DOI: 10.7860/NJLM/2023/60175.2711

Case Report

Pathology Section

Hydatid Cyst in Humerus- A Rare Case Report

RAVINA RAMESH PUROHIT[1], HEMALI J TAILOR[2], CHINTAL GHANSHYAMBHAI PATEL[3], RAHUL MODI[4], MILI SANJAY JADAV[5]

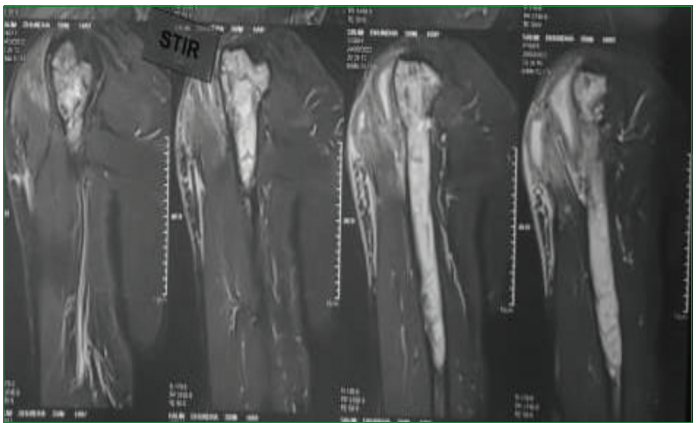
ABSTRACT

Hydatid disease is caused by ingestion of eggs of *Echinococcus* species and formation of cyst in organs where the parasite larvae are deposited. *E.granulosus*, a cestode commonly causes hydatid disease in humans. This case report highlights an unusual presentation of the hydatid cyst. The authors hereby, present a case of 37-year-old male patient with discharging sinus over left upper arm since 13 days along with complain of pain and swelling over left upper arm since 5 years which gradually increased. Laboratory analysis was performed and the patient underwent Ultrasonography (USG) and Magnetic Resonance Imaging (MRI) studies. Histopathological examination confirmed the diagnosis as hydatid cyst. Extensive curettage was done and postoperative albendazole was given. Ossesous hydatid disease, a rare diagnosis, should be included in the differential diagnosis for cystic lytic lesions of bone. To the best of our knowledge, not more than handful of cases of hydatid disease of the humerus has been reported in our country, making this a rare case report.

Keywords: Echinococcus, Humerus, Hydatidosis

CASE REPORT

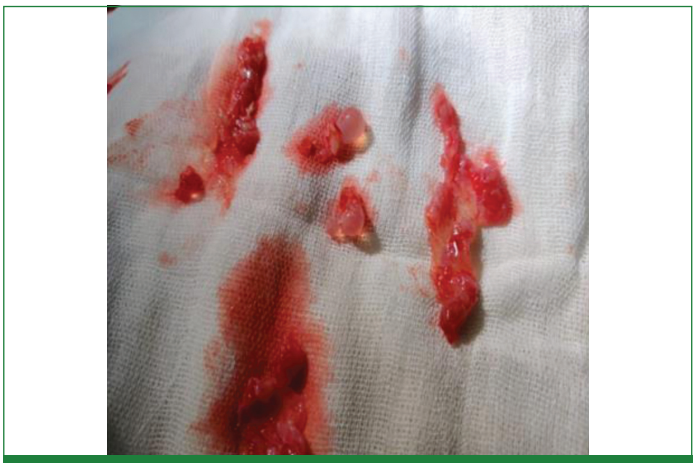
A 37-year-old male patient attended the tertiary care centre of the South Gujarat with complain of discharging sinus over swelling present over left upper arm since 13 days. Apparently the patient was asypmtomatic 5 years ago then he developed swelling over left upper arm, which was gradually increasing in size and was painful. Examination of swelling site revealed a diffuse, firm and tender swelling over left upper arm. MRI of the left arm [Table/Fig-1] showed a thick walled collection with multiple internal loculations and septations approximately measuring 8×3.9×7.8 cm^3 in the deep intermuscular and intramuscular region along the lateral aspect of proximal humerus involving the deltoid muscle showing possibility of soft tissue hydatid cyst with intraosseous extension. Ultrasonography (USG) revealed no hydatid cyst present in liver or any other organ. Chest X-ray revealed no abnormality. Therefore, based on clinical and radiological data cystic lesion of the bone was suspected.



[Table/Fig-1]: Magnetic Resonance Imaging (MRI) of the left arm- showing well-defined altered marrow signal lesion seen in the proximal humerus involving the head and extending upto the mid shaft. There are multiple internal loculations and septations present.

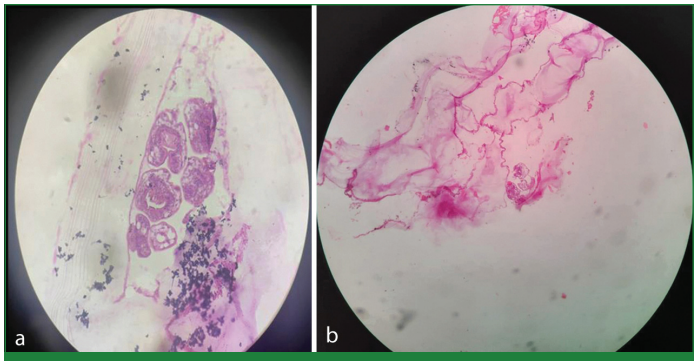
The Department of Pathology received multiple soft tissue bits [Table/Fig-2,3] labelled as "Left-sided proximal humerus septate cyst" measuring 3x1x0.5 cm^3 in size, greyish white in colour. Few colourless cysts were also identified.

[Table/Fig-2]: Gross image of the received specimen (HP 930/22) shows multiple small colourless cyst along with surrounding tissue.

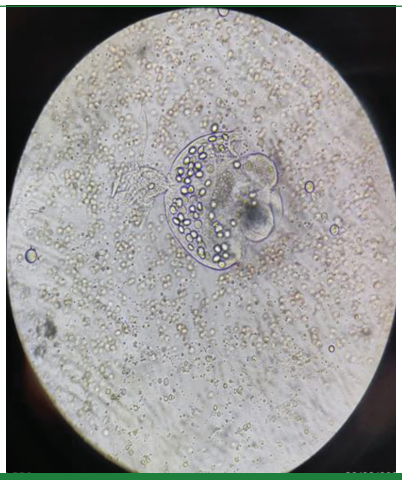


[Table/Fig-3]: Gross image of the received specimen (HP 930/22) shows multiple small colourless cyst.

Microscopic examination showed chitinous cyst wall, daughter cyst with protoscolices along with mixed inflammatory infiltrates [Table/Fig-4a,b]. Overall histological features were that of hydatid cyst as was also evident in wet mount of the slide [Table/Fig-5]. Extensive curettage followed by bone cementing was done for the patient.



[Table/Fig-4a]: (H& E, 40x) showed chitinous cyst wall, daughter cyst with protoscolices along with mixed inflammatory infiltrates.
[Table/Fig-4b]: (H& E, 4x) showed chitinous cyst wall, daughter cyst with protoscolices along with mixed inflammatory infiltrates.



[Table/Fig-5]: Wet mount image of cyst showing protoscolex along with calcerous bodies.

Postoperatively, 12 weeks Albendazole therapy was prescribed and the patients were asked for follow-up after 3 months.

DISCUSSION

Hydatid disease is caused by *Echinococcus granulosus* worm. Hydatid cysts commonly affect the liver and the lung [1]. Although hydatic cyst can form disease in any organ except hair and nails, it usually affects liver, spleen, and lungs with involvement of these three organs responsible from 90% of infections [2]. Primarily bone involvement of hydatid cyst occurs in 1% to 2% and among them 30% to 50% involves the vertebrae, 15% involves pelvis, and less frequently involves long bones [2]. Countries in the temperate zone- China, Central Asia, Mediterranean region, Australia, and parts of South America have the greatest prevalence of cystic echinococcosis while Andhra Pradesh and Tamil Nadu have the highest prevalence of hydatid disease in India [1].

Hydatid disease is caused by ingestion of eggs of *Echinococcus* species and formation of cyst in organs where the parasite larvae are deposited. *E.granulosus*, a cestode commonly causes hydatid disease in humans. Humans are the accidental intermediate host infected by ingestion of food infected with eggs shed by dogs or foxes. For *E.granulosus* dog is the definite host and for *E.multilocularis* fox is the most important definitive host.

After ingestion, *Echinococcus* eggs reach portal venous circulation after passing through duodenal mucosa and cause disease after arriving to liver and the lung [2]. Liver and lung act as filters for the parasite. On the other hand, some eggs may localise to peripheral organs after passing from lung. The larva lodges in capillaries and incites mononuclear and eosinophilic inflammatory reaction, in this process majority of the larvae are destroyed but few encyst [3]. Cyst progressively increases in size, disrupting the structure and function of the organ involved. Infection in humans is usually asymptomatic but large cyst can cause pain [3].

Intraosseous invasion occurs through three methods: (a) An expansive cyst which compresses and displaces the surrounding tissues, leading to pressure-induced atrophy of the bone; (b) ischemia through obstruction or compression of the nutrient vessels; (c) osteoclast proliferation around the compressed bone tissue [1]. Extraosseous invasion results from osseous disruption or pathologic fracture. This exogenous proliferation constitutes the hydatid abscess, which is a cold, migratory abscess, similar to a tubercular abscess [1]. Osseous hydatidosis may lie silent for many years and gets detected only after a complication such as a pathologic fracture, neural deficit, and infection or fistulation of the abscess.

Differential diagnosis of a cystic lesion in humerus includes infections due to typical and atypical agents (including tuberculosis, brucellosis, and parasitic infections), fibrous dysplasia, simple bone cysts, osteosarcoma, intraosseous gangliomas, or osteomyelitis [2]. Diagnosis is difficult due to lack of any specific biochemical or radiological finding. Histopathological examination is the key for the definitive diagnosis of the cyst material. Ultrasound evaluation has a limited role in musculoskeletal hydatid disease unless a soft-tissue component is identified, which may show characteristic cystic appearance [4].

Mondal SK and Sengupta SG study presented a case of Hydatid cyst in the radial bone, the patient had complain of only pain over forearm but discharging sinus was not present similarly as in present study [5]. Joshi N et al., study presented a case of sacral Hydatid cyst with complain of increasing back pain and bilateral radiculopathy, thus mimicking a case of sciatica [6]. Patino JM and Ramos Vertiz AJ represented a case of hydatidosis of complete humerus with complain of diaphyseal humerus fracture [7]. This study was similar to the present study but with contrast that in present study only upper part of humerus was involved. All the above studies showed us that hydatid cyst presented with common complains of pain along with other symptoms. Thus, the authors can see that hydatid cyst can occur in the bone at any locations and manifest symtoms according to the site involved.

Bone hydatidosis is difficult to treat and carries high morbidity due to frequent recurrences [1]. Early diagnosis and treatment is important to prevent complications [8]. Caution is required if surgical removal of cyst is considered as it may inflict an anaphylaxis reaction and/or dissemination of organism can result from spillage of the cyst content [3].

CONCLUSION(S)

Hydatid disease of the bone, although a rare entity, must be kept in mind while dealing with cystic lesions of the bone in the endemic regions. Timely diagnosis and treatment can minimise the morbidity and also save the limb like in our case where the humerus bone was affected.

REFERENCES

[1] Bhatnagar N, Kishan H, Sura S, Lingaiah P, Jaikumar K. Pelvic hydatid disease: a case report and review of literature. J Orthop Case Rep. 2017;7(4):25-28.
[2] Serbest S, Tiftikci U, Uludag A. Unusual localisation of a primary hydatid cyst: scaphoid bone: a case report. Medicine. 2016;95(17):e3290.
[3] Kumar V, Abbas AK, Aster JC. Robbins basic pathology e-book. Elsevier Health Sciences; 2017 Mar 8.
[4] Paruchuri RK, Kapoor A. Osseous hydatid disease- a series of cases. Indian J Musculoskelet Radiol. 2020;2(2):120-24.

[5] Mondal SK, Sengupta SG. Hydatid cyst of radial bone. JNMA: J Nepal Med Assoc. 2009;48(176):321-23.
[6] Joshi N, Hernandez-Martinez A, Seijas-Vazquez R. Primary sacral hydatid cyst. A case report. Acta Orthopædica Belgica. 2007;73(5):674.
[7] Patino JM, Ramos Vertiz AJ. Hydatidosis of the complete humerus. Treated with radical resection and endoprosthesis. Case report. International Journal of Surgery Case Reports. 2019;65:296-300. doi: 10.1016/j.ijscr.2019.10.083
[8] Vlok M, Buckley HR, Domett K, Willis A, Tromp M, Trinh HH, et al. Hydatid disease (Echinococcosis granulosis) diagnosis from skeletal osteolytic lesions in an early seventh-millennium BP forager community from preagricultural northern Vietnam. American Journal of Biological Anthropology. 2022;177(1):100-15.

PARTICULARS OF CONTRIBUTORS:
1. Resident (3rd Year), Department of Pathology, Goverment Medical College, Surat, Gujarat, India.
2. Associate Professor, Department of Pathology, Goverment Medical College, Surat, Gujarat, India.
3. Tutor, Department of Pathology, Goverment Medical College, Surat, Gujarat, India.
4. Assistant Professor, Department of Pathology, Goverment Medical College, Surat, Gujarat, India.
5. Resident (1st Year), Department of Pathology, Goverment Medical College, Surat, Gujarat, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Ravina Ramesh Purohit,
Room No. 154, 10th Floor Building, New PG Hostel, New Civil Hospital Campus, Majuragate, Surat, Gujarat, India.
E-mail: rajpurohitravinasingh@gmal.com

PLAGIARISM CHECKING METHODS: [Jain H et al.]
- Plagiarism X-checker: Sep 27, 2022
- Manual Googling: Dec 12, 2022
- iThenticate Software: Dec 14, 2022 (17%)

ETYMOLOGY: Author Origin

AUTHOR DECLARATION:
- Financial or Other Competing Interests: None
- Was informed consent obtained from the subjects involved in the study? NA
- For any images presented appropriate consent has been obtained from the subjects. NA

Date of Submission: **Sep 17, 2022**
Date of Peer Review: **Nov 03, 2022**
Date of Acceptance: **Dec 15, 2022**
Date of Publishing: **Apr 01, 2023**