

Mass of the neck: an extremely rare location of hydatid disease

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SUMMARY: Mass of the neck: an extremely rare location of hydatid disease.

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Hydatid disease (HD) is a parasitic zoonosis which was first described by Hippocrates in the ancient years. Liver and lungs are the most commonly affected organs, while neck involvement is extremely rare. We report a case of a 75-year old male, who presented with an

isolated right-side cervical mass. After clinical examination and screening tests, HD was diagnosed. Pre-operative and post-operative treatment with albendazole was administered and en-block surgical excision of the mass was performed. Histopathological examination of the specimen confirmed the diagnosis of HD. At one-year follow-up, the patient remained asymptomatic without any signs of recurrence. Although HD of the neck is an extremely rare entity, physicians should always include it in the differential diagnosis of neck masses.

KEY WORDS: Hydatid disease - Echinococcus.

Introduction

HD is one of the commonest zoonoses. It is caused by the larvae of dog and fox tapeworms (cestodes) of the genus *Echinococcus* (family Taeniidae). Six species of *Echinococcus* have been recognized, but the most common members of the genus are *Echinococcus granulosus* which causes cystic echinococcosis (CE) and *Echinococcus multilocularis*, which causes alveolar echinococcosis (1).

Humans may be affected accidentally. Most commonly affected organs are liver and lungs, although any organ can be involved. However, occurrence of Hydatid cyst (HC) in the head and neck region, remains an extremely rare manifestation (2).

We present a case of a 75-year old male, with a follow-up of a whole year, who presented with an isolated HC of the neck.

Case report

A 75-year old male, with no reported domestic animals, presented to our department complaining of a mass in the right side of the neck, starting about 3 cm below the angle of mandible, and which as mentioned has been gradually enlarged over a period of 2 years. In clinical examination, the man was in good general health, having a prominent right-side neck mass of 4 cm x 6 cm (Figure 1). This mass was well defined, mobile and firm in consistency. Patient never complained about pain and there were no signs of inflammation on the overlying skin. The ear, nose and throat clinical examination were normal. The abdomen was soft and the chest roentgenogram was normal. Complete blood count was normal, except from slight eosinophilia.

Computer-assisted tomography (CAT) scan of the cervical region was performed, in order to assess features and exact location of the lesion. Results showed a cyst located in the subcutaneous tissue of the neck, being compatible radiographically with hydatid cyst (Figure 2). Though fine-needle aspiration (FNA) was proposed, it was not performed to avoid local disperse.

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Figure 1 - Mass of the right side of the neck.



Figure 2 - Axial post contrast CAT scan of the neck.

Furthermore, CAT scan of the abdomen and thorax was decided, which excluded hydatid disease in other organs, diagnosing the patient with an isolated hydatid cyst of the neck.

Surgical excision of the cyst was planned. The patient had a preoperative treatment with albendazole 400mg twice a day for 2 weeks prior to surgery. Subsequently, the patient was operated under general anesthesia and a complete excision of the cyst was performed (Figures 3, 4). The surgical wound was irrigated with hypertonic saline and povidone-iodine.

Histopathological examination of the specimen confirmed the diagnosis of hydatid cyst, by showing a single grey white cystic mass with multiple layers measuring 4,4 cm x 6,5 cm x 2 cm. The cyst was multilocular. Patient was discharged after a total of 2 days and his hospital course was uneventful. He was additionally given a post-operative treatment with albendazole 400 mg twice a day for 4 weeks.

After a one-year follow-up, there was no evidence of recurrence and the patient was free from any visceral or soft tissue hydatidosis.

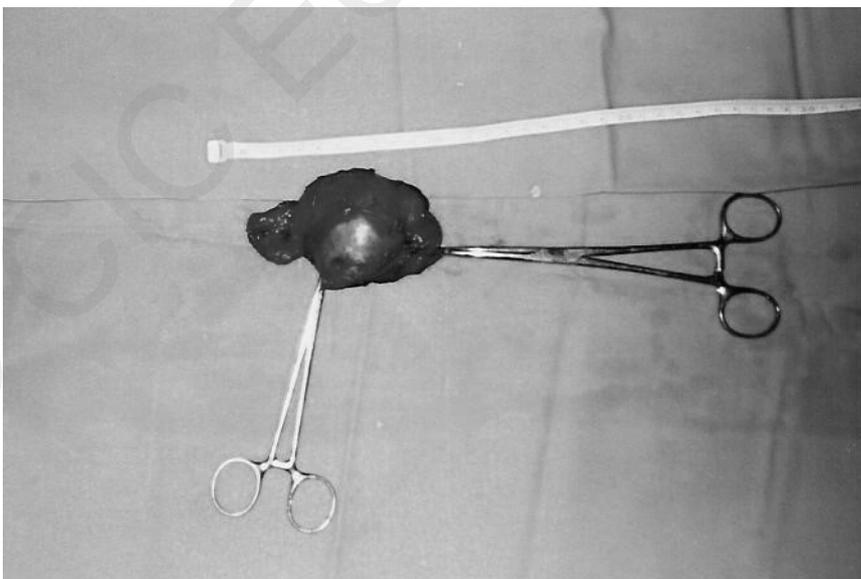


Figure 3 - Macroscopic appearance of hydatid cyst after complete surgical excision.

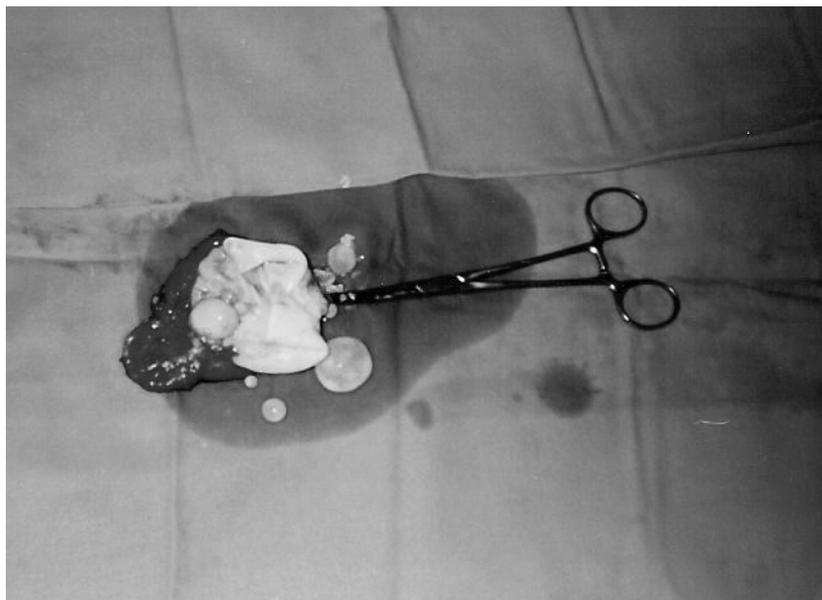


Figure 4 - Hydatid cyst after complete surgical excision.

Discussion

CE or HD is a parasitic zoonosis caused by the larval stage of cestodes *E. granulosus*. The definitive host of this parasite tapeworm is the dog and humans are one of the intermediate hosts. *E. granulosus* has life span of 5 to 20 months. The tapeworm's length is 5 mm. *Echinococcus* grows up in the intestine of the definitive host and releases eggs which are excreted with the feces and are ingested by intermediate hosts, like humans (3).

HD is endemic in the Middle East Africa, South America, New Zealand, Australia, Turkey and Southern Europe. Infestation by *Echinococcus granulosus* in humans most commonly occurs in the liver (55-70%) followed by the lung (18-35%). In about 5-13% of cases there can be simultaneous affection of both organs. Incidence of unusual sites is about 8-10%. Symptomatic cysts have been reported occasionally in the spleen, kidney, peritoneal cavity, skin and muscles. Rarely the cyst is located in the heart, brain, vertebral column, neck or thyroid gland (4).

The cyst of *E. granulosus* may take many years to produce clinical symptoms or it could remain asymptomatic. Patients of any ages may be affected. Most often, liver and lungs are affected, with both those organs amounting for 90% of all cases. Symptoms depend on mass effect or complications of loss of cyst integrity. Symptoms therefore depend on locality and

size of cysts, speed of growth and complications of rupture. They vary from vague pain to major rupture that can lead to severe anaphylactic reaction (5).

Many affected patients have a single cyst. However, the presence of an isolated hydatid cyst of the neck is extremely rare (4). Head and neck hydatid cyst can be observed at all ages. The clinical features are usually limited to incidental discovery of a slow-growing, painless, non-inflammatory swelling. In some cases, the swelling is so large that can be responsible for compression of adjacent organs.

HD in the head and neck region is rare and only a few cases have been reported in the literature (6). Rarity of the disease in this region presents a diagnostic problem for physicians. HC is rarely considered in the differential diagnosis of cystic lesions in the head and neck, especially in non-endemic areas in absence of HD elsewhere in the body.

Diagnosis of HD mainly depends on the personal medical history, radiological imaging and serological tests. Imaging techniques like ultrasound, CAT scan and magnetic resonance imaging (MRI) remain more sensitive than serum diagnosis (7).

Tests like hemagglutination, latex agglutination, skin test (Casoni intradermal test), Enzyme-linked Immunosorbent Assay (ELISA), and western-blot are widely used especially in abdominal disease, but they have low diagnostic sensitivity and specificity in extra hepatic hydatidosis. Nevertheless, these tests have a

role in the follow-up of treated patients (8).

The role of fine needle aspiration cytology (FNAC) in HC diagnosis was considered controversial and usually not preferred due to the possibility of anaphylactic reaction, dissemination of the disease and recurrence as a result of spillage of contents. However, FNAC currently represents the gold standard to confirm the diagnosis of hydatid cyst (9, 10). It is a feasible, safe and high-sensitivity technique that can identify features such as scolax and the lamellar membrane.

Ultrasound, CAT and MRI scans have almost 100% specificity and sensitivity and can reveal the morphological characteristics of the cyst, its exact position in relationship with the surrounding tissues, as well as the exact number of the cysts. Imaging tests can also help to differentiate HC from other lesions such as abscesses or tumors (11).

Medical treatment with anthelmintic drugs, such as mebendazole and albendazole, has been introduced since 1977 as preoperative and/or postoperative chemotherapy. These agents can offer clinical improvement, increased life expectancy, shrinkage of the cyst and sterilization of the cyst fluid, making surgery safer. A short course of chemotherapy after surgery reduces the risk of recurrence (12).

Surgical excision still remains the most effective treatment of HC of the neck. The general principle

for surgical treatment is the complete en-bloc resection of the cyst and utmost care should be taken to prevent rupture of the cyst, as spillage of the content can lead to anaphylactic reaction, recurrence and multiple hydatidosis (13). Packs immersed in hypertonic saline solutions placed in the operative field are widely used in order to prevent spillage of scoleces and daughter cysts (14). Subtotal pericystectomy or resection of the prominent dome should only be performed in case of adhesions to neurovascular structures or the upper aerodigestive tract (15).

Regular immediate and long-term postoperative surveillance is recommended, especially ultrasound and hydatid serology (16).

Conclusion

HD is a common and generally benign condition, which mainly affects the liver and lungs, but can possibly develop in any site of the human body. Although HC rarely occurs in the head and neck, it should be kept in the differential diagnosis of any cervical swelling, even in non-endemic areas.

Imaging techniques and high emphasis on avoiding rupture of the cyst during operation can achieve a satisfactory clinical outcome with excellent prognosis.

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