Primary hydatid disease of breast: a case report

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Received: 17 April 2015 / Accepted: 1 March 2017 / Published online: 11 March 2017
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Abstract Human hydatid disease, or echinococcosis, is a helminthic infection that leads to the formation of fluid-filled cysts in the liver, lungs and other organs. Breast, is a rare primary site of hydatid disease. We report a case of a 45 years old female who presented with a breast lump of 2 years duration. On fine needle aspiration cytology a diagnosis of fibrocystic changes of the breast was made and ultrasonography breast showed a thick walled infected cystic lesion. Intraoperatively, a diagnosis of hydatid cyst was made which was confirmed on histopathology.

Keywords Hydatid disease · Breast · Histopathology

Introduction

Echinococcosis is a zoonotic infection caused by the larval stages of tapeworms belonging to the genus *Echinococcus*, which are parasites of animals (Beaver et al. 1984; Gutierrez 1990; Thompson 1986). The adult worms parasitize the intestine of wild and domestic dogs and cats. The intermediate hosts are many, ranging from sheep, goat, cattle, horses to medium sized and small rodents depending

on the species (Gutierrez 1990). Human beings and a variety of other animals are accidental intermediate hosts (Helminths 1980). *Echinococcus granulosus* produces the most common form of hydatidosis in humans, referred to as “unilocular hydatid disease”.

The incidence of the parasite differs from country to country. The incidence is higher in the sheep-raising, developing and underdeveloped countries around the Mediterranean sea, the Middle East, South America and Asia (Romero-Torres and Campbell 1965). Insufficient hygienic conditions promote the spread of the disease. Our state Kashmir located in India is endemic for hydatid disease (Khuroo 2002; Wani et al. 2012).

This disease is endemic in cattle and sheep rearing regions of the world with India being one such country having high prevalence in Kashmir, Andhra Pradesh, Tamil Nadu and central India (Salati et al. 2008; Wani et al. 2014; Tyagi and Gaeg 2014; Akhter et al. 2011).

Cystic echinococcosis is a public health problem in Kashmir, North India, owing to the ongoing transmission and asymptomatic infection in most areas of the state (Fomda et al. 2015).

Although the parasite can grow in any part of the body, breast is a rare primary site of involvement mainly reported in literature as case reports only (Alamer et al. 2013).

Case report

45 year old female, a known case of hypothyroidism and hypertension on treatment presented with the complaints of swelling in the right breast associated with mild pain for the past 2 years. The swelling was gradually increasing in size without any history of fever or nipple discharge. On examination there was a large

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swelling about 5×4 cm in the right breast, involving the upper outer quadrant. The swelling was soft to cystic in consistency with mild tenderness, and free overlying skin. There was no lymphadenopathy in the right axilla and the contralateral breast and axilla were also normal.

Fine needle aspiration cytology (FNAC) was done which yielded 60 ml of clear fluid and a diagnosis of fibrocystic changes of the breast was made on microscopy (Figs. 1 and 2).

Ultrasonography (USG) breast revealed a thick walled round to oval complex infected cystic lesion measuring $69 \times 61 \times 54$ mm in the outer quadrant of right breast with internal septations and associated inflammatory changes in the adjacent breast parenchyma (Fig. 3). A USG guided aspiration of cyst fluid was done for biochemical analysis and detection of mycobacterium tuberculosis (MTB). Biochemical analysis of the cyst fluid revealed the presence of protein, LDH and amylase. PCR for MTB DNA was negative. Routine hematological investigations like CBC, LFT and KFT were within the normal limits. Chest radiography & USG abdomen were unremarkable. The cyst along with the adjacent inflammatory tissue (pericyst) were removed subsequently. Grossly, the cyst was pearly white in colour measuring 7×7 cm and the inner surface was covered with sand like particles (Fig. 4). The pericyst measured 6×3 cm with irregular inner surface. On Histopathological examination of the specimen the lamellated membrane of hydatid cyst was seen with chronic inflammatory infiltrate rich in eosinophils in the surrounding fibrocollagenous tissue (Fig. 5). Thus, a diagnosis of hydatid breast was made (Fig. 1). The patient was discharged without any post-operative complications.

Fig. 1 FNAC showing apocrine cell groups in a fluid background

Fig. 2 FNAC showing macrophages and few apocrine cells against a fluid background

Fig. 3 Oval complex cyst with internal septations

Fig. 4 Pearly white hydatid membrane with surrounding pericyst

Fig. 5 Lamellated membrane of hydatid with scolex

Discussion

Hydatid cyst is due to *Echinococcus* species, a Cestode parasite commonly known as the small tape worms of carnivorous animals. There are predominantly two species affecting humans; *E. granulosus* and *E. multilocularis* causing Cystic Echinococcosis (Hydatid disease) and Alveolar Echinococcosis respectively (Khuroo 2002). The other two species found very rarely in humans are *E. vogeli* and *E. oligarthrus*. This infection produces the important cestode disease in humans, not only because of the number of people infected around the world, but also because of the resulting morbidity and mortality. The adult worm resides in the intestine of dogs and cats. Sheep, cattle, goat etc. are intermediate hosts. Humans are accidental intermediate hosts and are infected by ingesting the eggs of the parasite. The most common sites of human disease are the liver [$>65\%$] and lungs [20%] (Khader et al. 2013; Eckert and Deplazes 2004; McManus et al. 2003). Other less commonly affected areas include other intra-abdominal sites such as the kidneys, spleen, and the peritoneal cavity. Occasionally, spine, brain, heart and bone maybe affected (Wani et al. 2012; Khader et al. 2013). Breast, is a rare site of involvement accounting for only 0.27% of all cases of Hyadid disease (Vege et al. 1994; Tukul et al. 1997; Taori et al. 2004). Classically, the patient presents with a long standing history of breast lump, which should be differentiated from fibroadenoma in the young and from a carcinoma in the older patients. Hydatid cyst in the breast usually appears as a mass on physical examination. Ultrasonography and mammography are very effective in the evaluation of this mass. Despite its high cost, MR imaging has also been used in further evaluation of the mass (Tukul et al. 1997). The definitive diagnosis is made by histopathological examination of the cyst. The morphology of the cyst is characteristic. Grossly, the cyst varies in size and appearance depending on the viability but

usually three distinct layers are apparent. The outer layer (pericyst), is a fibrous reaction of the tissues to the cyst. The second, the acellular or lamellated layer (ectocyst), is produced by the parasite and is the most characteristic because it is white, with a rather uniform thickness, made of a friable substance resembling the albumin of a boiled egg. The third layer (endocyst), is the germinal layer, seen better in viable cysts as a thin, semi transparent membrane from which the larvae grow. Microscopically, the cyst has the same three layers described grossly and a slight to moderate chronic inflammatory infiltrate around the fibrous layer (Gutierrez 1990; Thompson 1986). Complete excision of the cyst without spillage can be a curative procedure for primary hydatid cyst of the breast.

We conclude, that although breast is a rare site of primary hydatid disease even in the endemic areas, it should always be kept as a differential diagnosis in the cystic lesions of the breast. Awareness of this entity can help in arriving at a pre operative diagnosis, even without cytological examination, which is important in the management of the cyst.

Acknowledgements Funding was provided by Dr Sheema Sheikh.

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