Primary Orbital Hydatid Cyst: A Case Report

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Abstract- Hydatid cyst is a rare parasitic infection that involved all organs. It caused by the larval stage of Echinococcus tapeworms. Hydatid cyst of the head and neck is a very rare condition, even in areas where Echinococcus infestation is endemic. Orbital hydatid cyst is extremely rare and accounts for less than 1% of all hydatid cysts. Herein a 24-year-old man with primary orbital hydatid cyst is introduced. He complained from proptosis and diplopia. MRI images revealed a lesion with low signal intensity on T1-weighted images and high signal intensity on T2-weighted images, which displaced the optic nerve inferiorly and the globe inferolateral. The cyst was enucleated via frontotemporal craniotomy and superior orbitotomy approach. Histopathological examination of the fluid confirmed the diagnosis of hydatid cyst. Treatment of the orbital hydatid cyst is surgical excision followed by the systemic use of albendazole.

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Keywords: Hydatid cyst; Exophthalmos; Echinococcus

Introduction

Hydatid cyst is a rare parasitic infection that involved all organs. It caused by the larval stage of Echinococcus tapeworms. It is commonly seen in some cattle and sheep raising regions of the world like Africa, the Middle East, Australia and South Africa (1,2). Although hydatid disease affects any organ or soft tissue, liver (60-70%) and lungs(30%), followed by the CNS are the most frequent sites of hydatid cyst (3,4). Orbital hydatid cyst is extremely rare and accounts for less than 1% of all hydatid cysts (2,5). The most common presentation of hydatid cyst of the orbit is proptosis which forms an important differential diagnosis of unilateral exophthalmos in patients. In this paper, we report a new case of orbital hydatid cyst that we removed through a combination of intra and extra cranial approach.

Case Report

A 24-year-old man referred to the maxillofacial clinic with complaints of exophthalmia on his right eye with duration of 4 months (Figure 1). He had no pain and tenderness. He had moderate proptosis and diplopia in the upward gaze. His visual acuity was 8/10 in his right eye and 10/10 in the left side. Fundoscopic examination by ophthalmologist indicated that the optic disc of the right eye was mild pale and atrophic whereas the left one was normal. There were no any other abnormalities in his systemic examination. Axial, Sagittal, and coronal MRI images revealed a lesion with low signal intensity on T1-weighted images and high signal intensity on T2-weighted images, which displaced the optic nerve inferiorly and the globe inferolateral (Figure 2). Laboratory tests were normal. Indirect hemagglutinin test was performed to exclude parasitic disease, but it was negative.

The patient candidate for enucleated of the cyst via a left frontotemporal craniotomy and superior orbitotomy approach. A unilocular cyst was found in the superomedial aspect of the orbital cavity. An aspiration with a syringe was done, and a transparent clear fluid was aspirated which is characteristic for a hydatid cyst. The cystic wall ruptured accidentally during the dissection. The liquid contents were aspirated, and the outer thick fibrous wall was removed. The site of the operation was washed for five minutes with hypertonic saline solution. After hemostasis, the bone was fixed with miniplate in its position and flap sutured. Post-operatively, the patient received oral albendazole,

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antibiotic and steroid.

The postoperative period was uneventful. Histopathological examination of the fluid confirmed the diagnosis of hydatid cyst. His right side proptosis was completely resolved within 10 days after surgery (Figure 3). Ultrasonography of the abdomen and chest X-ray were done after operation to rule out systemic involvement, but they were normal. He discharged from the hospital 6 days after the operation. The patient was advised to maintain regular follow up constantly.

Figure 1. Right exophthalmia with 4 months duration

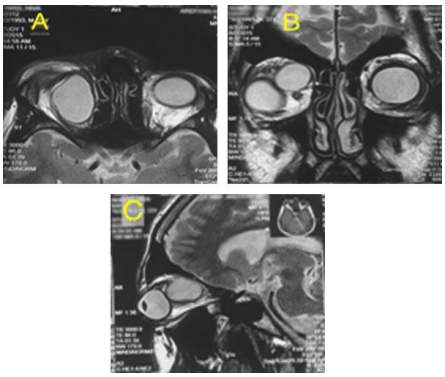

Figure 2. Axial (A), coronal (B) and Sagittal (C) MRI images revealed a lesion with low signal intensity on T1-weighted images and high signal intensity on T2-weighted images, which displaced the optic nerve inferiorly and the globe inferolateral

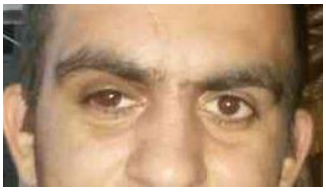

Figure 3. Proptosis was resolved in 10 days after operation

Discussion

Hydatid disease is a worldwide health problem that is endemic in many parts of the world. This disease is prevalent in Africa, the Middle East, Eastern Europe, Australia and Mediterranean countries (1,2). This disease is caused by Echinococcus. The three types of echinococcosis are cystic echinococcosis caused by E. granulosus, alveolar echinococcosis caused by E. multilocularis, and polycystic echinococcosis caused by E. vogeli or E. oligarthrus (6). The most common cause of orbital hydatid cyst is Echinoccus granulous (7). The definitive host is a dog and the adult worm of the parasitic lives in the proximal small intestine of these animals. The eggs are released into the environment by the feces and ingested by the intermediated hosts such as sheep and cow. The cycle is completed by a definitive host such as dog; eat the viscera of the intermediate host. Human is the accidental host in the life cycle of Echinoccus by the ingestion of contaminated vegetables. The most involved organ is the liver bye destination via the portal system (6o-70%) (1).

Orbital involvement is rare and takes place in 1% of all cases (2,7,8). Orbital hydatid cyst can involve patients at any age, but it is more prevalent among young patients with no sex predilection. The most frequent clinical finding is nonpulsatile, nontender exophthalmia (in our case). The degree and direction of the proptosis depend on the location of the cyst. Other clinical presentation includes chemosis, lid edema, visual impairment and restriction of extraocular mobility. Lid edema and limited mobility were not all present in our patient whereas mild visual impairment and proptosis were. An orbital hydatid cyst may involve different locations within the orbit. It is well accepted that the most frequent sites are the superomedial or superolateral angle of the orbit (9). In our patient hydatid cyst was found within medial rectus muscle. Left-side lesions were more common than right-side ones because left common carotid artery arises directly from the summit of the aortic arch (10). Although the orbital imaging finding suggests the diagnosis of hydatid cyst, the exact diagnosis is confirmed by pathologic and clinical evaluation. The various radiological investigations including ultrasound (US), CT scan and MRI in correlation with MRI Spectroscopy help to confirm the correct diagnosis. MRI is the choice diagnostic imaging in orbital hydatid cyst.it appears as a well-defined fluid-filled cyst which is thin-walled and usually unilocular.

Magnetic resonance imaging demonstrates rates a cystic lesion with long signal intensity on T1-weighted images and high signal intensity on T2-weighted images (11). Orbital abscess, epidemiologic cyst, mucocele, hematic cyst, lymphangioma, and teratoma are being considered as differential diagnosis of hydatid cyst in orbital imaging. Casoni's test was found to be negative in our case, and it is now widely accepted that Hematologic and serologic (Casoni's test) findings are not reliable in orbital hydatid cyst due to the high rate of normal reports (9).

Definitive treatment in orbital hydatid cyst is total

surgical excision. Various surgical approaches have been used to access thy cyst, including transcranial, transconjunctival, orbitotomy, transmaxillary approach (12). The use of these approaches depends on the location and size of the cyst as the skill and ability of the surgery. We used fronto-orbital approach because of the location of the cyst. The most complication during the surgical treatment of hydatid cyst is rupturing of the cyst and spillage of its content which leads to anaphylaxis and secondary local recurrences (13). Complete aspiration of the liquid and in situ irrigation with hypertonic saline or hydrogen peroxide is necessary. The cyst ruptured during removal in our case but fortunately didn't happen anaphylaxis. We irrigated the site with hypertonic solution because of nontoxic feature of the neural tissue.

The standard therapy of hydatid cyst is surgery but, Adjuvant medical therapy is the important step in the treatment of the hydatid cyst to decrease the number of relapses and hydatid cyst size (14). Albendazole is the choice in this disease that starting 2-4 weeks before surgery which is more effective if combined with praziquantel (15). In some cases as ours, diagnosis was made during operation, so medical treatment starts postoperatively. In our case albendazole had been prescribed with the dosage of 10 mg/kg daily for 12 weeks.

This case indicates that orbital hydatid cyst should be considered in the differential diagnosis of unilateral proptosis especially in patient living in the endemic region.

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