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SHORT COMMUNICATION

Isolated musculoskeletal hydatid disease: diagnosis on fine needle aspiration and cell block

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Abstract Hydatidosis is a zoonotic infection caused by *Echinococcus granulosus*. The most common sites of involvement are liver and lungs. Isolated musculoskeletal hydatidosis in absence of visceral involvement is rare and it mimics bone or soft tissue neoplasm. Fine needle aspiration cytology and cell block aids in diagnosis in such unusual location. Here we present one such rare case of isolated musculoskeletal hydatidosis diagnosed on fine needle aspiration cytology and cell block which was mimicking as fibrous dysplasia on radiology.

Keywords Hydatidosis · Cell block · Musculoskeletal · Fibrous dysplasia

Introduction

Hydatid disease (HD) is a zoonotic infection caused by *Echinococcus granulosus*. Isolated musculoskeletal hydatidosis is rare. Here we report a unique case of isolated musculoskeletal HD diagnosed on fine needle aspiration (FNA) and cell block. Hydatidosis in musculoskeletal system mimics as bone and/or soft tissue tumor. In such rare cases, FNA cytology (FNAC) and cell block aid in rapid diagnosis.

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Case report

A 22 year-male presented with right arm and forearm swelling for 8 months. Physical examination revealed a painless slow growing, soft, non tender swelling and suspected clinically as soft tissue sarcoma (Fig. 1a). Radiograph showed a lytic lesion with cortical disruption, internal septation and calcification involving shaft of ulna with metaphyseal extension suggestive of fibrous dysplasia (Fig. 1b). Subsequently FNAC of forearm swelling yielded 160 ml of clear fluid and the material was processed for cytology and cell block. The May Grunwald giemsa (MGG) and Papanicolaou stained smears showed occasional fragment of acellular lamellated membrane, numerous polymorphs, few lymphocytes and occasional osteoclastic giant cells (Fig. 2a, b). The residual material within the hub of the needle was rinsed with formalin, the tissue sediment was processed to prepare paraffin embedded block. Hematoxylin and eosin (H&E) stained cell block sections showed lamellated membrane, numerous scolices and hooklets aligned in two rows (Fig. 2c–f). The core of the hooklets were acid fast on Ziehl Neelsen stain (Fig. 2g, h). Solid phase ELISA IgG tests were positive for both Echinococcus as well as cysticercal antibodies (IBL International GMBH-Kit). Subsequent T2-weighted magnetic resonance imaging (MRI) showed hyper-intense cystic lesion with curvilinear membrane inside ulna. Similar hyper intense lesion was also seen in soft tissue of arm (Fig. 1c, d). Additional imaging did not show any other visceral involvement.

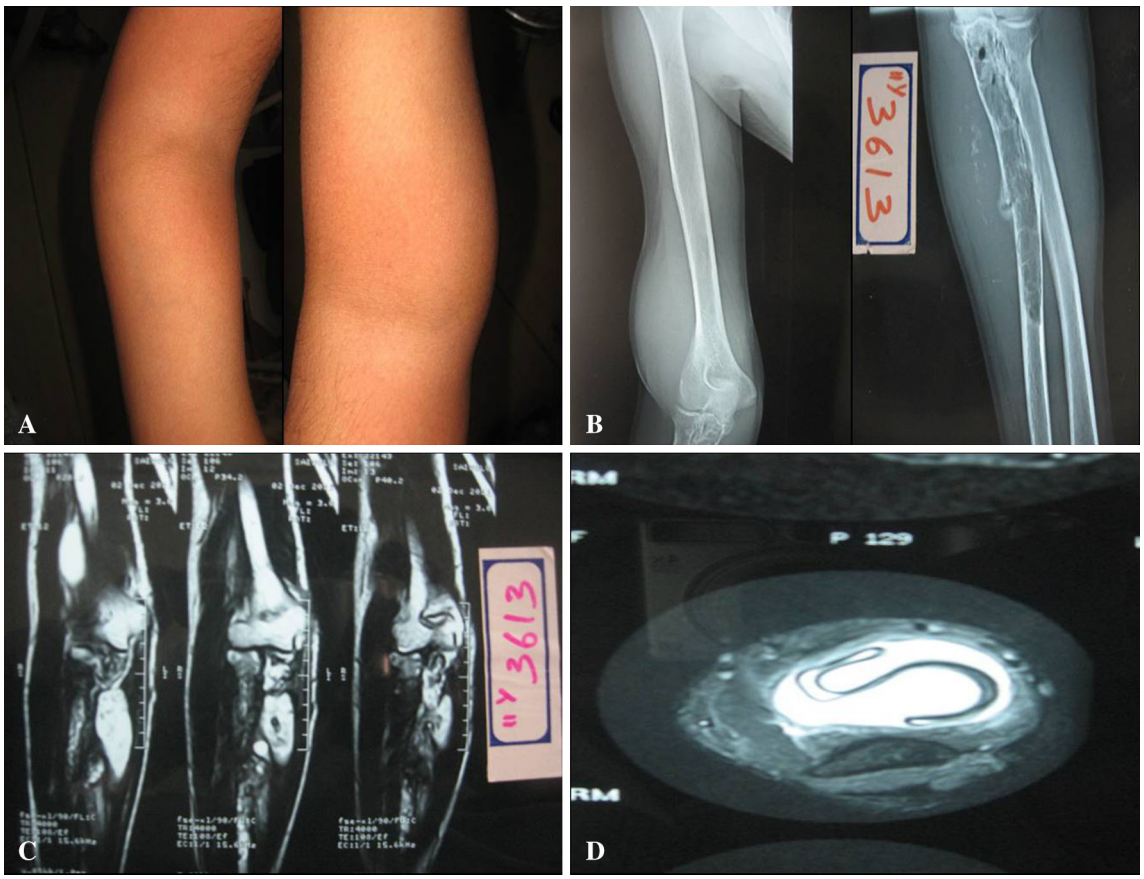

Fig. 1 **a** Swelling in right arm and forearm. **b** Radiograph showing lytic lesion in ulna with ground glass matrix and endosteal scalloping in the distal end of humerus. **c** Coronal T2 weighted MRI of right upper arm shows a lesion with hyperintense signal lateral to distal metadiaphysis of humerus and proximal end of ulna. **d** MRI STIR axial image through the soft tissue arm and ulna shows hyperintense lesion with curvilinear membrane

Discussion

Hydatid disease is distributed widespread in Mediterranean region, Asia, South America, Europe and Australia (Kireşi et al. 2003). The highest prevalence in India has been reported from Andhra Pradesh, Saurashtra, and Tamil Nadu. The most common sites of hydatidosis are liver and lungs and may be explained by filtering efficacies of hepatic and pulmonary barriers. Several reports of musculoskeletal hydatid cysts are summarized (Table 1) (Drimousis et al. 2006; Kocakusak et al. 2004; Tatari et al. 2001). It accounts for 1–5.4 % of all cases of HD. These cases are associated with involvement of other visceral organs like liver or lungs. Isolated musculoskeletal HD in absence of visceral involvement is very rare. Various case reports of HD diagnosed on FNA cytology have been described well. However an extensive literature search has revealed only rare reports of FNA diagnosis of primary musculoskeletal HD. It may be attributed to unfavorable environment due to excessive lactic acid in muscles. Gupta et al. (2008) reported two cases of primary soft tissue hydatidosis diagnosed on FNAC. Previous studies have described involvement of chest wall, sartorius, biceps brachii, supraspinatus and gluteus muscles in HD (Mohan Rao et al. 2011). The cysts lodge into bone due to high vascularization viz. vertebra, long bone epiphysis, skull, ribs etc. Polat et al. (2003) describe hydatid involvement in different large and small bones. The present case showed primary involvement of ulna and soft tissues of arm. The parasite replaces the osseous tissue over a period of time and destroys the cortex. It then spreads from bone to the surrounding tissue such as muscles and simulate soft tissue neoplasm (Polat et al. 2003).

FNAC is a rapid and sensitive method of demonstration of hooklets in necrotic lesions of HD (Babu et al. 2008). Cell blocks can be useful adjuncts to smears for establishing a more definitive cytological diagnosis especially when large quantity of fluidy aspirate produce dilution effect on cytology smears. This procedure is highly recommended in situations where there is discrepancy between imaging and serological results pertaining to unusual locations. In the present case cell block clinched

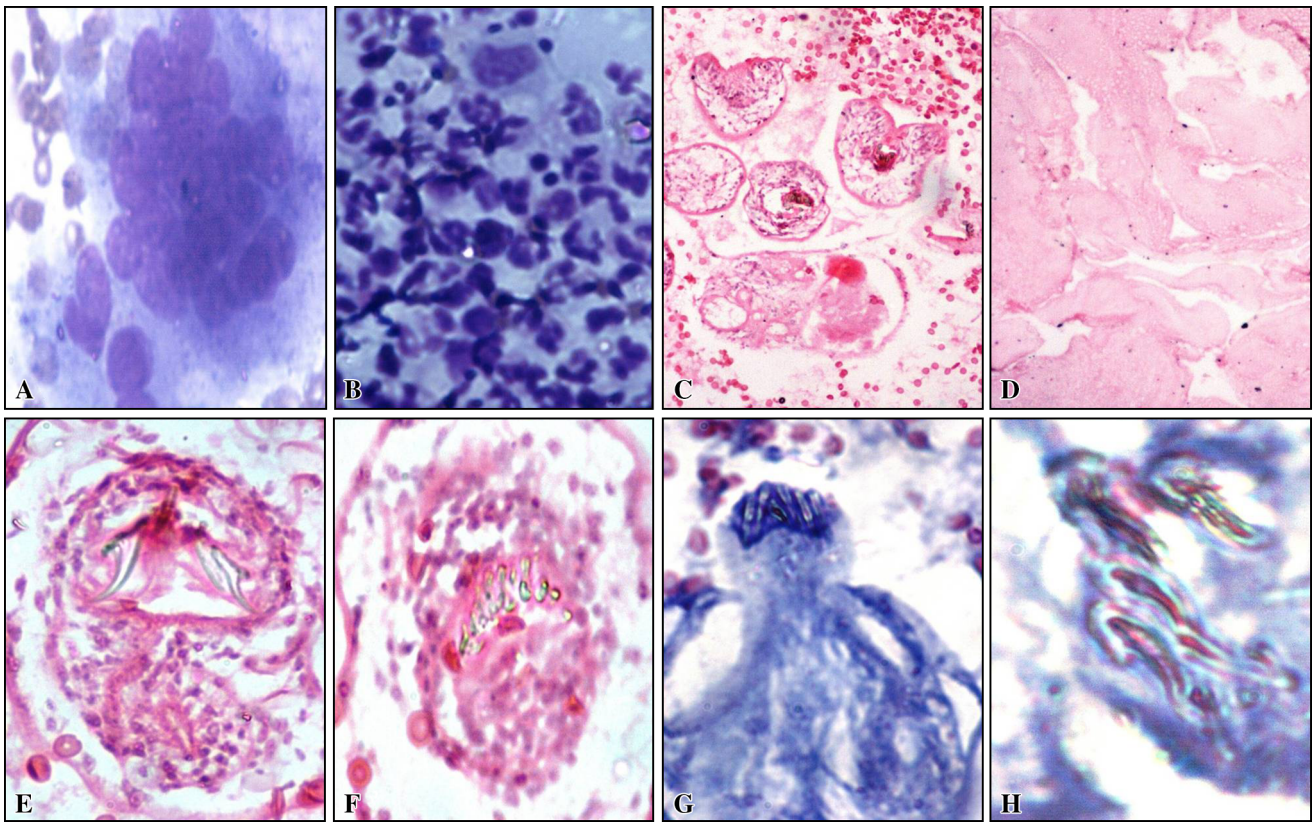

Fig. 2 FNAC **a** osteoclastic giant cell (MGG × 400×). **b** Dense collection of acute inflammatory cells (MGG × 100×). **c–f** Cell block preparation. **c** Numerous scolices (H&E × 400×). **d** Acellular lamellated membrane (H&E × 400×). **e**, **f** Scolices with hooklets, seen in aligned two rows (H&E × 400×). **g**, **h** Acid fast core of hooklet (Ziehl Neelsen Stain × 1000×)

Table 1 Reported cases of musculoskeletal Hydatidosis (Merkle et al. 1997; Rieber et al. 1989; von Sinner et al. 1991; Torricelli et al. 1990; Kocakusak et al. 2004; Tatari et al. 2001)

Authors	Number of cases	Sites of involvement
Merkle et al.	8	Iliopsoas, left adductor, left femur, gluteus
Rieber et al.	1	Paravertebral structures
von Sinner et al.	1	Pelvic
Torricelli et al.	14	Bone with adjacent soft tissues
Kocakusak et al.	1	Vastus lateralis
Tatari et al.	1	Supraspinatus

the final diagnosis owing to conclusive evidence of lamellated membranes as well as hooklets and scolices. Cell block preparations from aspiration samples are thus a simple way to provide additional information as well as increases diagnostic yield by allowing a larger amount of material for study.

It was believed that aspiration of hydatid cysts were exceedingly dangerous due to dissemination of the disease However, recent studies have shown that though there is 1 % risk of leakage and anaphylactic shock during aspiration this can be minimized by using a thin bore needle. There was no spillage of contents or allergic reaction following aspiration in our case.

Musculoskeletal HD is often misdiagnosed as soft tissue tumors because of lack of specific clinical symptoms and radiological signs. In case of extremity involvement, the common finding is the palpable soft tissue mass (Basarir et al. 2008). The present case simulates clinically as a soft tissue tumor. On ultrasonography muscular hydatidosis present as multiseptate cystic mass showing daughter cysts, floating membranes, calcification and hydatid sand (Loudiye et al. 2003). Bony involvement mimics osteomyelitis in early stages of the disease and in later stages the cyst progressively enlarges filling the medullary cavity. Erosion of bone leads to osteolysis and it radiologically mimics aneurysmal bone cyst, giant cell tumor, cystic metastasis and fibrous dysplasia. Fibrous dysplasia can closely resemble long-standing cases of HD in which the marrow cavity is extensively invaded by the parasite. These lesions can also present as multilocular lesions as in the present case. As the cortex is eroded the lesion extends into the surrounding soft tissue. Early recognition and extraosseous extension of HD on computed tomography (CT) and magnetic resonance (MR) imaging are possible. The classic

MR findings include a multivesicular cyst with a low intensity "rim sign" on T2-weighted images representing ectocyst (Kireşi et al. 2003).

Serological tests may aid in diagnosis but are not always positive in all histopathologically proven cases. Hence, negative test does not rule out the diagnosis of echinococcosis (Loudiye et al. 2003). Serological cross reactivity is known between *E. granulosus* and *Echinococcus multilocularis.* In the present case positivity for cysticercal antigen by ELISA may be due to cross reactivity for cystodal antigens. Arazi et al. (2005) found positive indirect hemagglutination test in 27 % cases in their case series of musculoskeletal echinococcosis. Imaging remains more sensitive than serodiagnostic techniques and characteristic scan in the presence of negative serologic results still suggests the diagnosis.

Conclusion

FNAC and cell block aids in the diagnosis of hydatid cyst in unusual locations in soft tissues and bone where clinical presentation mimics tumor. This is second report on bone involvement aspiration cytology and we emphasize utility of cell block preparation which clinches the diagnosis of musculoskeletal HD. Once the diagnosis is established, the appropriate surgical management minimizes recurrence.

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