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CASE STUDY

SUBMENTAL CYSTICERCOSIS: AN UNCOMMON SITE FOR A COMMON INFECTION

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ABSTRACT

Cysticercosis is a condition which occurs when humans are infested by larvae of *Taenia solium.* It is a cause of increasing health burden in developing countries. Solitary extraneural cysticercosis may mimic soft tissue lesion; hence it is important to consider cysticercosis as a differential diagnosis during evaluation of such lesions. Here we present a case of submental cysticercosis in a young male child along with a brief review of literature.

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INTRODUCTION

Cysticercosis is a parasitic infection caused by the ingestion of eggs of the *Taenia solium* parasite through contaminated food or ingestion of infected pork. Most common site of infestation is brain, termed as neurocysticercosis. Other less common sites of cysticercosis is muscles, eyes, lungs and liver. It can especially present as a diagnostic challenge in a paediatric patient where lymphadenopathy or congenital swellings like branchial cyst or thyroglossal cyst are likely diagnosis of a neck swelling. Here we present an uncommon case of cysticercosis presenting as a sub mental swelling in an 8-year-old male child.

CASE REPORT: An 8-year-old male child presented to the hospital with complaints of submental swelling and off & on fever. He had mild cough without sputum production. No other significant history was present. There was no history of tuberculosis or any other infection in the past.On general examination, a small swelling measuring approximately 1x1 cmwas noted in the submental region (Figure 1a). The swelling was mobile, soft and non-tender. No other palpable swelling or lymph node was noted. Complete blood count revealed mild eosinophilia; however, rest of the parameters were within normal limits. Biochemical investigations were also well within the range of normal reference values. Chest X-ray did not reveal any abnormality. Ultrasound of the neck showed a small oval hypoechoic lesion measuring 9.2 X 7.6 mm with internal echoes and fine septa over sub mental region in subcutaneous plane; suggestive of a cystic lesion(Figure 1b). Fine needle aspiration (FNA) was done from the submental swelling and yielded fluidy aspirate. Smears prepared were air dried and fixed in 95% ethyl alcohol and subsequently stained with Giemsa and Papanicolaou stain; respectively. Smears examined showed mixed inflammatory cells consisting of neutrophils, eosinophils, lymphocytes along with proteinaceous background (Figure 2a).

After a careful screening, an occasional large fragment of fibrillary material with interspersed small nuclei could be demonstrated, which was suggestive ofbladder wall of parasite, i.e. cysticercus (Figure 2b-d). A final diagnosis of submental cysticercosis was made. The patient was started on albendazole 400mg once daily for 21 days. After completion of antihelminthic therapy, the swelling significantly decreased in size. The patient is currently doing well.

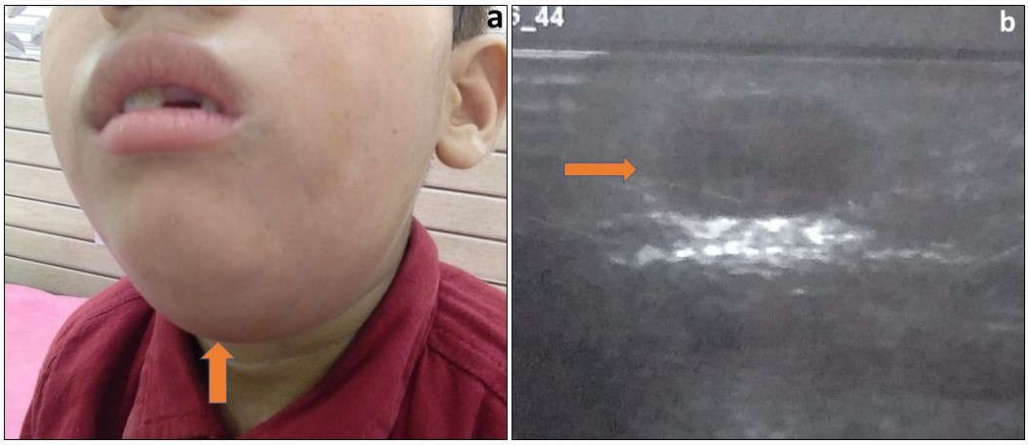

Figure 1a-b. a- Submental swelling measuring 1.5x1.5 cms; b- USG neck showing a small oval hypoechoic lesion with internal echoes&fine septa over submental region in subcutaneous plane

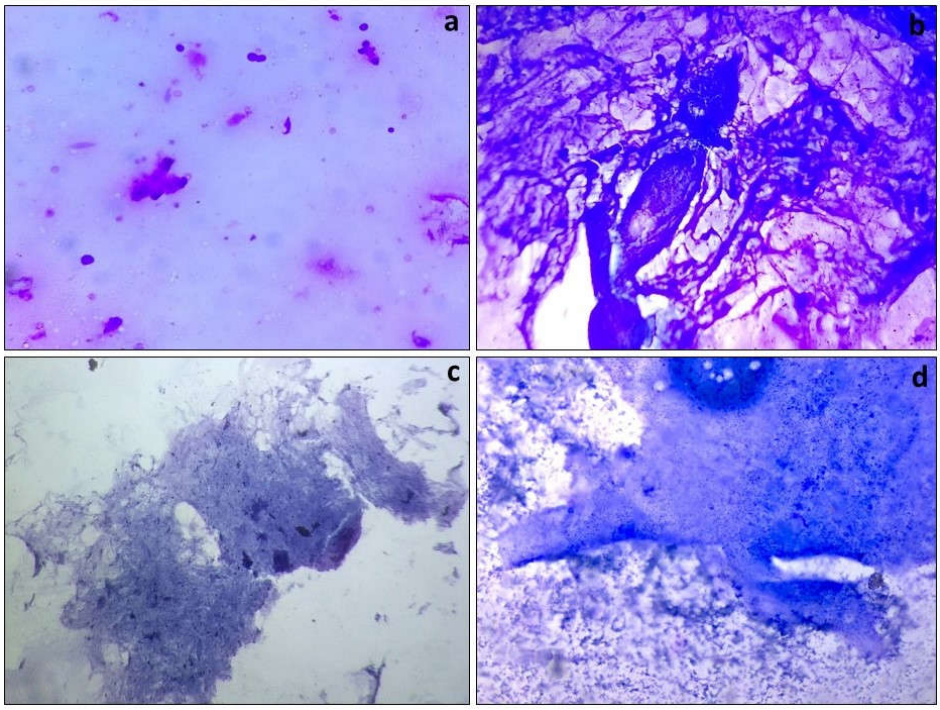

Figure 2a-d. a- Smear showing mixed inflammatory cells in a proteinaceous background; bd: Large fragments of fibrillary material with interspersed small nuclei; suggestive of bladder wall of *T. solium*(b- Giemsa, 200X; c-d- Pap, 200X, 400X)

DISCUSSION

Cysticercosis in children is relatively rare as compared to the adults (Allan *et al*., 1991; Chawla *et al*., 2004; Uledi, 2010). The most common site involved is the central nervous system whereas the second most common site is skeletal muscle. Symptoms are secondary to mass effects or an allergic response. A small number of cases of isolated cysticercosis in neck are reported however all were reported either in adults or were travel or diet related (Chawla *et al*., 2004; Uledi, 2010; Gupta, 2010; Elhence *et al*., 2012). In our case the patient was a child with no such history. Normally, humans are the definitive hosts for Taenia solium, the life cycle of which begins with ingestion of viable larvae in inadequately cooked pork. Ingested eggs hatch in the small intestine, releasing oncospheres that penetrate the bowel mucosa and enter the bloodstream to travel to various tissues where they develop to form an encysted larval form of *T. solium* known as cysticercosis cellulosae (Uledi, 2010; Uledi, 2010; Gupta, 2010; Elhence *et al*., 2012; Chakravarti, 2014; Meher, 2006; Jain *et al*., 2012).

When the larva dies, it induces an aggressive granulomatous inflammatory response, leading to the characteristic organ-specific symptoms. Only a few cases of cysticercosis have been reported in neck region (Gupta, 2010; Elhence *et al*., 2012; Chakravarti, 2014; Meher, 2006; Jain *et al*., 2012; Virk, 2009; Mittal *et al*., 2009). Every case of cysticercosis should be investigated for the presence of other lesions on other sites. Serology can be done to detect the cysticercal antibodies in the serum or cerebrospinal fluid. Biopsy from the lesion remains the gold-standard in providing a definitive diagnosis. Histopathology confirms the diagnosis by showing scolices, hooklets and cyst wall (Jain *et al*., 2012; Virk, 2009; Mittal *et al*., 2009; Tanechpongtamb, 2005). In the head and neck site, other than neurocysticercosis, the sites commonly involved are eyes, buccal mucosa, tongue and lips. However, cysticercosis of the neck has also been reported in the muscles such as mylohyoid, masseter, sternocleidomastoid and omohyoid in a handful of case reports (Virk, 2009; Mittal *et al*., 2009; Tanechpongtamb, 2005). Clinical diagnosis of isolated subcutaneous neck swelling especially in a child as cysticercosis is extremely challenging.

The usual differential diagnosis in such a case is inflammatory lymphadenopathy, cystic hygroma, branchial cyst, thyroglossal duct cyst, lymphangioma, epidermoid cysts, bronchogenic cysts, ranula or arterio-venous malformations (Jain *et al*., 2012; Virk, 2009; Mittal *et al*., 2009; Tanechpongtamb, 2005). Ultrasonography may help in diagnosis by showing presence of opacities in hypoechoic cystic lesions; however, it is not confirmatory (Chakravarti , 2014; Meher *et al*., 2006; Jain *et al*., 2012; Virk, 2009; Mittal *et al*., 2009; Tanechpongtamb, 2005). FNAC generally confirms the diagnosis by demonstrating the parasitic wall but in a case like ours with significant inflammation on smears, a careful search for the same is warranted. Available treatment options for cysticercosis depending on the extent of involvement, location & size, include anti-parasitic therapy, anti-seizure therapy, and surgery (Virk, 2009; Mittal *et al*., 2009; Tanechpongtamb, 2005). All the members of the family should also be screened. Awareness programmes should be given to the public regarding importance of basic personal hygiene.

CONCLUSION

To conclude, the possibility of an isolated submental cysticercosis should be considered in evaluation of soft tissue lesion in neck region. Not only can cysticercosis be seen at unusual sites but also in patients who have no history suggestive of dietary ingestion of pork. Cytological examination plays a key diagnostic role in cysticercosis at unusual locations.

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