



Multicentric Musculoskeletal Hydatid Cysts - A Rare Case Report

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Hydatid disease is one of most common parasitic diseases affecting humans. Though no organ is exception, the hydatid cysts preferably develop in the liver and the lungs. Though the musculoskeletal involvement is known to be rare, it should be considered in the differential diagnosis of soft tissue tumors.

We report an unusual case of musculoskeletal hydatid disease presenting in multiple locations over a period of four years duration in a 34-year-old male patient. The disease manifested with painful swellings, initially on the lateral aspect of right hip, then on the left hip and later in the left shoulder region. They were treated by incision and drainage/surgical excision in combination with anti-helminthic medication. Final functional outcomes were found satisfactory.

Keywords: Hydatid Cyst; Musculoskeletal Echinococcosis; *Echinococcus granulosus***Introduction**

Hydatid Disease (Cystic Echinococcosis) is a zoonosis caused by the tapeworm *Echinococcus granulosus*. In the life cycle of *E. granulosus*, humans are the intermediate hosts whereas the dogs act as definitive hosts. Man gets infected by accidental ingestion of eggs from the faeces of definitive host. The ingested eggs gain into the circulation after penetrating the intestinal mucosa and start developing the cystic lesions in the suitable anatomical sites [1].

Manifestation of symptoms depends on the size, location, organs involved, compressive effects on adjacent tissues, complications viz rupture, anaphylaxis etc. Since this is slow growing lesion, majority of them are understandably asymptomatic in the early stages.

Infection with *E. granulosus* results in development of solitary or multiple hydatid cysts; majority of which occurs in the liver (70%) and in the lungs (20%). Involvement of musculoskeletal tissues is uncommon (1% to 4%) even in the endemic regions [2]. If involved, the disease has been frequently reported with pelvic, thigh, and paravertebral muscles [3].

In suspected cases, careful clinical history, physical examination along with serological, radiological and histopathological investigations are necessary for confirming the diagnosis.

Here, we present a case of primary intramuscular and subperiosteal hydatid cyst appeared in three different locations in a 34-year-old male patient over a 4-year period of time. We also would like to discuss the pitfalls in diagnosis and management of this otherwise a rare entity.

Case History

Site 1

In August 2016, a 30-year-old male presented with complaints of painful swelling on the lateral aspect of right hip region for four weeks duration. There was no history of trauma, fever, weight loss or any other constitutional symptoms. Clinical examination revealed diffusely tender, firm swelling measuring 00x00 cm over right the greater trochanter. Plain x-ray (Figure 1a and 1b) showed calcification with no attachments on bony structures.



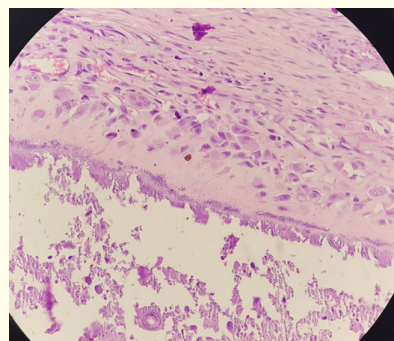
1a



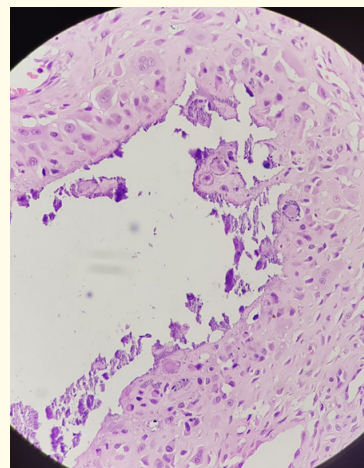
1b

Figure 1a and 1b: Radiographs of right hip. Note the calcification near right greater trochanter. 1a. Pre-op, 1b. Post-op.

As the presentation resembled soft-tissue mass on clinical and radiological examination, the patient was planned for an excision and biopsy. Upon the exploration, the gluteus medius was found necrosed; about 50 ml of creamy thick fluid found collected under the deep fascia. The fluid was drained and the devitalized tissues were excised leaving a reasonably good margin of healthy tissues. Thorough saline irrigation was given. Scrapings and other tissues were sampled for biopsy. Diagnosis of echinococcus infection was made only after the histopathological examination (Figure 2a and 2b). Anti-helminthic drug albendazole was given postoperatively for period of 3 weeks.



2a



2b

Figure 2: Biopsy from right hip. 2a -The germinal layer, adventitia and calcified contents of the parasitic cyst. (400× H&E). 2b -The germinal layer with scolex head, calcification and adjacent giant cell reaction (400× H&E).

Site 2

In November 2019, the same patient presented with almost similar complaints on the left hip region. No pain or swelling on the previously operated site, i.e. the right hip region. MRI investigation was carried out (Figure 3a and 3b).



3a



3b

Figure 3a and 3b: Magnetic resonance images of the left hip.

During the surgical procedure, about 5 x 4 x 4 cm sized cystic swelling was found on the anterolateral aspect, superficial to the gluteus medius muscle (Figure 4). Wide excision was performed followed by copious saline lavage. Biopsy confirmed the same tape-worm infection. Similar postoperative protocol was followed.

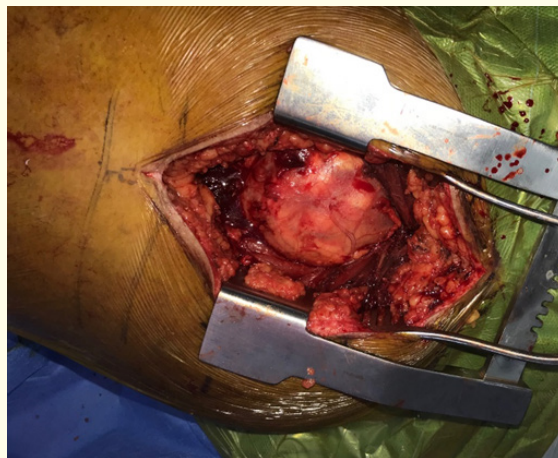


Figure 4: Per-op photo of the left hip.

Site 3

In March 2020, the above said patient reported with diffuse swelling about the left shoulder. MRI images showed three cystic masses, (measuring 16 x 6 x 5 mm, 11 x 4 x 3 mm and 10 x 4 x 5 mm) occupying at the tendinous portions of infraspinatus and subscapularis muscles (Figure 5). Additionally, a well-circumscribed cyst measuring 50 x 45 x 25 mm was seen eroding the cortex of lateral end of the clavicle, with reactive hyperostosis in the remaining part of the bone (Figure 6a and 6b).



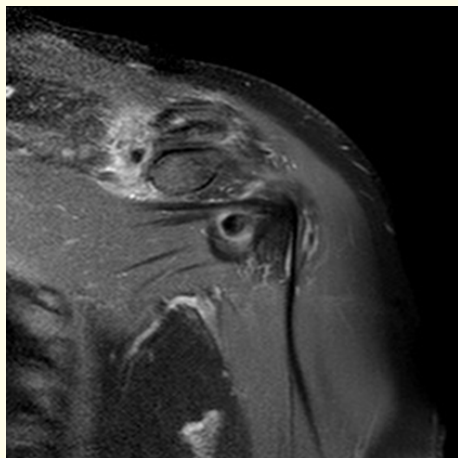


Figure 5: Magnetic resonance images of the left shoulder.



6a



6b

Figure 6a and 6b: Radiographs of the left shoulder; 6a. Pre-op, 6b. Post-op.

Accessibility to all the aspects of cysts was met with difficulty. After considering multivesicular nature of the cysts and complex anatomy of the shoulder, percutaneous aspiration-injection-reaspiration (PAIR) technique was attempted and unsuccessful. Hence, the lesions were removed piecemeal through incisions made over the swellings. Saline wash and post operative medication were followed as before.

Among the three sites described here, in the first two (the hip regions), the cysts were confined to the intermuscular planes, whereas in the third (left shoulder), the pathology extended beyond the cortex of left clavicle. In all sites, we used hypertonic saline (the most widely used and easily available scollicidal agent) for irrigation to prevent recurrence or dissemination due to inadvertent intra-operative spillage.

Discussion

Hydatid cyst is a parasitic disease caused by *Echinococcus granulosus*, and is an endemic infection in Mediterranean countries, the Middle East, Africa, Asia, South America, and Australia [4]. Despite being a common zoonotic disease, the musculoskeletal involvement in the humans is rare [5].

Owing to its low prevalence in bone and muscle tissues and its similar insidious growth pattern with that of soft tissue tumors, definitive pre-operative diagnosis of muscular hydatid cysts poses great challenges to the clinicians. In fact, the most common symptom of isolated intramuscular hydatid cyst is a slow growing painless mass [6].

Many a times, the diagnosis is made either intraoperatively or after the histopathological results. We would like to remind here that, in our case during its first presentation, the diagnosis of hydatid cyst was made only after the surgery and confirmed by the biopsy report. When more than one muscle or its surrounding tissues are involved, as depicted in the left shoulder of this patient, encroachment of adjacent bones can happen [7].

Most studies show that the musculoskeletal hydatid cysts usually present as isolated swellings and are not seen to be associated with other organs involvement [8]. This was true in our patient as well, as he did not suffer any symptoms suggestive of internal organ involvements or space occupying lesions.

Several serological tests like enzyme-linked immunosorbent assay (ELISA), Western blot, indirect hemagglutination antibody assays are available for the diagnosis of *E. granulosus* infection [9]. These tests show positivity over 90 percent cases of liver hydatid cysts, and less than 30 percent in musculoskeletal cysts [6].

Computed tomography (CT) gives information such as size, site, calcifications and number vesicles in the cysts. It also shows the relationship with the surrounding tissues [10].

MRI is the diagnostic modality of choice as far as the musculoskeletal hydatid cysts are concerned. It provides a better visualization regarding the extent of the lesion and its relations with adjacent neurovascular structures [11].

Although periosteal reaction, osteosclerosis or calcification are not specific for hydatid bone disease, when dealing with cases of chronic osteomyelitis, hydatid infection should be considered in differential diagnosis especially in the endemic regions [12].

Surgical removal of the cyst is the best treatment approach for surgically accessible muscular cysts [13]. Chemotherapy with albendazole remains the drug of choice both in pre and postoperative adjuvant therapy. Mebendazole and praziquantel are the other alternatives. The importance of antihelminthics intake lies on reducing the chances of metastatic infections and recurrences [14]. For inoperable cases, albendazole in combination with percutaneous aspiration-injection-reaspiration (PAIR) is considered a safe option [15].

Limitations of the Study

Relapses/recurrences are high if surgery is carried out without complete and effective antihelminthic coverage [16]. In addition to complete excision, duration of chemotherapy is considered a key factor in achieving effective eradication. Also, the responses to treatment will consume longer time to if the cysts are larger [17]. WHO suggests preoperative coverage for a period of 4 - 30 days [18].

We must have been aware of the above mentioned evidences and should have initiated antihelminthic agents prior to the procedure and properly monitored during the post-operative periods. The efficacy of albendazole increases if given for 3 months, but our case had taken only for 3 weeks which was a major (among the oth-

ers) drawback in the management.

We accept that inadequate intake of albendazole and insufficient post-op monitoring might have contributed to successive development of cysts in this patient, albeit on different locations.

Conclusion

Primary musculoskeletal hydatid cyst is a rare condition. It requires high index of suspicion during the diagnosis of soft tissue swellings, particularly in the endemic areas. Definitive diagnosis requires a multidisciplinary approach which includes clinical, radiological and histological examinations. Surgical procedures combined with albendazole yields a better functional outcome. Most importantly, albendazole must be administered before and after the surgical procedures to avoid recurrence and secondary hydatidosis.

Disclosures

None.

Conflict of Interest

The authors declare that there is no conflict of interest regarding the publication of this manuscript.

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