CASE REPORT

Primary hydatid cyst presenting as a mass in the supraclavicular region: an unusual case report and literature review

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Abstract

Hydatid disease, also known as echinococcosis or hydatidosis, is an infectious disease caused by the cestode Echinococcus. Echinococcus granulosus is the most common Echinococcus species affecting human beings. It may affect any organ and tissue in the body, in particular the liver and lung. Musculoskeletal or soft tissue hydatidosis accounts for about 0.5%-5% of all echinococcal infections in endemic areas, and is almost always secondary to the hepatic or pulmonary disease. Even in regions where echinococcosis is endemic, hydatidosis of cervicofacial region is extremely rare. Herein, we present an exceptionally rare case with an unusual localization of primary hydatid cyst in the right supraclavicular region of the neck without involvement of the lungs or pleura.

Keywords: Hydatid disease, Echinococcus granulosus, Supraclavicular region.

Introduction

Hydatid disease, also known as echinococcosis or hydatidosis, remains a serious health problem in endemic countries, like India. It is a zoonotic infection caused by the larval forms (metacestode) of Echinococcus granulosus that lives in the small intestines of adult dogs.[1] In humans hydatid disease commonly involves the liver (70%) and the lungs (25%).[2] The spleen, kidneys, bile ducts, mesentery ,heart, brain and muscluloskeletal or soft tissues are less frequent sites of involvement.[2,3] Definite diagnosis is mostly based on cross-sectional imaging techniques such as ultrasound (US), computed tomography (CT) or magnetic resonance imaging (MRI).[3]

Even in regions where echinococcosis is endemic, hydatidosis of the neck is rare and its incidence is unknown. In this report, a patient with an unusual localization of hydatid cyst in the right supraclavicular region of the neck without involvement of lung or pleura is discussed.

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Case Report

A 28 year female presented to our department with chief complaints of slowly progressive, painless swelling in right supraclavicular region for 6 years. On examination a 6x3cm, soft, non tender, transilluminant, non compressible and non expansile solitary swelling in right supraclavicular region was seen. Computed tomography showed a well defined, multilocular, cystic lesion in the right supraclavicular fossa (Figure-1a). Magnetic resonance imaging showed multilocular cystic lesion in the right supraclavicular fossa, with clavicle, subscapularis and scalenus antecus as anterior, posterior and medial limitations with preserved fat planes (Figure-1b and c.) Ultrasound abdomen and Chest x- ray were negative for any liver and lung cystic lesions. FNAC was consistent with hydatid cyst. The patient was given tablet Albendazole 400 mg twice a day for 2 months. Partial response with medical treatment in the form of reduction in size was seen, hence surgical excision was undertaken. Intra operatively there was a thick walled, multilobulated cyst in the supraclavicular region within the muscles of scalenus anterior and posterior and going underneath the

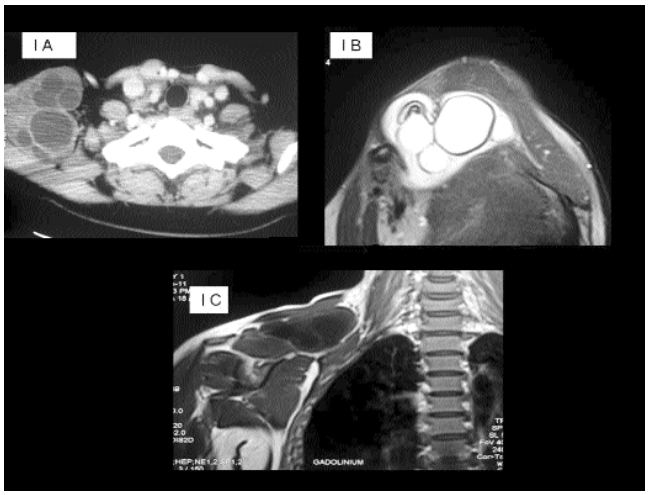

Figure-1: (a) Contrast enhanced CT scan, axial images ,showing well defined multiloculated cystic lesion in the lower right neck infiltrating muscles. (b) T2 weighted MRI, axial image, showing well defined multiloculated cystic lesion consistent with diagnosis of hydatid cyst. (c) T1 weighted MRI, coronal image showing multiloculated cyst lower part of the neck.

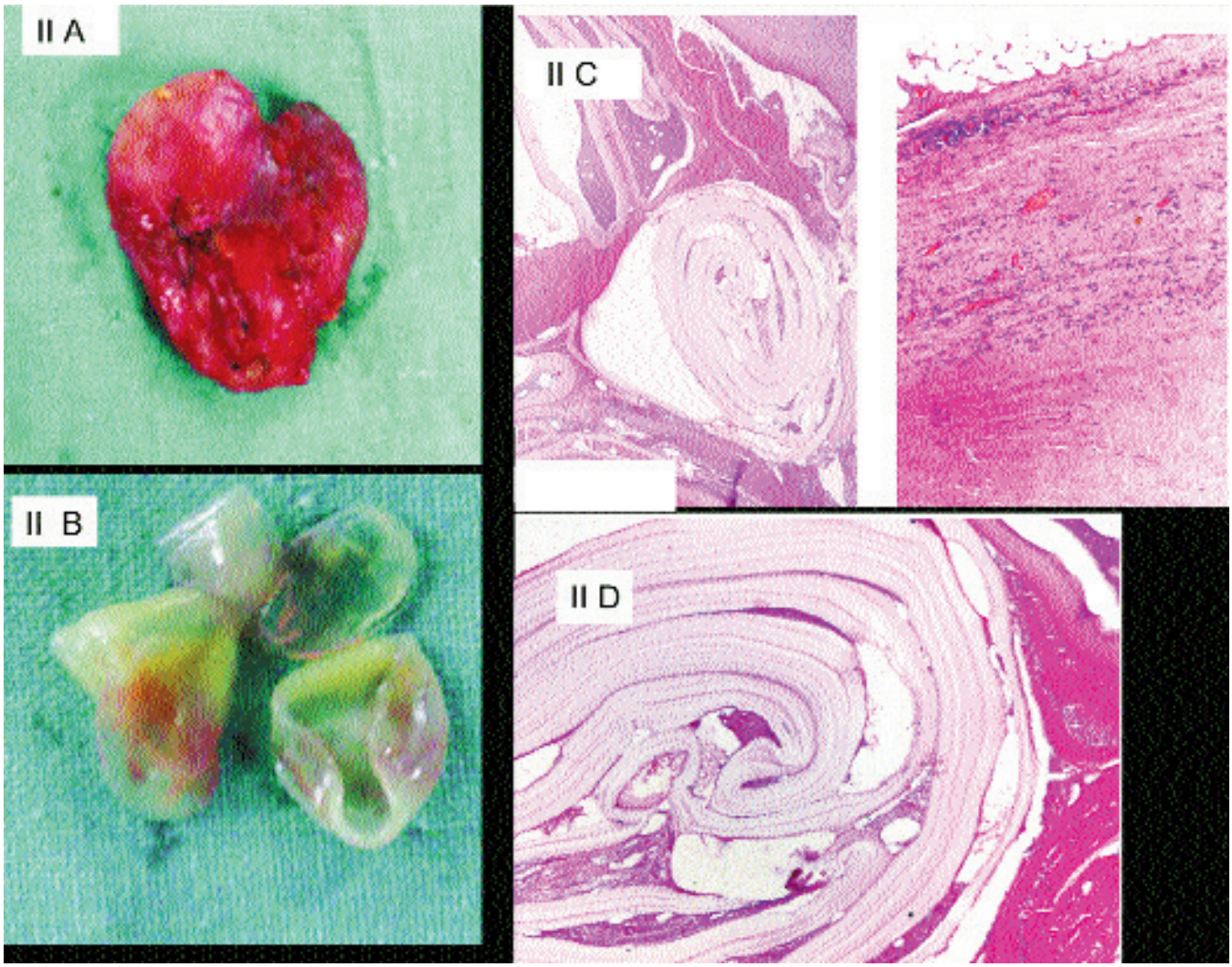

Figure-2: (a) Resected specimen. (b) Showing daughter cysts after cutting open the specimen. (c) Photomicrograph showing eosinophilic acellular laminated. Ectocyst focally lined by inner germinal layer. (d) Showing pericyst composed of fibrocollagenic tissue with inflammatory infiltrate (Haematoxylin & eosin, x200).

deltoid muscle. The cyst was excised in toto (Figure-2a, b) and sent for histo-pathological examination which was consistent with hydatid cyst (Figure-2c, d). Post operative check CT scan was showing complete excision of the cyst. At 6 months follow-up, the patient is absolutely asymptomatic.

Discussion

Hydatid cyst is an infectious disease which is most commonly caused by the cestode parasite (tapeworm), Echinococcus granulosus and less commonly by echinococcus multilocularis.[4] Dogs being the main host and cattle, sheep, horse and pig act as intermediate host. Parasite eggs that penetrate the organism hatch in the small intestine of the main host, pass into portal venous system or lymphatic system and reach the liver and lungs, and finally form hydatid cyst lesions. Moreover, they can cross the hepatic sinusoid or pulmonary capillary barriers, and embryos get into systemic circulation and can settle in any of the organs and structures in the body.[1,4] Hydatid cyst frequently involves the liver and lungs, and rarely the bones, brain, eye, heart, kidney and spleen.[3] Atypical localization of hydatid cyst may challenge the diagnosis.[2] Although the disease is generally asymptomatic, it may exhibit clinical symptoms depending on the size and location of the cyst, and the pressure of the growing cyst.[4] In our patient, there were no symptoms except for pain in upper arm and shoulder and mobile lump in the right supraclavicular area.

Detailed patient's history ,thorough physical examination, radiologic imaging including ultrasound, computed

tomography (CT) and MRI helps in diagnosis. Indirect haemagglutination, latex agglutination and ELISA can give false positive and false negative results. We performed ELISA which was not suggestive of hydatidosis. Abdominal and chest X-rays, ultrasound and CT scans should be performed to rule out involvements of other organs, particularly liver and lungs.[3]

Clinical symptoms depend on the anatomic host area. It may imitate benign and malignant tumours, cysts, abscess, haematoma, pseudocyst and congenital cysts.[6,9] Beji et al[6] reported a case of cervico-mediastinal hydatid disease presenting with a right-lower-neck mass. Alvarez et al[7] reported a case of primary mediastinal hydatid cyst presenting with a painful right supraclavicular mass, dyspnoea on exertion and dysphagia.

Fine neede aspiration has been carried out in our patient due to a low index of suspicion. It can theoretically lead to rupture of the cyst. In literature fnac is still being carried out routinely to prove the diagnosis[8] and in some cases as a therapeutic measure.[9] No fnac related complications were noted. In the present study fnac resulted in a decrease in size of the swelling thereby assisting in surgical excision. No complications following fnac were noted.

Total surgical excision of the cyst without rupture is the single effective treatment to prevent recurrence and to get excellent prognosis.[3,10,11] Albendazole can be used postoperatively and preoperatively to reduce the size of the cyst.[10,11]

Conclusion

Hydatid disease is a widespread public health problem in developing countries. The possibility of hydatid disease, especially in endemic regions, should always be kept in mind. Radiologic imaging modalities in such cases are mandatory for the diagnosis of unilocular or multilocular hydatid cyst with thin borders, thin walls, inner membranes, and a distinct appearance of characteristic cystic mass. The prognosis is excellent in hydatid cyst cases treated with total removal of the cyst without rupture.

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