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Case report

Unusual clinical presentation of spinal echinococcal cyst imitating a malignant tumour

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ABSTRACT. Spinal echinococcal cyst is very uncommon and may have variable clinical presentations. We describe an exceptional case of intradural and extramedullary spinal echinococcal cyst, misguided as a spinal tumour on radiological examination and was diagnosed as echinococcal cyst on fine needle aspiration biopsy smears and subsequently on histopathological examination.

Key words: hydatid disease, echinococcal cyst, spine, FNAB, *Echinococcus granulosus*

Introduction

Hydatid disease in humans is caused by the tapeworm *Echinococcus granulosus*. Primary echinococcal cyst is common in the liver, spleen, and lungs [1]. Spinal echinococcal cyst is very uncommon and may have variable clinical presentations. Here we are presenting an exceptional case of intradural and extramedullary spinal echinococcal cyst, misguided as a spinal tumour on radiology.

Case report

An 11-year-old female child presented with weakness while walking and standing, for 5 years. She also had complaints of backache, bladder instability and difficulty in squatting. She was advised a Computerised Tomography scan which revealed an intradural, extramedullary cystic lesion measuring 2.3 cm at D12-L1 level, compressing and displacing the spinal cord. Radiologists and clinicians suggested a diagnosis of cystic tumour with possibilities of cystic schwanomma or an ependymoma. Subsequently, CT guided fine needle aspiration biopsy (FNAB) was done which yielded 2 ml of straw colored fluid. The smears were air dried as well as wet fixed in 95% ethyl alcohol and were stained with May Grunwald Giemsa, and haematoxylin and eosin method. FNAB samples showed, giants cells and *E. granulosus* scolices with rostellum of hooklets and free lying hooks (Figs.1,2). Further histopathological sections revealed different layers of echinococcal cyst. Scolices of parasite were seen developing from an out pouching of the germinal layer. The middle laminated membrane and a marked foreign body

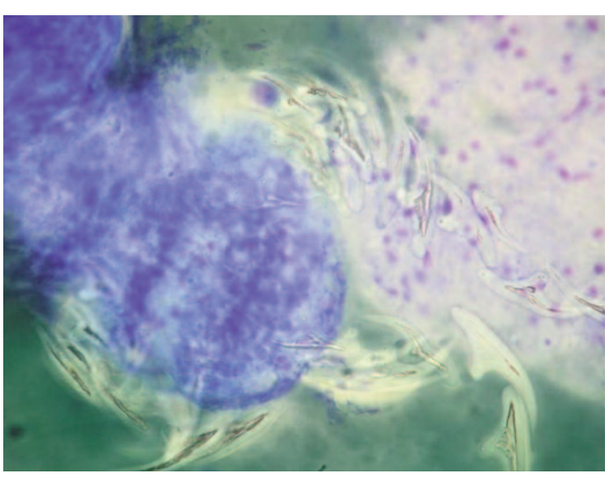

Fig. 1. Multiple hooks of *E. granulosus* from a hydatid cyst fluid obtained by a fine needle aspiration biopsy (May-Grunwald-Giemsa staining, 40×)

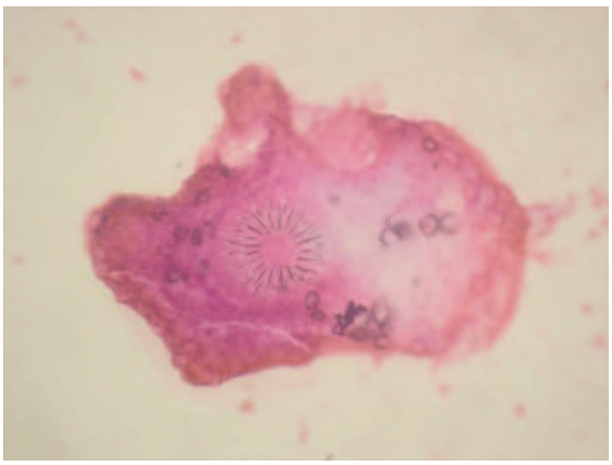

Fig. 2. Scolex of *E. granulosus* from a hydatid cyst fluid obtained by a fine needle aspiration biopsy (H and E, 40×)

type giant cell reaction was noted. So a final diagnosis of spinal echinococcal cyst has been made on the basis of parasitological and histopathological examinations. The case is so exceptional because of the: 1. Young age of the patient; echinococcal cysts are rarely diagnosed in children because of long period of parasite development; 2. Unusual and difficult location in spine; 3. Diagnostic difficulties because of watery fluid in biopsy specimen.

Discussion

Hydatid disease in humans is caused by the cystic (metacestode) stage of the tapeworm *E. granulosus*. Canines are the primary host of the parasite. The life cycle of *E. granulosus* may also involve sheep, cattle, goats, and humans. This infection is transmitted orally via eggs shed in the feces of infected animals. Primary hydatid cysts are located in the liver, spleen, and lungs [1]. Musculoskeletal involvement is secondary and uncommon, with an incidence of less than 2.5%. It affects the pelvis and sacrum, metaphyses of the long bones, skull, spine, and ribs in decreasing order of incidence. Spinal location is rare, with an incidence of less than 1% [2]. Signs and symptoms produced by spinal echinococcal cyst depend upon the size and location of the cyst. The clinical manifestations of spinal hydatid disease are non-pathognomonic, and symptoms are usually related to compression of the spinal cord. Generally, the first symptoms are back and radicular pain. Weakness of the limbs occurs in the advanced stage of the disease and paraplegia is reported in 25% to 84% of cases [3].

Diagnosis can be made on fine needle aspiration cytology and histopathology. FNAB is the next step in diagnosis after radiology and if watery fluid is aspirated hydatid disease should be suspected [4]. However, watery clear fluid is more likely to be seen in non-parasitic simple cysts located in internal organs or tissues. Interestingly, fine needle aspiration cytology enabling the diagnosis of hydatid cyst without procedure-related complication is reported in literature [5]. FNA smears may reveal classic scolices, hooklets and laminated membrane. However probable minimal complications (mild anaphylaxis) can be managed by antianaphylactics [6].

Histopathological evaluation shows three layers 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, settling into the dependent parts of 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 2mm thick and is easily ruptured. It is selectively 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 [7]. There may be marked foreign body type giant cell reaction [8].

A preoperative cover with an effective antihelminthic agent like albendazole to eliminate a risk of spreading *E. granulosus* protoscoleces with a potential risk of developing secondary ehinococcosis is necessary. The choice of the treatment in spinal hydatid disease has been surgery for nearly all cases. The preferred operative procedure is usually laminectomy. The total removal of the cysts without rupture should be the aim of the operation. Irrigating the wound with hypertonic saline or diluted Betadine solution after the cyst removal helps destroy and disrupt the parasites. In the early postoperative stage, adjuvant antihelmintic drug therapy must be given [9,10].

Conclusions

Spinal echinococcal cyst is a rare manifestation, it should be kept as a differential diagnosis of cystic mass lesions of spinal cord. The earliest confirmatory preoperative diagnosis of hydatid cyst in rare locations like spine is possible by FNAB and it can act as a simple diagnostic tool in such cases.

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