IDCases 37 (2024) e01994

Contents lists available at ScienceDirect

IDCases

journal homepage: www.elsevier.com/locate/idcases

Case report

A case report of primary hydatidosis of ulna and adjacent soft tissue

Torgot Ghani [a], Abdullah Ahmad [b,*], Akhtar Mohammad Totakhail [c], Naser Sardar [d], Kristina Ghani [e], Abdul Razaq Siawash [b]

[a] Wazir Mohammad Akbar Khan Hospital, Kabul, Afghanistan
[b] Prof. Dr. Siawash Surgical Orthopaedic Hospital, Kabul, Afghanistan
[c] Paktia University Faculty of Medicine, Paktia, Afghanistan
[d] Ibn Sina Emergency Hospital, Kabul, Afghanistan
[e] Balkh University Faculty of Medicine, Balkh, Afghanistan

ARTICLE INFO

Keywords:
Case report
Osseous hydatidosis
Primary ulna hydatidosis
Pathologic fracture

ABSTRACT

Hydatidosis, is a zoonotic disease prevalent in sheep-raising regions globally. Musculoskeletal hydatidosis is uncommon and usually remains asymptomatic over a long period. The detection of musculoskeletal hydatidosis often signifies extensive cyst spread within the bone marrow cavity, making treatment difficult with a high recurrence rate. Unlike the conventional surgical approach for visceral hydatid cysts, treating osseous hydatidosis requires a strategy akin to oncologic therapy. We report a rare case of primary hydatidosis affecting the ulna and adjacent soft tissue in a 58-year-old woman. She presented with a painless forearm mass evolving over six years, accompanied by recent onset tenderness and restricted elbow joint mobility. Imaging revealed a cystic mass in the forearm, an intra-ulnar bone lesion, and an olecranon fracture. The primary diagnosis of musculoskeletal hydatidosis was made based on clinical findings and radiological imaging. Treatment involved resection of 2/3 of the proximal ulna and pericystectomy for the soft tissue hydatid cyst. Diagnosis of musculoskeletal hydatidosis is vital for effective preoperative planning, as internal fixation often fails without eradicating the infestation. Treatment typically involves radical operation with wide excision of the affected bone and adjacent joint structures, coupled with chemotherapy. Clinicians in endemic regions should consider musculoskeletal hydatidosis in the differential diagnosis of osteolytic lesions and slow-growing cystic masses. Diagnosis relies on clinical, serological, and radiological assessments.

Introduction

Hydatid disease, is caused by Echinococcus species, mainly involves Echinococcus granulosus and less commonly Echinococcus multilocularis, and Echinococus oligarthrus. A hydatid cyst of Echinococcus Granulosus often leads to cyst formation in carnivores like dogs, wolves, and foxes, as well as in intermediate hosts like sheep, goats, and cattle, with humans serving as incidental intermediate hosts in this context [1]. Humans can become infected with the parasite through direct contact with contaminated animals or by ingesting the parasite through the fecal-oral route. Hydatidosis poses a significant risk to more than 270 million people in Central Asia, covering regions including Mongolia, Kazakhstan, Kyrgyzstan, Tajikistan, Turkmenistan, Uzbekistan, Afghanistan, Iran, Pakistan, and western China. This constitutes around 58 % of the overall population in the region [2]. Echinococcosis primarily affects the liver (50–70 %), lungs (20 %), and spleen (5–8 %), while the involvement of other organs such as the kidneys, brain, bones, spinal cord, and heart are rare, with bone infections averaging 1.5 %. Among bone cases, 60 % are located in the spine, pelvis, and hip joint, with approximately 28 % occurring in the femur, tibia, and humerus, and 8 % in the ribs and scapula. Isolated infections are occasionally found in other parts of the musculoskeletal system [3].

Osseous hydatidosis is considered one of the most severe forms of Echinococcus infection, and its treatment closely resembles oncological therapy rather than the surgical treatment of visceral hydatidosis. Medical treatment alone is insufficient for eliminating the infestation, and it must be coupled with a wide surgical excision similar to radical procedures for oncologic conditions [4]. The diagnosis of hydatid disease should be considered if the individual lives in a region where the infestation is known to be prevalent, or if the individual has moved from or visited an area endemic to the disease [6].

The diagnosis is typically determined through a combination of

* Correspondence to: Siawash Surgical Orthopaedic Hospital, Maiwand Road, Kabul 1003, Afghanistan.
E-mail address: drabdullahahmad786@gmail.com (A. Ahmad).

https://doi.org/10.1016/j.idcr.2024.e01994
Received 31 March 2024; Accepted 23 May 2024
Available online 24 May 2024

clinical, radiographic, and biological data. However, in some cases, a definitive preoperative diagnosis may be challenging to establish despite thorough diagnostic evaluations. As a result, the final diagnosis is often confirmed only during surgery or through a biopsy of the lesion [4].

This report presents a rare case of hydatidosis affecting the ulna and forearm soft tissues in a 58-year-old female, diagnosed after a pathological fracture of the olecranon and treated with radical excision of the proximal ulna and soft tissue pericystectomy.

Case presentation

A 58-year-old patient from a pastoral society in Afghanistan was presented at our hospital due to elbow pain, tingling, and numbness in the right hand and a forearm mass. The mass had been present for six years and gradually expanded in size. The tingling and numbness, which were intermittent at first, have been almost persistent for two years. Recently, after a fall, she developed elbow pain along with a decreased range of motion of the elbow.

On physical examination, a 14×12cm lobulated, non-tender, mobile mass was noticed on the anterior of the elbow joint. There was no erythema or increased temperature present. The elbow was tender on palpation. A radiological X-ray of the elbow revealed an olecranon pathologic fracture along with round forearm mass. The CT scan of the elbow joint demonstrated an expansile lytic lesion involving the proximal ulna with a pathologic fracture of the olecranon. The destructive changes involving the ulna extended to the proximal mid-shaft with multiple breaks and associated large fluid collections around the elbow joint. This communicated with the joint space, extending along intermuscular planes and reaching up to the subcutaneous planes, suggestive of infective or neoplastic etiology (Fig. 1). However, ultrasonography studies showed multilocular soft tissue cystic lesions of the forearm with several floating membranes.

She was hypertensive, but otherwise, vital signs were normal. Routine blood investigations, including complete blood count, liver and kidney function tests were normal. Serology against Echinococcus was negative. Abdominal ultrasonography and chest X-rays were clear.

Given the clinical and radiological evidence, a provisional diagnosis of primary hydatidosis of ulna and adjacent soft tissue was made. The patient prepared for the operation with Albendazole (20 mg/kg), five days prior to the operation. The operation was performed under general anesthesia with 200 mg of hydrocortisone administered prophylactically for possible reaction after cyst rupture. After the skin incision, the cyst appeared subcutaneously (Fig. 2, a). Pericystectomy was done carefully, and after exposing the ulna, 2/3 of the proximal ulna were resected along with the olecranon (Fig. 2, b). Unfortunately, there was spillage of cystic content at the operation site with no reaction occurring. The whole operated area was rinsed with 20 % hypertonic saline and the wound was closed with layers after placing a drain. The gross appearance of the pathology was typical for hydatidosis (Fig. 3), and the histopathological studies confirmed the diagnosis.

After surgery, the elbow was placed in a splint at a 90-degree flexion for three weeks. After three weeks, we advised a functional brace to be used and, passive range of motion and active-assisted motion to be started. The patient was discharged three days after the operation. Albendazole (20 mg/kg) was directed to be taken twice daily for the next three months.

Due to the nomadic lifestyle of the patient, she is followed by phone communication and clinical feedbacks from local clinics. At the ten-month postoperative mark, she is pain-free and can hold her forearm against gravity.

Discussion

Echinococcosis, a prevalent zoonotic disease, is endemic in many nations where sheep farming is common [5]. Individuals who are involved in livestock farming, specifically raising sheep, cattle, horses, and camels, as well as those who have contact with domestic or wild canids, are at risk of contracting the disease. The risk of transmission can also increase through exposure to contaminated water and food sources. [7]. Over 90 % of hydatid cysts are observed in the liver and lungs [6]. Hydatidosis of bone is rare, accounting for only 0.5 % to 2.5 % of human hydatidosis cases, it represents one of the most severe forms of this infection [4]. Hydatid cyst eggs can be transmitted to humans through the fecal-oral route via contaminated food or water. When these eggs reach the gastrointestinal tract, the acidic environment triggers their release of oncospheres. These oncospheres are then absorbed through

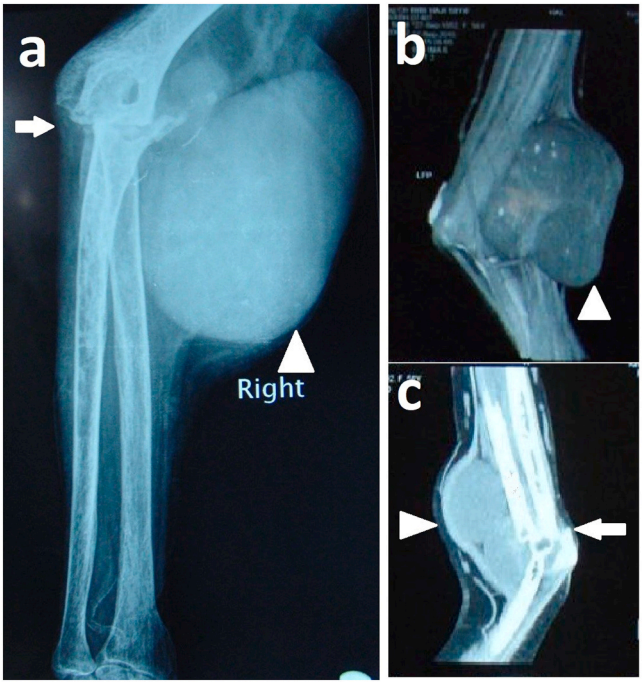

Fig. 1. Radiographic images of right Forearm reveal the following findings: a) Digital x-ray displays a pathologic fracture of olecranon (arrow), accompanied by forearm soft tissue mass (arrowhead). b) 3D CT scan show cases a lobulated cystic lesion in the forearm (arrowhead). c) Sagittal CT scan of forearm exhibiting pathologic fracture of olecranon (arrow) and soft tissue cystic mass (arrowhead).

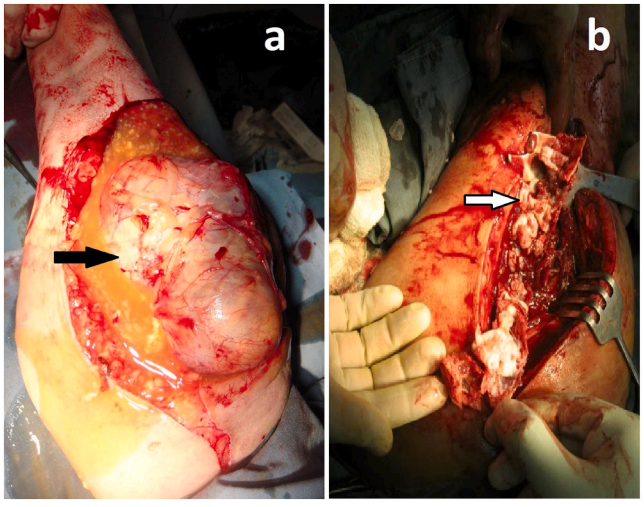

Fig. 2. Intraoperative images of forearm hydatidosis, a) lobulated cystic mass (black arrow) is observed immediately after skin incision, pericystectomy was done for soft tissue hydatidosis. b) hydatid cysts are identified within the ulna (white arrow).

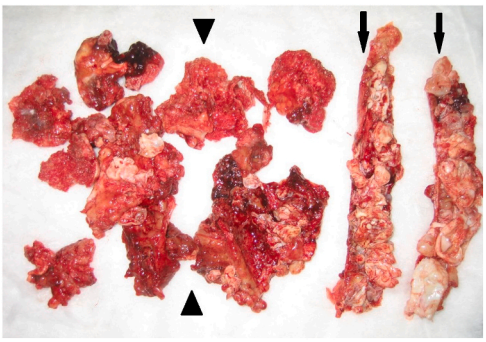

Fig. 3. Arrows indicate the resected ulna image post-resection, filled with hydatid cysts. Arrowheads denote forearm soft tissue hydatidosis after pericystectomy.

the small intestine and subsequently filtered by the liver and lungs [1]. Primary hydatid disease outside the liver and lungs usually happens when larvae manage to evade the capillary beds of the liver and lungs, which function as filters, and enter the bloodstream [2]. The hydatid larva undergoes slow development within the medulla of the bone, due to the dense nature of bone tissue. Microvesicles are produced through exogenous budding and spread throughout the spongy tissue of the affected bone, forming a lesion that initially conforms to the rigid bony structure. The hydatid cyst then develops multiple exogenous daughter cysts in the absence of any adventitial pericyst, which insinuates themselves between the bony trabeculae, destroying them. Ultimately, this process results in extensive damage to the medullary bone caused by the multivesicular cysts [3]. In contrast to the presence of cystic formations in visceral tissues, hydatid larva infestation in bone tissue does not result in cyst formation. Instead, bone infestation is termed echinococosis or osseous hydatidosis. Extraosseous invasion occurs when parasitic formations develop in the surrounding soft tissues due to osseous disruption or pathologic fractures. These external vesicular growths form hydatid abscesses, which contain a seropurulent exudate. These abscesses are characterized as cold and migratory [4].

Osseous hydatidosis may lie dormant in the bone for as long as 40 years. The disease seldom manifests during childhood; it is essentially a disease of adults [4]. Osseous hydatidosis often remains asymptomatic for a prolonged period, with symptoms typically manifesting after a pathological fracture, secondary infection, or the emergence of compressive symptoms on nearby soft tissues [8]. Conventional laboratory tests in patients with bone hydatid are usually normal and serological examination is a more specific diagnostic method; however, due to the inconsistent detection rate of this test, pathological examination of intraoperative specimens remains commonly used in clinical practice for the definitive diagnosis. [7]. The primary diagnostic approach for bone hydatidosis relies on the findings obtained from X-rays and computed tomography scans [8]. Ultrasonography is considered the gold standard for diagnosing soft tissue hydatidosis, while magnetic resonance imaging is particularly valuable for confirming the diagnosis [9]. Additionally, Marzouki et al. highlights the sensitivity of ultrasonography in detecting floating membranes, daughter cysts, and hydatid sand within purely cystic lesions, making it highly suitable for the initial evaluation, even in rare localizations [1]. The discovery of multiple white cystic structures intraoperatively strongly suggests the presence of echinococcosis [7].

By the time bone hydatidosis is recognized, the cyst may have extensively spread within the bone marrow cavity, leading to frequent failure of attempts at local lesion excision or internal fixation placement. To fully eliminate the lesion, it is often necessary to perform a complete resection of the entire bone, extending up to the joint boundary [7]. The treatment of choice for soft tissue hydatidosis is pericystectomy [1]. Strategies such as utilizing hypertonic saline to wash the surgical area during the procedure, conducting postoperative assessments, and administering albendazole before and after surgery are considered effective in reducing the risk of echinococcosis recurrence [10]. According to the literature, resection arthroplasty should be contemplated primarily in cases where infections persist despite repeated salvage efforts and in patients who are medically fragile and unable to endure prolonged or multiple surgical interventions [11]. As osseous hydatidosis is notorious for recurrence and is compared to malignancies, treatment for this case involved removing olecranon and 2/3 of the proximal ulna, following medical literature advice for aggressive action when standard treatments fail.

Conclusion

Musculoskeletal hydatidosis should be considered in any differential diagnosis of osteolytic lesions and slow-growing cystic masses in the musculoskeletal system, especially in endemic regions. Diagnosis relies on clinical, serological, and radiological findings. Although long-term survival is possible, the disease is not easy to eradicate, to address this surgery with a broad safety margin is the best treatment option for osseous hydatidosis.

Ethical approval

Ethical approval was waived by the authors 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.

Funding

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

CRediT authorship contribution statement

Abdul Razaq Siawash: Supervision, Writing – review & editing. **Kristina Ghani:** Data curation. **Naser Sardar:** Data curation. **Akhtar Mohammad Totakhail:** Data curation, Formal analysis, Methodology. **Torgot Ghani:** Conceptualization, Data curation, Writing – original draft. **Abdullah Ahmad:** Data curation, Investigation, Writing – original draft, Supervision.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Acknowledgements

Not applicable.

References

[1] Marzouki A, Naam A, Abdulrazak S, Soumaré B, Lahrach K, Boutayeb F. Musculoskeletal Echinococcus infection as a rare first presentation of hydatid disease: case report. Published 2017 Jul 17. Patient Saf Surg 2017;11:21. https://doi.org/10.1186/s13037-017-0136-y.

[2] Ahmad A, Ghani T, Hanifi AN, Faez SA, Baset Z, Malakzai HA. Recurrent spinal hydatidosis causing Gibbus deformity: report of a rare case. Published 2023 Oct 13. IDCases 2023;34:e01912. https://doi.org/10.1016/j.idcr.2023.e01912.

[3] Reddy IV, Kumar AHA, Samorekar B, Babu BA, Mettu AK. Complicated hydatid cyst of ulna- a rare case report. RD01-RD03 J Clin Diagn Res 2017;11(5). https://doi.org/10.7860/JCDR/2017/21804.9773.

[4] Zlitni M, Ezzaouia K, Lebib H, Karray M, Kooli M, Mestiri M. Hydatid cyst of bone: diagnosis and treatment. World J Surg 2001;25(1):75–82. https://doi.org/10.1007/s002680020010.
[5] Madhar M, Faik OM, Chafik R, El Haoury H, Fikry T, Schuind F. Primary subcutaneous hydatid disease of the forearm. Hand Surg Rehabil 2016;35(3):229–30. https://doi.org/10.1016/j.hansur.2016.01.005.
[6] Bayram M, Sirikci A. Hydatic cyst located intermuscular area of the forearm: MR imaging findings. Eur J Radio 2000;36(3):130–2. https://doi.org/10.1016/s0720-048x(00)00188-1.
[7] Wang X, Huang J, Su L, Ma Q, Ma C, Xie Z. Complete excision of giant clavicular hydatid cyst: a case report. Published 2023 Mar 22. BMC Infect Dis 2023;23(1):178. https://doi.org/10.1186/s12879-023-08149-4.
[8] Khan MS, Hashmi PM, Khan D. Eradication of advanced pelvic hydatid bone disease after limb salvage surgery - 5-year follow-up: a case report. Published 2015 Apr 21 J Med Case Rep 2015;9:21. https://doi.org/10.1186/1752-1947-9-21.
[9] Alatassi R, Koaban S, Alshayie M, Almogbil I. Solitary hydatid cyst in the forearm: a case report. Int J Surg Case Rep 2018;51:419–24. https://doi.org/10.1016/j.ijscr.2018.09.038.
[10] Agholi M, Heidarian HR, Montaseri Z, Khajeh F. Muscular hydatid cyst in Iran: a case report. Int J Surg Case Rep 2023;103:107867. https://doi.org/10.1016/j.ijscr.2022.107867.
[11] Sanchez Sotelo J, Zarkadas P, Throckmorton T, Morrey BF. Elbow resection for deep infection after total elbow arthroplasty: surgical technique. e5. Published 2012 Mar 14. JBJS Ess Surg Tech 2012;2(1). https://doi.org/10.2106/JBJS.ST.K.00017.