Primary Intraorbital Hydatid Cyst in a Child

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Abstract: Intraorbital hydatid disease is quite rare and its incidence has been reported to range from 0.3% to 1.0% among patients with Echinococcosis. The authors report a 4-year-old boy with primary intraorbital hydatid cyst who had presented with proptosis of the left eye. Because of high risk for cyst rupturing, the cyst was aspirated-injected simultaneously and reaspirated and then removed totally.

Key Words: hydatid cyst, intraorbital, child

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Hydatid disease is caused by the larval form of *Echinococcus granulosus*.[1,2] Orbital localization is very uncommon and only few cases have been reported in the literature. The incidence of orbital hydatid disease has been reported to range from 0.3% to 1.0% among patients with Echinococcosis.[3] The symptoms include progressive proptosis with or without pain, disturbance in ocular motility, visual deterioration, diplopia, eyelid edema, conjunctivitis, and chemosis.[1,3,4] Surgery is the primary treatment of choice in these cases but complete excision of the lesion without rupture from the intraobital region is difficult so chemotherapy should be applied if a cyst ruptures.[4,5]

CASE REPORT

A 4-year-old boy was admitted to neurosurgical department with a 4-week history of proptosis of the left eye. His physical examination revealed moderate left eye proptosis. A neurological examination showed movement restriction of the left eye. There were no specific laboratory signs of hydatid disease. Serological tests for hydatid disease were negative. The eosinophil count was within normal limits. Chest radiography and abdominal ultrasonography were all normal. Cranial magnetic resonance imaging (MRI) demonstrated a well-defined retrobulbar homogenous unilocular lesion in posterior-superior of the left orbit which was hypointense on T1-weighted images and hyperintense on T2-weighted images (Figs. 1A, B). The patient underwent surgical resection of the cyst via left frontotemporal craniotomy and orbitotomy. Intraoperatively, the outer capsule of the exposed cyst was identified and an effort was made to dissect it from the retrobulbar tissues by using hypertonic saline-soaked cotton patties. Because of the high risk of the cyst rupture, although cyst fluid leakage aspirated by injector, hydrogen peroxide was injected to cyst simultaneously by other injector. This way we aimed to reduce the effect of the cyst content. Then the cyst was reaspirated, membrane was dissected and removed totally (Fig. 1D). At the end of the surgery, operative field was thoroughly irrigated with 9% hypertonic saline solution. The proptosis and movement restriction of the left eye resolved completely after surgery. Postoperative cranial computed tomography demonstrated no evidence for cyst (Fig. 1C). The patient was maintained on Albendazole treatment (10 mg/kg twice daily). Histopathologic examination revealed a hydatid cyst. The postoperative period was uneventful.

DISCUSSION

Hydatid cyst is a cyclozoonotic infection of the larvae form of *E. granulosus*,[1,2,6] which is rarely seen in organs other than the liver (60%) and lungs (20%).[4] It results from the ingestion of food contaminated with eggs of tapeworm *E. granulosus*. Human being is the intermediate host.[2] It may involve almost every organ or tissue in the body via portal and systemic circulations.[4] Echinococcosis is endemic to Africa, the Middle East, the Mediterranean region, New Zealand, and South America.[6] Orbital hydatidosis represents a quite rare clinicopathologic entity and its incidence varies from 0.3% to 1.0% of all hydatid disease cases. It can be differentiated to primary, where cyst or cysts are solely located into the orbital cavity and secondary, when orbital lesions represent a component of disseminated multiorgan systemic disease.[3] This case was accepted as a primary infection because of no findings in laboratory and radiologic examinations and previous history of liver and lung cysts. The most commonly clinical findings are exophthalmus, papilledema, diplopia, chemosis, conjunctivitis, visual impairment, and restriction of extraocular motility.[1,3–5] Our case had proptosis with movement restriction of the left eye. The diagnosis was based on the history of the patient, physical examination, imaging, and serological tests. There is an indisputable superiority of MRI to computed tomography in the detection of the lesion and the delineation of its relationship to the adjacent ocular structures. In addition, negativity of the serological tests cannot exclude the existence of solitary orbital hydatid cyst.[3] Although serological tests were negative, cranial MRI demonstrated a hydatid cyst in the left orbit in our case.

Definitive treatment of a hydatid cyst is total surgical excision without rupture and for this aim various surgical approaches have been used.[1–5] Each approach carries certain advantages and disadvantages. The anatomic location of the cyst, its size, its extension into the cranial cavity, and experience of the surgeon's are the important factors for

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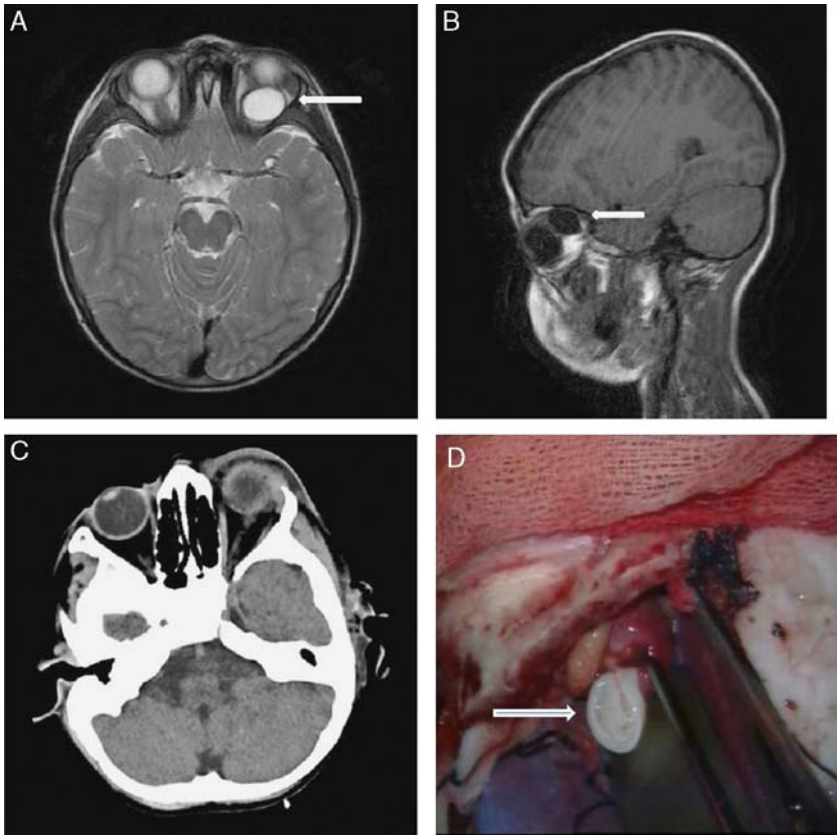

FIGURE 1. White arrows are showing a retrobulbar homogenous unilocular lesion in posterior-superior of the left orbit which was hyperintense on T2-weighted image (A) and hypointense on T1-weighted image (B). Postoperative cranial computed tomography demonstrated no evidence of the cyst (C). White arrow is showing total removal of the cystic membrane (D).

determining the most suitable approach.[2,3,5] Two main approaches, the transcranial and lateral orbital approaches are used to expose the intraorbital lesions.[2] The most important complication in surgical treatment is rupture of the cyst during excision, which can cause anaphylactic reaction or recurrences.[1–5] However, Selçuklu et al[5] reported that complete resection of the cyst without rupture is almost impossible. Turgut and colleagues reported that rupture or puncture rate was 64% on surgical exploration.[6,7] Irrigation is also a very important part of surgery and if the cyst ruptured accidentally the operative field should be irrigated generally with hypertonic saline, hydrogen peroxide, or formalin.[2–7] Also some authors recommend aspiration of the cyst to prevent rupture and thus to reduce the risk of anaphylaxis, incomplete removal, and secondary implantation.[8,9] We selected frontotemporal craniotomy. Although maximum effort was made to dissect it from the retrobulbar tissues, because of high risk for cyst rupturing, we aspirated cyst fluid leakage by injector and hydrogen peroxide was injected to cyst simultaneously by other injector. Then the cyst was reaspirated and membrane was dissected and removed totally. Postoperative adjuvant chemotherapy with Albendazole is recommended especially if begun 14 to 28 days before surgery.[2,3,5] We also administered Albendazole therapy to decrease the risk of relapse.

CONCLUSIONS

(1) We believe that surgical therapy is the treatment of choice in orbital hydatidosis, but complet excision of the lesion without rupture from the intraorbital region is difficult. So before removing the cyst, simultaneous aspiration-injection and then reaspiration could be applied in the presence of high risk for cyst rupturing.
(2) Increased migration and worldwide traveling has increased the importance of orbital hydatid cyst. So the physicians should keep this pathology in mind in the differential diagnosis of orbital mass, especially in nonendemic areas.

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