



Primary Cervical Muscular Hydatid Cyst: A Rare Case and Review of Literature

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Abstract

Hydatid disease caused by *Echinococcus granulosus* and *Echinococcus multilocularis* commonly present with pulmonary and hepatic cysts. Primary spinal muscle cyst is a rare presentation. We report a case of hydatid cyst within paraspinal muscles presenting with cervical mass. The hydatid disease serological test was negative. Neither hepatic nor pulmonary cysts were found. Surgical resection was planned due to provisional diagnosis of muscular cystic neoplasm. During surgery, a cyst containing clear liquid was found. The cyst wall was excised and surgical field was irrigated with hypertonic saline. Postoperative pathological examinations revealed a muscular hydatid cyst.

Keywords: Hydatid cyst; Cervical; Muscle

Introduction

Hydatid disease remains an important health issue in livestock of developing countries where veterinary control is infrequent [1]. *Echinococcus granulosus* and less commonly *Echinococcus multilocularis* are primary species responsible for hydatid cysts in humans [2,3]. Humans act as an intermediate host and dogs as the final and definitive host [4]. The larval phase of *Echinococcus* penetrates the intestinal wall and is commonly transferred to the liver *via* portal circulation [5]. Consequently hepatic hydatid cyst is the most common. Primary soft tissue hydatidosis without hepatic or pulmonary involvement is a rare entity in hydatid disease. Especially paraspinal muscular involvement has been rarely reported [6].

We report a case of cervical paraspinal muscular hydatid cyst without identifiable hepatic or pulmonary hydatid involvement.

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Received Date: 21 Jan 2019

Accepted Date: 18 Feb 2019

Published Date: 20 Feb 2019

Citation:

Raswan US, Samoon N, Bhat I. Primary Cervical Muscular Hydatid Cyst: A Rare Case and Review of Literature. *Ann Clin Case Rep.* 2019; 4: 1603.

ISSN: 2474-1655

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Case Presentation

A 40-year old male came to neurosurgery department with one year history of a slowly growing occipito-cervical mass with mild pain on neck movement and no fever. Clinical examination revealed 7 cm × 7 cm × 5 cm round swelling to be cystic and fluctuant with no local inflammatory response or spasm of the cervical muscles. Physical and neurological examination revealed no abnormalities. Chest X-ray and sonography of liver were normal. Serological testing for hydatid disease was negative. Magnetic Resonance Imaging (MRI) showed multilocular cystic lesion in right paravertebral region at C1-C2 level extending to posterior triangle, exhibiting low signal on T1W1 with high signal on T2W1 measuring 7.7 cm × 7.4 cm × 4.2 cm in size (Figure 1 and 2). The cystic lesion was round and well delineated by a wall that showed no enhancement after injection of contrast. Differential diagnosis of a cystic neoplasm or an infective lesion was made. Both clinical and laboratory examinations were normal. The patient underwent medical therapies including antibiotics, but his complaints were not alleviated. His past history was unremarkable. The cyst was removed surgically through a posterior midline approach (Figure 3). Cyst was entered; it contained clear liquid. Multiple cysts were removed and the wall of the cyst was excised (Figure 4); sent for histopathological examination. As precautionary measure the surgical field was irrigated with hypertonic saline to deal with spillage of cyst contents.

Histopathology revealed laminated membranes of hydatid cyst with surrounding reactionary inflammation (100X), and squash smear shows laminated membranes of hydatid (400X) too (Figure 5 and 6).

The patient was discharged under good surgical condition and Albendazole treatment was initiated in the early postoperative stage: 400 mg twice a day for 8 weeks. No recurrence was seen

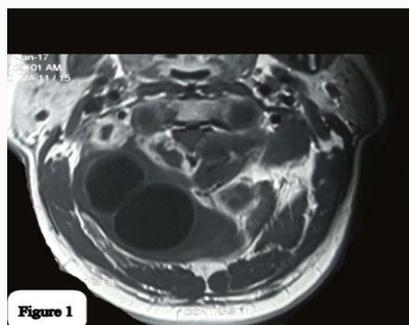


Figure 1: MRI (T1 axial)-Multilocular cystic lesion in right paravertebral region at C1-C2 level extending to posterior triangle, exhibiting low signal on T1 weighted.

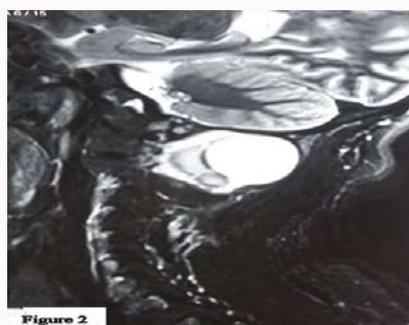


Figure 2: MRI (T2 sagittal)-Cyst shows high signal on T2 weighted image.



Figure 3: Per-operative image showing the cystic lesion seen through the posterior midline approach.

at 6 months.

Discussion

Primary hydatid cyst in soft tissue and muscles without hepatic or pulmonary cysts is extremely rare. A proposed explanation for this observation is effective filtering effect of hepatic and pulmonary circulation which traps the echinococcus larvae. In 10% to 15% of cases, however, larvae can escape from this filtering effect and form hydatid cysts in other organs [7]. Sener et al. [8] provided an alternative mechanism for bypassing the hepatic and pulmonary circulation in formation of primary widespread spinal and paraspinal hydatid cysts. Based on presence of porto-systemic anastomoses in various anatomical locations, they proposed that the larvae penetrate the intestinal muscle and may directly enter the inferior vena cava system through small venous connections at the intestinal walls instead of entering the portal circulation. This mechanism probably explains the epidural location of hydatid cysts in our patient.



Figure 4: Post excision, red-brown surrounding reactionary capsule of the cyst with whitish-yellowish multiple hydatid cysts.

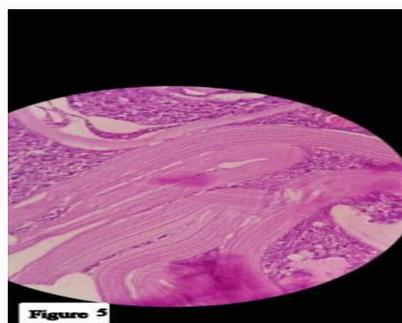


Figure 5: Histopathological photomicrograph show laminated membranes of hydatid cyst with surrounding reactionary inflammation (100X), haematoxylin and eosin stain.



Figure 6: Photomicrograph of a squash smear shows laminated membranes of hydatid (400X), haematoxylin and eosin stain.

A palpable slow-growing mass is the most constant clinical finding in soft tissue hydatid disease. Symptoms related to their compressive effects on adjacent organs are another common finding [9]. Overall, paravertebral muscular hydatidosis presents with non-specific symptoms such as edema, pain and tenderness [10]. Thus physicians must be aware of this diagnosis in patients from endemic regions or with positive history of hydatid disease. Differential diagnosis includes congenital cysts, pseudocysts, cystic tumors, abscess and hematomas [7]. Our patient had mild pain on neck movement and was initially thought to be a cystic tumor on radiology.

MRI represents the gold standard for diagnosing spinal hydatid cyst. On MRI, hydatid cysts appear as well-circumscribed, cystic lesions, with CSF-like signal intensities. The cyst wall is usually thin and regular with no septations. The cysts are hypointense on T1-

weighted images. On T2-weighted images, they appear hyperintense with sharply defined, hypo intense cyst wall which shows mild enhancement following intravenous gadolinium, reflecting the vascularity of the pericyst in case of muscle hydatid cyst. But there is no contrast enhancement either in extradural or intradural hydatid cysts [11].

In our case serological tests before surgery were negative. Serological tests such as haemagglutinin, complement fixation and ELISA may aid in diagnosis but are not positive in all cases of muscular hydatid cyst [12]. Therefore complete reliance on serology for definitive diagnosis is not recommended [13]. The hydatid cyst capsule may play a role in fast-negative results in serological tests because of isolation of the parasite from the host immune system by the cyst capsule. Another proposed explanation is inadequate Th-2 cell activation and cytokine production that is implicated in immunoglobulin expression in cystic echinococcosis [14].

Surgical removal is the most effective treatment of hydatid cyst [15]. Surgeon must be careful to avoid spillage of cyst to avoid fatal anaphylaxis, recurrence and multiple hydatidosis [16]. If presurgical diagnosis is hydatid cyst, preliminary aspiration and instillation of hypertonic saline (20%), silver nitrate (0.5%), formalin, and other chemical could be used to prevent seeding of the cyst contents and to inactivate the protoscolices [17]. Therapy with nontoxic scolical agents or combination chemotherapy with Mebendazole is of therapeutic value in the treatment of patient with recurrence or a high risk contamination [18]. There was complete cyst removal with intentional aspiration of cyst contents and no spillage in the present case. Albendazole is suggested to be given post operatively for 1-3 months [19]. We treated the present case with Albendazole for eight weeks postoperatively. The diagnosis of hydatid cyst is confirmed by histology [20]. Histopathological evaluation shows three layer of hydatid cyst, the inner most germinal layer which is thin and translucent on gross. The embryonic tape worm, scolices develops from an out pouching of the germinal layer and form hydatid sand, setting into the dependent part of the cyst. The cyst fluid is crystal clear, as it is transudate of serum containing proteins and is therefore antigenic. The middle laminated membrane is white 2 mm thick and is easily ruptured. It is electively permeable to nutrients but not to bacteria. The outer layer or pericyst is a rigid protective layer with a few millimeters thickness, representing response of the host to the parasite [16]. Our case also demonstrates histopathological, a cellular, thick, lamellar cyst wall the surrounding host reaction, which is composed of the inflammatory fibrous tissue, form a dense pseudo capsule around the cyst confirm the diagnosis.

Conclusion

The case is a reminder that hydatid cyst should be kept in mind as differential diagnosis of cervical masses especially in countries where Echinococcus infestation is endemic. Imaging techniques, though sensitive cannot always pin-point the exact etiology of cystic lesion. Surgical resection is the goal and spillage of cyst content should be avoided.

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