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Case Report

Six senses while considering hydatid cyst as a differential for a swelling at nape of the neck: a case report

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ABSTRACT

While cervical swellings usually are located in anterior midline like thyroglossal cyst, thyroid swellings, or in antero-lateral aspect of neck like cold abscess, branchial cyst, lymphangioma, cervical lymphadenopathy etc. Nape of the neck swelling is even less common with differentials including lipoma, sebaceous cyst, lymphangioma, etc. Hydatid cyst (HC) is often missed as a differential resulting in intraoperative surprises. This case report might change the mind of the readers to keep HC in back of their minds while approaching a case of swelling of the neck. Here we report a case of 15 years' female who presented with swelling of nape of neck which on evaluation was inclining towards lipoma/epidermal cyst. With an intention for surgical exploration and excision, the patient was taken for operation, where we discovered it to be HC and the same was later confirmed by histopathology as well. Because of its rare presentation the primary diagnosis of HC is often missed out in spite of having sensitive cytology and imaging modalities. Hence, by reporting this case we intend to emphasize six facts a clinician, a radiologist and also a pathologist must consider while keeping primary HC at an unusual site as a differential diagnosis.

Keywords: Hydatid cyst, Echinococcosis, Soft tissue swelling, Fine needle aspiration cytology

INTRODUCTION

Hydatid cyst (HC) is known by many names such as hydatosis or echinococcosis, which occurs in four different forms: cystic echinococcosis caused by *Echinococcus granulosus*; alveolar echinococcosis caused by *E. multilocularis*; polycystic echinococcosis caused by *E. vogeli* and unicystic echinococcosis caused by *E. oligarthrus*. Of all these, the unilocular cystic form caused by *E. granulosus* is far more common. The definitive hosts are dogs, wolves and foxes, while intermediate hosts are sheep, cattle, and horses. Humans are accidental intermediate hosts therefore do not play any role in the biological transmission. Humans are occasionally infected by handling dogs as well as by oral ingestion of *Echinococcus* eggs through contaminated food or water which hatch in the small intestine and pass into the portal venous system or lymphatic system to reach the liver and lungs.[1]

Sometimes, they can cross the hepatic sinusoids or pulmonary capillary barrier to enter systemic circulation and can affect any body parts. Also, few postulate that the larvae may enter the lymphatics of the intestinal wall and bypass the liver through the cisterna chyli to enter systemic circulation. Although the most commonly involved organ in human is liver (65-75%) and lungs (15-25%) but rarely 5-10% cases can involve any organ of the body including heart, bone, muscles, soft tissue, spleen, kidney, brain, eye etc.[2] Multi-organ involvement is seen in 20-30% of the cases. Though secondary lesions at an intra-abdominal-extra hepatic site are found in many cases, there are also reports of primary involvement of peritoneum, omentum and mesentery of bowel.[3]

Primary involvement of extra abdominal site has been reported in many case reports worldwide, but exact incidence is yet to be estimated. Involvement of head and neck region by HC is very rare, and only few cases have been reported till date in literature.[4]

CASE REPORT

A 15 years female presented with a swelling over nape of the neck of insidious onset, gradually progressive since last 1 year and was associated with mild pain/discomfort on movement of neck. It was not associated with fever, cough, chest pain, loss of weight or appetite.

On local examination a globular swelling of size 4×4 cm present at the nape of neck, soft in consistency, non-tender, non mobile, not fixed to overlying skin with no signs of inflammation or spasm of cervical muscles cervical lymph nodes were normal.

While blood investigations were within normal limits. MRI of cervical spine revealed a round well-defined cystic lesion of 3×3 cm in the left posterior paraspinal muscle at the level of C2 and C3 without any extension to spinal cord. Radiologist mentioned lymphatic cyst and benign epithelial cyst as differential. FNAC report presented by the patient had an impression of a benign lipomatous lesion with hemorrhagic aspirate. In light of all these, we proceeded for PAC with intentions for exploration and excision under GA.

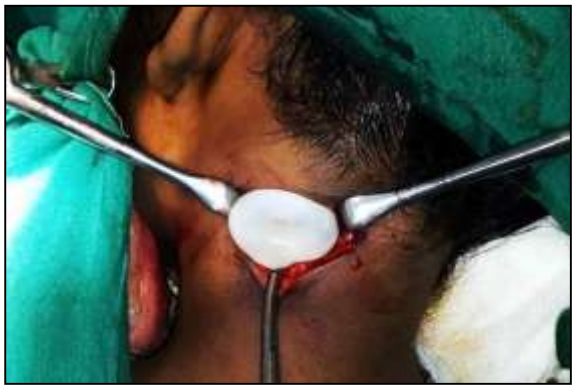

Figure 1: Intraoperative finding of 3×3 cm HC present over nape of neck.

On exploration, operative finding revealed single cystic swelling of 3×3 cm at nape of neck, deep to trapezius with features suspicious of HC. Pericystectomy was done and specimen was sent for histopathological examination which confirmed the diagnosis of HC. Meanwhile we advised the patient for ultrasound abdomen to look out if this was a primary site or secondary to liver involvement. Lung involvement was ruled out by normal chest X-ray (CXR) done preoperatively for PAC workup. But both the investigations were unremarkable which is quite rare, making this nape of the neck lesion primary one. As we were not thinking of HC as a differential and blood tests as well as CXR were unremarkable, we did not subject patient to serological test for *Echinococcus* pre-op.

In postoperative period there were no signs of anaphylaxis and patient was doing well. Hence, was discharged on POD 2 with prescription of Albendazole 400 mg twice day for 4 weeks. The patient was followed up for 6 months and remained free of symptoms.

DISCUSSION

As evident from our case report and few others, the diagnosis of HC should be considered not necessarily among the top differentials still must be kept in the list as a rare possibility while assessing cystic swellings at any anatomical location in an endemic area in all age groups.[2]

Primary involvement of extra abdominal sites though extremely rare is not impossible as interesting reports of primary involvement of neck, supraclavicular, preauricular, pterygopalatine fossa, infratemporal fossa, eye etc are popping up worldwide.[3-5]

The point of reporting our case is to bring in light the following 6 facts in knowledge of every clinician and radiologist when they are anticipating primary Hydatid disease at rare locations as a differential:

Patients remain asymptomatic for very long. Depending upon size and location, swelling or pressure effect might be the only reason for presentation.[4] Clinically small swelling of neck may mislead the clinicians into common differentials like lipoma.[5]

Though we anticipate marked eosinophilia to aid our preoperative diagnosis.[6] However, no increase in eosinophil count has been reported in more than 90% of the past cases as well as in our case.[7]

Imaging modalities like chest X-ray, abdominal ultrasound, CECT abdomen/thorax, MRI etc has high sensitivity in detecting HD in liver/lungs. But primary at unusual site very closely mimic other, rather more common cystic entities can be missed as in our case because imaging modalities depend on performance variability of the radiologist, their familiarity with pathogenesis, awareness of radiological features and consideration of differentials by radiologist.[8]

Though fine needle aspiration has been associated with risk of anaphylactic reaction and increased recurrence rate due to cyst rupture. It could either be carried out as in our patient due to a low index of suspicion or carried out routinely as suggested by certain literature to aid the diagnosis.[9] In our case, patient already presented to us with the report mentioning a benign lipomatous lesion with hemorrhagic aspirate, which in fact was misleading can be attributed to faulty sampling and therefore can be improved by use of ultrasound guided FNA and proper training of technician.[10]

ELISA, Casoni skin test, latex agglutination, immune electrophoresis and direct hemagglutination are serological methods, used for diagnosis of HD. Among these, Ig G2 and G4 ELISA are considered good markers.[11] But immunodiagnostic tests particularly for unusual locations are associated with very high false negativity.[12] Hence, negative test cannot rule out possibility of primary of unusual site.

As in other similar case reports, diagnosis of a HC in present case was not considered until intraoperatively evident, and definitive diagnosis was made only by post-op histopathology which in fact is gold standard. During surgical removal of cysts great care must be taken to avoid spilling of cystic contents which can result in recurrence and anaphylaxis. Patient follow up seems critical in all cases in order to offer accurate diagnosis and definitive treatment and prevent recurrence.[13]

Besides surgery, non-conventional treatment like puncture aspiration injection and re-aspiration (PAIR) had been studied recently and was found safe and effective. In medical treatment, the imidazole group of drugs (mebendazole and albendazole) is widely used but is contraindicated in pregnancy, and in hepatic and renal impairment.[14] Surgery with adjuvant therapy (peri-and postoperative antiparasitic medical therapy such as albendazole) seems to remain the optimal method of treatment.[15] The recommended dose of albendazole is 400 mg orally twice a day for 1-5 months.

CONCLUSION

Because of its rare presentation the primary diagnosis of HC is often missed out in spite of having sensitive cytology and imaging modalities. Hence, by reporting this case we intend to emphasize that HC should also be kept as a differential diagnosis of neck swelling, particularly in countries like India where echinococcus is endemic. Cytology and imaging modalities though very useful are not flawless and can be misleading if we are not flexible enough while considering our differentials as a clinician or a radiologist. If at all suspicious better to take the help of radiology, cytology combined with serology (Ig G ELISA) to reach to a preoperative diagnosis, then to surprise yourselves intraoperatively and to reduce risk of recurrence and even anaphylaxis.

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