Isolated Cyst Hydatid: An Unusual Cause of A Mass in The Nape of The Neck

Şemsettin Okuyucu MD[1], Ali Balcı MD[2], Sinem Karazincir MD[2], Ertap Akoğlu Assoc. Prof. MD[1], Serhat Gözel MD[1], Şafak Dağlı Prof. MD[1]

[1] Department of Otorhinolaryngology Head and Neck Surgery, Medical Faculty of Mustafa Kemal University, Antakya, TURKEY

[2] Department of Radiology, Medical Faculty of Mustafa Kemal University, Antakya, TURKEY

ABSTRACT

Because of the rarity of hydatid cyst in the head and neck area, it may be overlooked by the clinicians. We report a case of primary hydatid cyst in the posterior cervical region without another organ involvement. A 54-year-old woman presented with a large, painless lump in the nape of the neck.

A computed tomography scan demonstrated a 5x5x4 cm hypodense mass which is located in the left scalen muscles. An axial magnetic resonance image revealed well-defined high signal intensity mass with low signal intensity rim, which is considered as a characteristic feature for hydatid cyst. She underwent surgery to excise the cyst. Histopathological examination confirmed the diagnosis as a hydatid cyst.

If a cystic mass in head and neck area exists, hydatid disease must be kept in mind, especially in endemic areas, to avoid the complications such as acute anaphylaxis or recurrences due to inappropriate management.

• ***Key Words:*** *Cyst hydatid, head, neck, mass.* ***Nobel Med 2009; 5(1): 46-49***

ÖZET

İZOLE KİST HİDATİK: ENSE BÖLGESİNDE KİTLE İÇİN SIRADIŞI BİR NEDEN

Kist hidatik baş boyun bölgesinde nadir görüldüğünden klinisyenlerin gözünden kaçabilir. Bu makalede başka organ tutulumu olmaksızın posterior servikal bölgede yerleşmiş bir kist hidatik olgusu sunulmuştur.

Elli dört yaşındaki bir kadın hasta, boynun ense kısmında büyük, ağrısız kitleyle başvurdu. Bilgisayarlı tomografide sol skalen kasların içinde 5x5x4 cm boyutlarında hipodens kitle görüldü. Aksiyel plan manyetik rezonans görüntülemesinde kist hidatik için tipik olan kenarları düşük sinyal yoğunluğunda çevrelenmiş, iyi sınırlı, yüksek sinyal yoğunluklu kitle saptandı. Cerrahiyle çıkartılan kistin histopatolojik incelemesi kist hidatik tanısını doğruladı. Özellikle endemik bölgelerede baş boyun bölgesinde kistik kitle varlığında, akut anafilaksi ve uygun olmayan tedavilere bağlı nükslerin engellenmesi için kist hidatik hastalığı akılda tutulmalıdır.

• **Anahtar Kelimeler:** Kist hidatik, baş, boyun, kitle, **Nobel Med 2009; 5(1): 46-49**

INTRODUCTION

Cyclo-zoonotic infection of the larvae form of *Echinococcus granulosus* (canine tapeworm) is called Hydatidosis. The life cycle *E. granulosus* needs two distinct types of mammalian hosts; dogs or other carnivores are definitive hosts, whereas sheep or other ruminants are intermediate hosts.[1] Humans do not participate in the complete life cycle but they are occasionally infected in case of ingestion of food or water that has been contaminated with feces of the infected dog.[2] After digestion of the outer capsule of the egg, the embryo (oncosphere) is released to enter a branch of the portal vein by penetrating through the duodenal mucosa. Most of these embryos become lodged in the hepatic capillaries, where they either die or to grow into hydatid cysts. If the larva passes through the capillary sieve, a systemic spread occurs.[3] Thus hydatid cyst develops most frequently in the liver (65%), the lungs (25%), rarely in the brain, the eye, the heart, the bone or other internal organs.[4]

The occurrence of hydatid cysts in the head and neck is rare even in countries where echinococcus infestation is endemic. It can be either isolated or with another organ involvement. We describe a patient with a primary hydatid cyst located in the posterior cervical region to emphasize the pitfalls in the differential diagnosis and treatment.

CASE REPORT

A 54-year-old female was referred to us with a painless mass in the nape of the neck. The patient had been complaining from this slowly growing swelling for nearly 3 years. She was asked for trauma, weight loss, fever and night sweats but she denied. She underwent several medical therapies including antibiotics, but her complaints were not alleviated. Her otorhinolaryngological past history was unremarkable. She was breeding sheep on her own farm.

Physical examination revealed a 5x5 cm firm, hard, round shaped mass located in the suboccipital area. The overlying skin was normal which there was no redness and was not warm. There was no cervical lymphadenopathy. Laboratory data were within the normal limits.

A Computed Tomography (CT) scan demonstrated a 5x5x4 cm hypodense homogeneous mass located in the scalen muscles on the left side with peripheral enhancement after intravenous administration of contrast agent injection. The lesion compressed the adjacent soft tissues. Adjacent cervical vertebra was normal (Figure 1). There were no septations within

Figure 1. Axial CT scan showing a well circumscribed, cystic lesion occupying the scalen muscles on the left side.

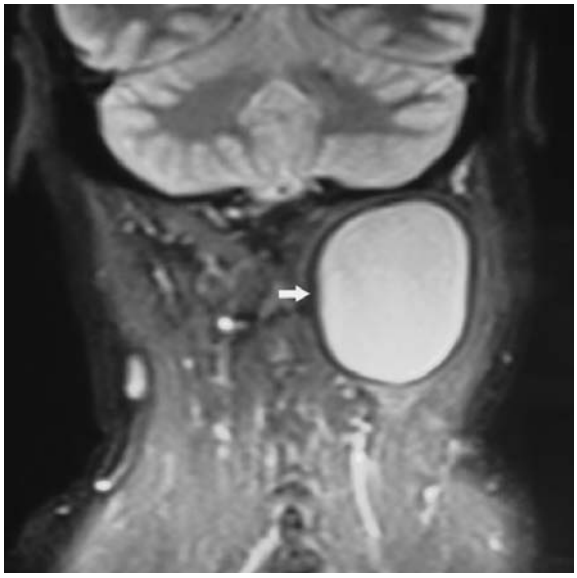

Figure 2. A coronal fat saturated T2-weighted MR image showing well-defined high signal intensity mass with low signal intensity rim (arrow), which is a characteristic feature for hydatid cyst.

the cystic mass. T1 weighted MR images showed a low signal intensity mass lesion. An axial fat saturated T2-weighted MR image through scalen muscles revealed well-defined high signal intensity mass with low signal intensity rim, which is considered as a characteristic feature for hydatid cyst (Figure 2). A chest X-ray and abdominal sonography were performed because the mass was thought to be a hydatid cyst. They both revealed no evidence of extracervical site for hydatid cyst.

Although serologic tests performed did not confirm the suspected diagnosis of hydatid disease, fine needle aspiration biopsy was avoided. Under general anesthesia an exploration of the neck was planned. The mass was located on the medial side of the trapezius muscle →

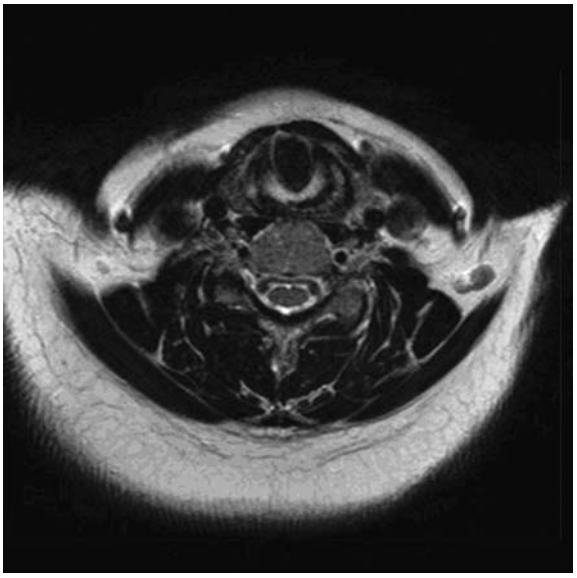

Figure 3. Axial T2 weighted MR image shows no evidence of recurrence after 17 months.

and thought to be cystic on palpation. A meticulous dissection was carried out to prevent any damage to the integrity of the capsule. Although spillage of the cyst contents were avoided, the operation field was irrigated with saline solution after the cyst was totally excised. Pathological examination of the operative specimen confirmed the diagnosis as a hydatid cyst.

Chemotherapy was administered with oral albendazol by 800 mg/day with in two doses for 8 weeks. After the operation the symptoms that disturbed the patient were diminished quickly. On regular follow-up visits, she was free of disease for 17 months (Figure 3). In addition, serologic tests confirmed no recurrences of the hydatid cyst.

DISCUSSION

Hydatid disease is a significant parasitic infection in endemic areas where sheep, dogs and man live in close contact such as Mediterranean countries, the Middle East, South America, New Zealand, Australia, and Southeast Asia; however, contrary to common belief, the incidence has not decreased significantly during the past decade.[5] As an accidental host men and women are affected equally. Echinococcus represents symptoms characteristic of a slow growing benign tumour with a variable period between initial infection and clinical symptoms.[6] The rate of growth of hydatid cysts is highly variable and ranges from 1 to 5 cm a year.[7] The slow-growing hydatid cyst is well tolerated until it causes dysfunction because of its size. The clinical course depends on the site of involvement and the pressure caused by the enlarged cysts.

A hydatid cyst has three layers. The outer layer or pericyst is a rigid protective layer with a few millimeters thickness. It represents the response of the host to the parasite. The middle laminated membrane is white, acellular, 2 mm thick and is easily ruptured. It is a selective permeable membrane that permits the passage of nutrients but is impervious to bacteria. The inner germinal (or germinative) layer is thin and translucent. Scolices, the infectious embryonic tapeworms, develop from an outpouching of the germinal layer and form hydatid sand, which settles in the dependent part of the cyst. The cyst fluid is crystal clear. It is a transudate of serum, contains proteins, and is antigenic. Although cyst rupture may be clinically silent, it can cause eosinophilia or anaphylaxis.[3] A single unilocular cyst is the typically presentation, however, 20-30% of cases may have multiple cysts in the same or multiple organs. Hydatid disease may mimic benign or malignant tumours, cysts, abscess hematomas, pseudocyst and congenital cystic masses including hemangioma, lymphangioma and branchial cleft cysts.[2, 8] Because of the rarity of the disease and lack of experience of clinicians in nonendemic areas, it may be difficult to diagnose a hydatid cyst when it is located in the head and neck area. The diagnosis of Echinococcus infection is based on the history of the patient, physical examination, diagnostic imaging and serologic tests.

Serological methods, used for the diagnosis of hydatid disease are direct hemagglutination, latex agglutination, immunoelectrophoresis, skin tests and ELISA.[8] Although serology is highly sensitive (80-100%) and specific (88-96%) for liver cyst infection, it is less sensitive for lung (50-56%) or other organ involvement (25-56%).[9] Therefore, positive serological results do not confirm the diagnosis of hydatid cyst, and negative results do not exclude it. Serologic tests are more important in the follow-up of treated patients, for whom a drop in titer indicates resolution and a rise is likely to indicate recurrence of the cysts.

Radiological imaging methods such as ultrasonography (US), Computed tomography (CT) and magnetic resonance imaging (MRI) play an important role in the diagnosis.[10, 11] While US helps to differentiate the solid lesion from the cystic lesion, the characteristic feature of hydatid cyst such as the "water-lily sign" could be seen in CT if germinative membrane is detached. For the cervical mass with a cystic component MRI is not only useful in differentiating the cyst from adjacent soft tissues, in showing the different intensity, the septas and the daughter cysts, it also gives excellent radiological data and alerts the surgeon of the need for a vascular procedure.[12] The patients with ecinococcus should undergo a systemic examination →

because the disease may locate in the other parts of the body.[4]

It is not recommended to aspirate the cyst for diagnosis because of the possibility to precipitate acute anaphylaxis or recurrences due to the spillage of daughter cysts.[13] In an opposite manner, in seronegative patients with hepatic image findings compatible with echinococcosis, US guided fine needle biopsy is advocated to confirm the diagnosis.[14]

Although the controversy exists, aspiration of the cyst contents and injection of a hypertonic saline solution followed by reaspiration was noted as a safe and effective treatment for a cyst hydatid located in parotid gland.[15] Surgical removal of the hydatid cyst is suggested as the most effective treatment.[16, 17] The surgeon must be careful to remove the cyst, totally avoiding spilling its contents since fatal anaphylaxis on spilling the contents of the cyst has been reported. For soft tissue disease, surgical strategy varies widely between radical clearance followed by lavage and simple instillation of scolicide.[2] Medical treatment alone is not effective but a combination may be used in the management of patients with recurrence and surgery is not available. Mebendazole or albendazole or praziquantel can be used as a chemotherapeutic agent.[4]

Although the occurrence of hydatid cysts in the head and neck is rare, clinicians have to consider this disease in the differential diagnosis of cervical masses especially in countries where echinococcus infestation is endemic.

C **CORRESPONDING AUTHOR:** Semsettin Okuyucu MD, Dept. of Otorhinolaryngology Head and Neck Surgery, Medical Faculty of Mustafa Kemal University, Antakya/TURKEY semsettinokuyucu@yahoo.com

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