

## 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 anterolateral aspect of neck like cold abscess, branchial cyst, lymphangioma, cervical lymphadenopathy. Nape of the neck swelling is even less common with differentials including lipoma, sebaceous cyst, lymphangioma. 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 reported 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 patient was taken for operation, where we discovered it to be HC which was later confirmed by histopathology as well. Because of its rare presentation the primary diagnosis of HC was 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

HC is known by many names such as hydatoris 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.<sup>1</sup>

Sometimes, they can cross the hepatic sinusoids or pulmonary capillary barrier to enter the systemic circulation and can affect any body parts. Although the most commonly involved organ in human are 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.<sup>2</sup>

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.<sup>3</sup>

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

## CASE REPORT

A 15 years female presented with a swelling over the 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.



**Figure1: Intraoperative finding of 3×3cm HC present over nape of neck.**

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.

On exploration, operative finding revealed single cystic swelling of 3×3cm at nape of neck, deep to trapezius with features suspicious of hydatid cyst. 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 was quite rare,

making this nape of the neck lesion primary one. As we were not thinking of hydatid cyst as a differential and blood tests as well as CXR were unremarkable, we did not subject the patient to serological test for echinococcus preoperatively.

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 a 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.<sup>2</sup>

Primary involvement of extra abdominal sites though extremely rare was not impossible as interesting reports of primary involvement of neck, supraclavicular, preauricular, pterygopalatine fossa, infratemporal fossa, eye was popping up worldwide.<sup>3-5</sup>

The point of reporting our case was to bring in light the following 6 facts in knowledge of every clinicians and radiologists when they are anticipating primary hydatid disease at rare locations as a differential: (1) patients remains asymptomatic for very long, depending upon size and location, swelling or pressure effect might be the only reason for presentation<sup>4</sup> Clinically small swelling of neck may mislead the clinicians into common differentials like lipoma;<sup>5</sup> (2) though we anticipated marked eosinophilia to aid our preoperative diagnosis.<sup>6</sup> However, no increase in eosinophil count has been reported in more than 90% of the past cases as well as in our case;<sup>7</sup> (3) imaging modalities like CXR, abdominal ultrasound, CECT abdomen/thorax, MRI had 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;<sup>8</sup> (4) 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.<sup>9</sup> 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;<sup>10</sup> (5) ELISA, Casoni skin test, latex agglutination, immune electrophoresis and direct

hemagglutination are serological methods used for the diagnosis of HD. Among these, Ig G2 and G4 ELISA were considered good markers.<sup>11</sup> But immunodiagnostic tests particularly for unusual locations are associated with very high false negativity.<sup>12</sup> Hence, negative test cannot rule out a possibility of primary of unusual site; (6) As in other similar case reports, the diagnosis of a HC in the present case was not considered until intraoperatively evident and a definitive diagnosis was made only by postoperative histopathology which in fact is gold standard. During surgical removal of cysts great care must be taken to avoid spilling of the cystic contents which can result in recurrence and anaphylaxis. Patient follow up seemed critical in all cases in order to offer accurate diagnosis and definitive treatment and prevent recurrence.<sup>13</sup>

Besides surgery, non-conventional treatment like puncture aspiration injection and re-aspiration (PAIR) has been studied recently and was found safe and effective. In medical treatment, the imidazole group of drugs (mebendazole and albendazole) was widely used but was contraindicated in pregnancy and in hepatic and renal impairment.<sup>14</sup> Surgery with adjuvant therapy (peri- and postoperative antiparasitic medical therapy such as albendazole) seemed to remain the optimal method of treatment.<sup>15</sup> The recommended dose of albendazole was 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 hydatid cyst 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 (IgG ELISA) to reach to a preoperative diagnosis, than to surprise yourselves intraoperatively and to reduce risk of recurrence and even anaphylaxis.

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