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Case Report

The Mandibular Angle Hydatid Cyst Mimicking Branchial Cleft Cyst: A Case Report

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Abstract

We report an unusual case of primary hydatid cyst of the mandibular angle without glands involvement, in the left supraclavicular region of the neck with no involvement of any other regions of the body. In July 2012, a 25-yr old woman, from Golestan Province, Northeast Iran was admitted to our ENT Clinic, with one-year history of a progressively increasing swelling, pain and gradually growing mass located in the left side of neck region. The patient was diagnosed by Fine Needle Aspiration Cytology (FANC) and histopathology examination. Hydatid cyst should be considered in differential diagnosis of soft tissue mass such as branchial cleft cyst (BCC) and or dermoid cyst in the cervical region especially in endemic areas. Moreover, FANC could be recommended as a valuable, rapid, simple, and safe procedure to diagnose hydatid cyst especially in unusual locations.

Introduction

Cystic hydatid disease (CHD), also known as hydatidosis, is a zoonotic parasitic infection caused by *Echinococcus granulosus* or dog tapeworm. Humans are accidently infected by oral ingestion of food or water contaminated with dog feces containing *Echinococcus* eggs (1). CHD is a helminthic disease with global distribution, especially in the Middle East including Iran (1-2).

In CHD, the liver (60%-70%), and lungs (15-25%), are the most common infected organs, but any organ of the body can be involved? The rates of the localization of hydatid cyst (HC) in different body organs vary in the literature (3). Even in region where hydatidosis is endemic such as Iran, cervical region and/or neck hydatidosis is rare and its incidence is unknown (2). Only a few cases of HC located in submandibular glands have been reported in literature (4, 5).

Diagnosis of HC in cervical region is difficult due to its rarity. Ultrasonography (USG), computed tomography (CT) scan, X-Ray graphy, Fine Needle Aspiration Cytology (FNAC) and biopsy (FNAB), magnetic resonance imaging (MRI) are valuable in identifying calcifications and the presence of daughter cysts. However, definite diagnosis should be confirmed by microscopic examination for hydatid sands (also known as protoscolices) supported by histopathology (2, 3, 6).

Herein, we report, an unusual case of primary HC located in the mandibular angle mimicking branchial cleft cyst (BCC).

Case Report

In July 2012, a 25-yr-old woman was admitted to ENT (ears, nose, and throat) clinic, Bandar-Torkman district, Golestan Province, Northeastern Iran, with one-year history of a progressively increasing swelling, pain and gradually growing mass located in the left side of neck region. She had no past medical history of hydatidosis. Physical examination showed a soft mass that was tender on palpation somewhat painful and moderately hard in the upper anterio-lateral surface left side of her neck. No other clinical sign was noticed present. Laboratory findings including sedimentation rate, and complete blood count (CBC) were normal and showed no eosinophilia. Ultrasound showed a unilocular cystic lesion, mimicking a branchial cleft cyst (BCC), 40× 26 ×24 mm in size, with about13 ml fluid in the anterior margin of sterneocleidomastoid muscle (SCM) and posterior to the submandibular gland. CT scan of the cervical region revealed a cystic lesion with thin borders and wall. The clinician recommended fine needle aspiration cytology (FNAC).

FNAC was inducted under aseptic conditions using a 22-gauge needle and about 2 ml of clear fluid was aspirated. The aspirated materials were smeared on the slides, also these were centrifuged, and then sediment was examined under light microscope. The microscopic examination revealed small number of protoscolices with numerous hooklets (Fig. 1).

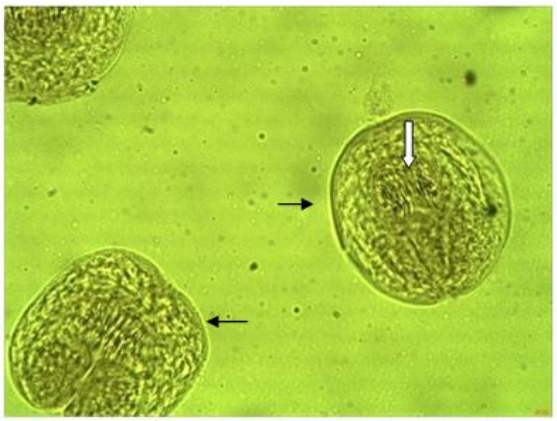

Fig. 1: Cytology direct smears revealed a few protoscolices (black arrow) and hooklets (white arrow) (40x)

Consequently, early diagnosis of hydatid cyst is confirmed by FNAC. The patient was examined for more works up and rule out of hydatidosis in other organs of the whole body. Chest radiograph was normal. Ultrasonography and CT scan showed no involvement of the liver, lungs, submandibular glands, and any other organs. Then, the mass was removed through surgery. The received tissue grossly showed white gelatinous membranous tissue 4×4 cm in size and 0.2 cm in thickness. Another fibroconnective tissue 0.5 × 0.5 ×0.5 cm in size received with the first one. Microscopic examination of the tissue revealed germinal, acellular laminated layers and a few pro-

toscolices (Fig. 2). In addition, separate fibroconective tissue showed chronic inflammatory reaction.

The patient was given albendazole (400 mg twice daily), 4 days prior to the surgery together with for 4 weeks post-operation. Throughout 20 months follow-up, there was no evidence of recurrence and hydatidosis in other locations of the body.

All examinations and works up were performed after the patient gave oral consent. All included the pictures in this report were taken in our research lab at Sari School of Medicine, Mazandaran University of Medical Sciences.

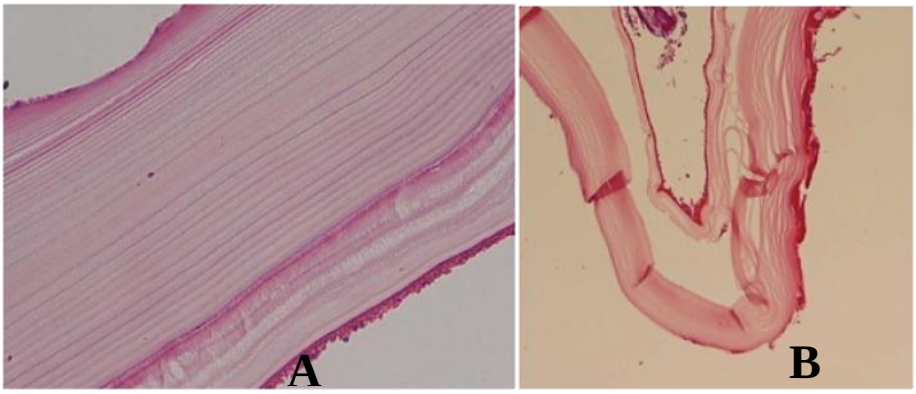

Fig. 2: Histopathology picture showing laminated layer (Hematoxylin and Eosin A, 40× – B, 10×)

Discussion

The primary HC occurrence in the neck is relatively uncommon and involvements of the submandibular glands are extremely rare. So far, a few case of hydatidosis in cervical region such as thyroid, parotid and submandibular glands have been reported from Iran and other parts of the world (2). However, mandibular angle involvement has not been previously reported. In the present report, the patient suffered from primary mandibular angel hydatidosis with no involvement of any other regions including submandibular glands. Thus, to our knowledge, this might be the first report of mandibular angle hydatid cyst from Iran and possibly the world.

The mandibular anglehydatidosis, caused due to systemic diffusion through lymphatic route, is a strong possibility in case of unusual presentation locations. The cyst might remain asymptomatic for a long period, presenting a slow development rate (7).

Unusual locations of HC have been reported around the world including the ureter, brain, uterus, heart, bones, kidney, spleen, cranium, and muscles, but soft tissue hydatid disease represents less than about 3% of all hydatid disease. Although it can involve many body organs, involvement of the submandibular region is very rare (2, 8).

The diagnosis of CHD mostly depends on the clinical history, serologic tests, and diagnostic imaging though not all these techniques are definitive. However, for the evaluation of mass lesions in the cervical region, FNAC and or FNAB, as gold standard, are very valuable and reliable procedures in the differential diagnosis of CHD (8, 9). FNA findings of HC have been well defined. FNA can be a safe, fast, easy diagnostic method in the evaluation of suspected HC with no complications (9, 10). However, its application generally is recommended and requires to sufficient experience, due to the potential risk of anaphylactic reaction and/or dissemination.

In our case, the patient had not any complications following FNAC. Thus, FANC could be suggested as a valuable, rapid, simple, and

safe procedure to diagnose hydatid cyst especially in unusual locations.

HC may involve mandibular angle without glands involvement. Given that throughout ultrasound, the mandibular angle hydatid cyst presents with a cystic mass, mimicking a branchial cleft cyst (BCC) and or dermoid cyst (11).

Conclusion

HC must be considered in differential diagnosis of soft tissue mass in the cervical region especially in endemic countries such as Iran. Therefore, it should be managed through surgery to prevent diffusing cyst to other regions of the body and anaphylactic reaction.

Acknowledgements

The authors declare that they have no conflict of interest.

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