctions. Callus formation is more evident, suggesting ongoing osteo integration. While consolidation remains incomplete centrally, signs are encouraging, and healing appears to be progressing steadily.

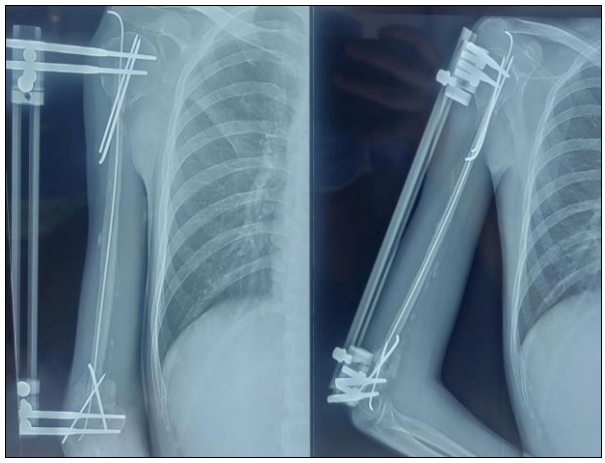

Fig 10: Six months post-operative x-ray showing sign of better osteo integration and healing.

Fourth-Stage Surgery: Definitive Internal Fixation with Plating: At six months following the fibular grafting and bone graft augmentation, the patient demonstrated radiological evidence of union at both the proximal and distal junctions of the fibular graft with the native humerus. Serial X-rays confirmed satisfactory graft incorporation and progressive callus formation, indicating a favourable biological response and mechanical stability.

In view of the progressing union and patient's clinical recovery, a definitive internal fixation was planned. The fourth-stage surgery involved:

- Removal of the external rail fixator under aseptic precautions.
- Exploration of the previous surgical site through a standard approach.
- A 12-hole dynamic compression plate (DCP) was contoured and applied along the lateral aspect of the humerus.
- Fixation was achieved using a combination of locking screws and cortical screws, ensuring both axial and rotational stability.
- Careful preservation of the fibular graft integrity and previously incorporated bone graft sites was ensured during dissection and screw insertion.
- The surgical wound was closed in layers over a negative suction drain.

Postoperative radiographs confirmed stable internal fixation, proper alignment of the humeral shaft, and maintained graft-host integration.

The following section includes intraoperative photograph and postoperative radiograph documenting the surgical steps and final construct.

Fig 11: Showing intraoperative image of fixation with long locking plate along with protected radial nerve.

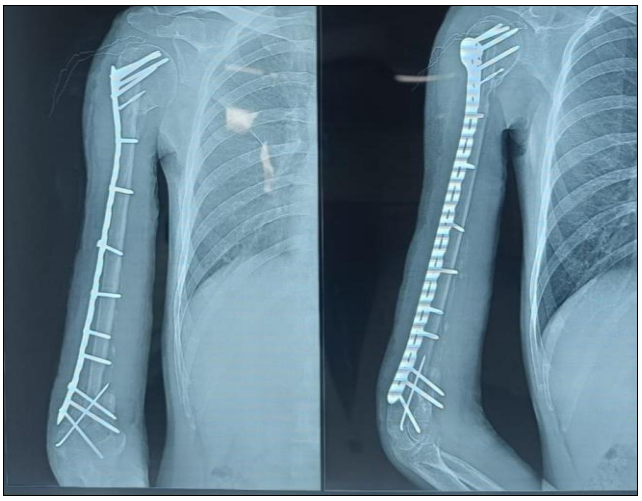

Fig 12: Immediate post-operative x-ray after 4th stage of surgery with long looking compression plating and removal of external fixation.

Follow up and outcome

At approximately 9 months following the definitive internal fixation with long humeral plating and bone grafting, the patient demonstrated significant clinical and radiological improvement.

- X-rays revealed satisfactory union at the graft-host bone junctions.
- There was clear evidence of graft incorporation, cortical continuity, and bridging callus formation, indicating successful osseous integration.
- The fibular graft remained structurally intact and well-aligned with native humerus.

Fig 13: Showing well incorporation of the graft and signs of union after around 9 month of definitive fixation surgery.

Fig 14: 11 months Post-operative X-ray picture after definitive fixation (fourth stage surgery).

At 18 months post definitive reconstruction, radiographs showed satisfactory progression of union with good consolidation at the graft-host junctions. However, clinical findings raised suspicion of chronic low-grade infection, including localized soft-tissue compromise, implant prominence along the lateral arm, and intermittent serous discharge. Despite these issues, the patient maintained a functional but limited range of motion at the shoulder and elbow.

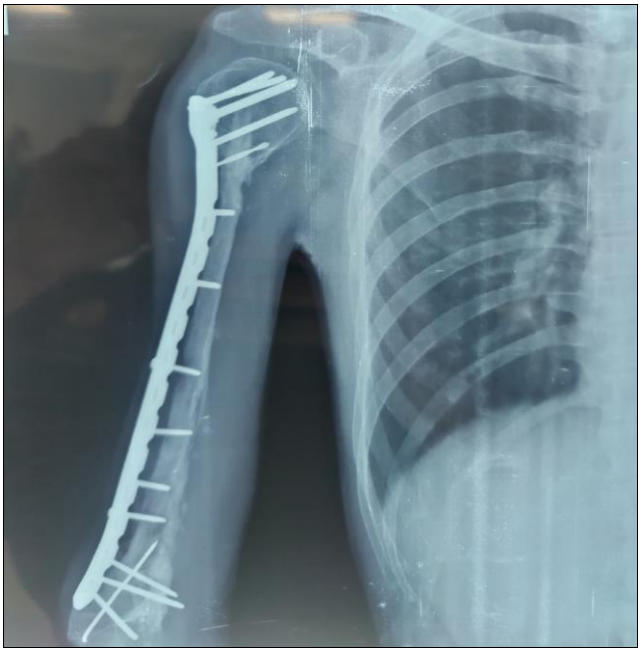

Fig 15: 18 months Post-operative X-ray picture after definitive fixation (fourth stage surgery).

Fifth-Stage Surgery: Implant Removal and Debridement

In view of the exposed hardware and ongoing infection, the patient was planned for a final surgical procedure involving implant removal and thorough debridement.

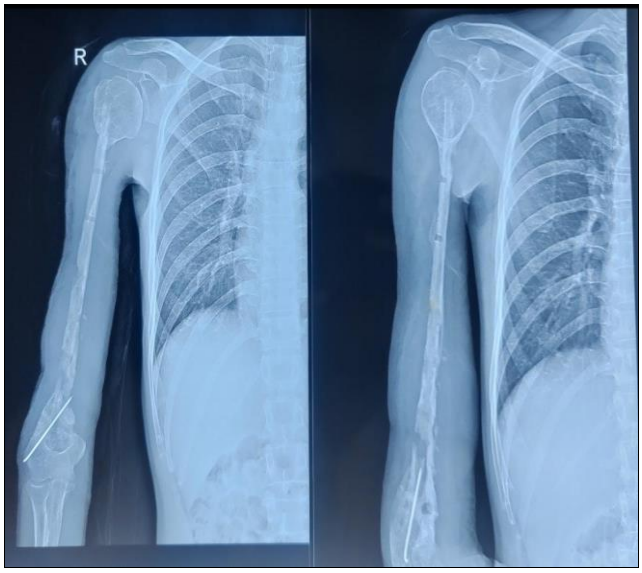

Fig 16: Post-operative X-ray picture after removal of plate

Postoperatively, osteogenesis was to be supported through the initiation of injectable teriparatide therapy. Following implant removal, the patient was initiated on a structured, supervised physiotherapy programme focused on improving shoulder and elbow range of motion, progressive upper-limb strengthening, and functional rehabilitation. The patient has shown steady improvement with gradual restoration of daily-use activities, although final functional capacity continues to improve with ongoing therapy. At present, the limb demonstrates partial but meaningful functional recovery, and the patient remains motivated and compliant with the rehabilitation protocol.

Discussion

Hydatid disease is a parasitic infection caused by the larval stage of *Echinococcus granulosus*, while hepatic (75%) and pulmonary (15%) involvement are most common, osseous hydatidosis represents only 0.5-4% of cases and is considered rare and aggressive [5].

Bone hydatidosis progresses insidiously due to the parasite's slow growth and lack of pericyst formation, leading to diffuse bone infiltration. The spine, pelvis, femur, and tibia are most frequently involved [6], with humeral involvement being extremely rare and sparsely reported in literature [7].

In contrast to soft-tissue hydatid cysts, osseous lesions do not form encapsulated cysts but instead follow a trabecular infiltration pattern, which compromises the bone's structural integrity, ultimately leading to pathological fractures, as seen in our patient [8]. These lesions often mimic chronic osteomyelitis, tuberculosis, or tumours, resulting in delayed diagnosis [9]. In our case, the pathological fracture was initially misinterpreted as a simple traumatic fracture until signs of chronic infection and osteolysis prompted further investigation. Histopathological confirmation remains the gold standard for diagnosis [10].

Radiologically, osseous hydatidosis appears as expansile, multiloculated, lytic lesions, often described as "honeycomb" or "soap bubble" patterns on imaging [11]. MRI and CT are superior in delineating soft tissue extension and bony involvement. Serological tests like ELISA or indirect hemagglutination can support diagnosis, though they may be negative in localized skeletal disease [12].

Surgical management is the cornerstone of treatment, with the goal of radical debridement and resection of infected bone, though this is often limited by the lesion's infiltrative nature.

Recurrence rates remain high (up to 30-50%) due to incomplete excision [13]. In our case, a staged surgical approach was employed:

- Initial debridement and infection control,
- Insertion of antibiotic-loaded cement spacers and external fixation,
- Followed by non-vascularised fibular grafting and internal fixation once infection was controlled.

This staged reconstructive strategy aligns with approaches described in previous reports [14, 15], especially when dealing with large segmental bone loss due to parasitic osteolysis.

The use of non-vascularised fibular autograft has been shown to be a reliable option for segmental defect bridging, offering structural support and osteoconductivity. In our case, autologous cancellous bone grafts at the graft-host junction enhanced union potential. Final stabilization was achieved with a long locking plate, offering rigid fixation and reducing the risk of mechanical failure.

Pharmacologically, Albendazole remains the drug of choice. It interferes with microtubule formation in the parasite, thus inhibiting nutrient uptake and leading to parasite death. Adjunctive use of Albendazole post-surgery reduces recurrence risk and complements surgical excision [16].

Our case demonstrates a rare but important differential for pathological fractures in endemic regions. Early suspicion, appropriate imaging, and confirmatory biopsy are crucial to prevent disease progression. Moreover, this case highlights the success of biological reconstruction using non-vascularised fibular grafting and multi-stage orthopaedic management in achieving union and limb salvage.

Conclusion

Hydatid disease of the humerus is a rare and often overlooked cause of pathological fractures, particularly in endemic regions. Its infiltrative growth pattern and radiological similarity to chronic infections or tumors frequently lead to delayed diagnosis. In our case, the disease was only identified after histopathological confirmation following initial surgical debridement, emphasizing the need for early suspicion and thorough evaluation of atypical bone lesions.

Successful management required a staged surgical approach, beginning with infection control and progressing to biological reconstruction using a non-vascularised fibular graft, followed by definitive internal fixation. The combined use of anti-helminthic therapy and sequential surgical intervention led to satisfactory union and graft incorporation. This case highlights the importance of individualized treatment planning and long-term follow-up in achieving functional recovery and preventing recurrence in rare presentations of osseous hydatidosis.

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