CASE REPORT

Primary Hydatid Cyst of the Neck

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Abstract

Hydatid cysts in the neck are relatively exceptional, even in areas where *Echinococcus granulosis* is endemic, such as Asia. Although liver and lung are frequent sites of involvement, it can involve all tissues, with neck remaining one of the most rare sites. It should come in the differential diagnosis of cystic lesion of neck, as the treatment options differ widely from common neck cysts. The role of radiological investigation is important and, in these cases, the involvement of other organs should be investigated. Serological tests may be helpful. The major treatment modality is surgical and the cyst should be excised as a whole, without being ruptured, to prevent any treatment complications, as the cyst fluid can initiate an anaphylactic reaction. Postoperative albendazole therapy is recommended particularly when there is intra-operative spillover. We report a case of an isolated hydatid cyst localized in the anterior triangle of the neck without any pulmonary or hepatic involvement.

Key Words: *Hydatid cyst. Neck cyst. Echinococcus granulosus.*

INTRODUCTION

Hydatid disease is a parasitic infestation caused by tapeworm *Echinococcus granulosis*. The adult phase of their lives is spent as parasites in gut of vertebrate animals. Although most cysts are caught in hepatic sinusoids, making it the most frequently involved organ,[1] few ova may pass through the liver and reach the general circulation to lodge in sites such as orbit, heart, lung, bones, bladder and other internal organs.[2-4]

Hydatid cysts in head and neck region are extremely rare, even in geographic areas in which echinococcal infestation is frequent.[2,5] The clinical course depends on the involved site, size and pressure caused by the enlarged cysts.[6]

Serological tests are widely used to confirm the diagnosis, although they are associated with false-negative and false-positive results.[7,8] Therefore, imaging modalities remain more sensitive than serodiagnosis, especially with unusual cyst locations and a characteristic scan should still suggest the diagnosis of echinococcosis.

CASE REPORT

A 7 years old boy presented in the Paediatric-surgical OPD in Liaquat National Hospital, Karachi, because of a slowly growing mass in the left side of the neck. His medical history was unremarkable. The physical examination revealed a 4.5 x 3 cms, painless immobile mass (cystic) in left anterior triangle of neck. The ultrasound examination showed a 4.5 x 3 cms cystic mass. Chest radiographs and abdominal ultrasound were normal. Serological tests for hydatid disease were negative. Patient underwent surgical intervention on 19th January 2011 and specimen was sent in Histopathology Laboratory of Liaquat National Hospital, Karachi.

On gross examination, a single egg shaped grayish white encapsulated cystic mass was seen of 4.5 x 3 cm. Cut surface was filled with clear fluid. Cyst wall was smooth and shiny and foci of pale granular areas. Representative sections were taken and submitted in two cassettes. Microscopically a benign cystic lesion was noted composed of external acellular cuticle and an inner cellular germinal layer where the scolices were attached (Figure 1). On the basis of these findings, hydatid cyst was confirmed histopathologically. The patient was put on Albendazole.

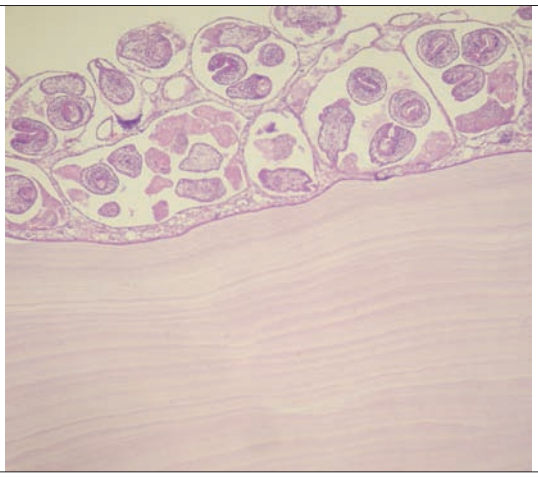

Figure 1: Upper half of figure shows cellular germinal layer and lower half demonstrates acellular cuticle (Hematoxylin and Eosin x 10).

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DISCUSSION

Hydatid cysts at unusual sites have been reported all around the world. Hydatid disease in the neck is quite rare and the diagnosis is remarkably difficult because of presence of simulating and more common cystic pathological conditions in the neck. The disease is endemic in the Mediterranean regions of the world and also in Australian and South Asian subcontinent. The liver is the most frequently involved organ (75%), followed by the lung (15%) and the remainder of the body (10%). The diagnosis of *Echinococcus granulosis* in an unusual location can be quite difficult. Involvement of sites other than liver and lung is rare but no site in the body is immune.[2,3] Among the other rare sites, bones, mostly the vascularized bones like vertebrae, long bones and epiphysis may be involved in 0.5 – 4% of the cases. Renal involvement is seen in 2 – 3% and brain hydatid cyst constitutes 2% of all intra-cranial space-occupying lesions in endemic regions. Cardiac hydatid disease occurs in 0.02 – 2% of cases. Other sites that may be involved include soft tissues, ovaries, pancreas, scrotum, inguinal canal and chest wall but neck is very unusual site of distribution.[1-5] This case was also missed because of the more common clinical differentials which were sebaceous cyst, branchial cleft cyst etc. If the patient had been diagnosed earlier than surgery by serological tests, the surgeon would have put him on anti-parasite drugs beforehand to decrease the chances of recurrence. As surgical rupture can cause parasitic cysts to develop, also chances of anaphylaxis are higher during the surgery. Luckily this patient did not develop either of these.

The serological tests including direct hemagglutination, latex agglutination, immunoelectrophoresis, skin tests and enzyme-linked immunosorbent assay are broadly used to substantiate the diagnosis.[8] However, because of chances of false positives and negatives, imaging modalities remain more sensitive than serodiagnosis, especially with unusual sites. However, in our patient, the ultrasound report did not diagnose the hydatid cyst, the unusual site being the reason behind the miss.

Histopathological evaluation of the excised specimen and the fine needle aspiration cytology usually leads to the diagnosis. Since puncture of the cyst may lead to an anaphylactic reaction due to spillage of hydatid fluid, the use of FNA is controversial at present.[9,10]

Few reports of infrequent incidence of hydatid cyst in different regions of neck have been published. These include report of hydatid cyst in lateral cervical region by Katilmis and Beji *et al.*[4] in submandibular gland by Memon[5], in the neck by Pampori[6] in parotid gland by Darabi[7] and two cysts in one patient, one in left submandibular gland, and the other in thyroid gland by Avcu *et al.*[8]

Just like our case, Iynen *et al.* also presented a case of 21-year-old woman with painless lump growing on the left lateral side of her neck.[10]

As already discussed, imaging modalities such as ultrasonography, CT and MRI are more sensitive than serologic tests, so these should always be kept in mind at unusual locations, to reduce the chances of morbidity and even mortality by anaphylaxis which is a major complication during intraoperative rupture. The preliminary diagnosis is confirmed by cytology or histology and in these cases; the involvement of other organs should be investigated. The cyst should be excised as a whole without being ruptured to prevent any complications.[9]

Treatment options for the uncomplicated hydatid cyst vary depending on site and size of cyst and these include needle aspiration under ultrasound guidance, laparoscopic approach, direct surgical intervention or medical treatment with the use of albendazole.[10]

In conclusion, hydatid disease should always be considered in the differential diagnosis in patients with cystic masses in head and neck region.

REFERENCES

1. Yilmaz YE, Ozlugedik S, Titiz A, Ozcan M, Unal A. Primary hydatid cyst of the neck: a case report. *B-ENT* 2007; **3**:83-6.
2. Tekin M, Osma U, Yaldiz M, Topcu I. Preauricular hydatid cyst: an unusual location for echinococcosis. *Eur Arch Otorhinolaryngol* 2004; **261**:87-9. Epub 2003 Jul 11.
3. Sajjad AS, Jalil S, Saleem Y, Suleman BA, Chughtai N. Hydatid cysts at unusual sites: reports of two cases in the neck and breast. *J Pak Med Assoc* 2010; **60**:232-4.
4. Katilmis H, Ozturkcan S, Ozdemir I, Guvenc IA. Ozturan S. Primary hydatid cyst of the neck. *Am J Otolaryngol* 2007; **28**: 205-7.
5. Memon AA, Shah SP, Ghumro AA. Hydatid cyst in a submandibular gland. *J Surg Pak* 2002; **7**:44-5.
6. Pampori R, Qazi S, Wani A, Latoo M, Hakeem A. Hydatid cyst in the neck: an unusual presentation. *JK-Practitioner* 2003; **10**: 130-1.
7. Darabi M, Varedi P, Mohebi AR, Mahmoodi S, Nabavizadeh SA, Erfan A, *et al.* Hydatid cyst of the parotid gland. *Oral Maxillofac Surg* 2009; **13**:33-5.
8. Avcu S, Unal O, Kotan C, Ozturk M, Ozen O. Submandibular and thyroid gland involvement of hydatid cysts: a very rare association with percutaneous treatment. *Diagn Interv Radiol* 2010; **16**:251-4. Epub 2009 Oct 9.
9. Hilmani S, Bertal A, Sami A, Ouboukhlik A, El-Kamar A, El-Azhari A. Primary hydatid cyst of the neck. *J Neuroradiol* 2008; **35**:188-9. Epub 2008 Feb 21.
10. Iynen I, Sogut O, Guldur ME, Kose R, Kaya H, Bozkus F. Primary hydatid cyst: an unusual cause of a mass in the supraclavicular region of the neck. *J Clin Med Res* 2011; **3**: 52-4.