Case Report / Olgu Sunumu

A rare, incidental submandibular hydatid cyst

Nadir bir insidental submandibüler kist hidatik

Murat Doğan, MD.,[1] Halil Polat, MD.,[1] Ömer Ayyıldız, MD.,[1] Mustafa Güreli, MD.[2]

[1]*Department of Otolaryngology, Kayseri Training and Research Hospital, Kayseri, Turkey*
[2]*Department of Pathology, Kayseri Training and Research Hospital, Kayseri, Turkey*

ABSTRACT

Hydatid cyst disease is a parasitic infestation caused by *Echinococcus granulosus*. It is transmitted via oral ingestion of eggs excreted by dog's stool. Liver and lungs are the most commonly involved organs. Primary involvement of submandibular region by hydatid cyst is extremely rare. In this article, we report a case of a lesion excised from submandibular region with an initial diagnosis of branchial cleft cyst, but shown to be a hydatid cyst in histopathological evaluation.

Keywords: Echinococcus; hydatid cysts; submandibular region.

ÖZ

Kist hidatik hastalığı *Echinococcus granulosus*'un yol açtığı bir parazit enfestasyonudur. Köpek dışkısı ile atılan yumurtaların oral yolla alınması ile yayılır. Karaciğer ve akciğerler en yaygın tutulan organlardır. Submandibüler bölgenin kist hidatik ile primer tutulumu son derece nadirdir. Bu yazıda, brankial kleft kisti öntanısıyla submandibüler bölgeden eksize edilen fakat histopatolojik değerlendirmede kist hidatik olduğu gösterilen bir lezyon olgusu sunuldu.

Anahtar Sözcükler: Ekinokokkus; kist hidatik; submandibüler bölge.

Hydatid cyst disease is a parasitic infestation caused by *Echinococcus spp.* The intermediate host of *Echinococcus spp.* are usually herbivores while adult parasites live in the small intestine of dogs and other canidae, which are definitive hosts. Humans serve as accidental hosts in the life cycle.

Echinococcus embryos are transmitted by contaminated food and soil or direct contact with dogs. Eggs attached to the small intestines of humans move through the systemic circulation into any tissue or organ, with liver and lungs being the most common.

Primary involvement of the submandibular region by hydatid cyst is extremely rare.[1,2] In this article, we discuss a mass lesion excised from the submandibular region with an initial diagnosis of branchial cleft cyst, but reported as hydatid cyst on histopathological evaluation.

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Correspondence / İletişim adresi: Murat Doğan, MD. Kayseri Eğitim ve Araştırma Hastanesi Kulak Burun Boğaz Hastalıkları Kliniği, 65200 Kocasinan, Kayseri, Turkey.
Tel: +90 505 - 629 82 62 e-mail *(e-posta):* drmdogan@hotmail.com

CASE REPORT

A three-year-old girl presented with a palpable submandibular region mass for three months. No significant feature was found in her history except urban residence. A subcutaneous, mobile and soft mass lesion (5x2 cm) was palpated at the right side of her neck on physical examination. A few small and soft masses were palpated lateral to the index mass. Laboratory analyses, biochemical tests, complete blood count were found to be within normal range. Serological tests were negative.

On sonography, a cystic mass (25x20 mm) medial to the submandibular gland and a few lymph nodes (largest at 15x10 mm) lateral to the cystic mass were observed (Figure 1 and 2).

The fine needle aspiration biopsy from the cystic mass was reported as inconclusive. Thus, it was decided to perform explorative neck dissection with an initial diagnosis of branchial cleft cyst based on available data. The cystic mass and adjacent solid mass were excised without rupture using blunt dissection (Figure 3 and 4). After bleeding control, the operation was completed without complications. The excised masses were sent for pathological evaluation.

The cystic mass was reported as hydatid cyst while the solid mass was reported as lymphadenopathy (Figure 5 and 6). No additional treatment was prescribed as the hydatid cyst was completely removed without rupture. The patient and her family were screened for Echinococcus and were found to be negative.

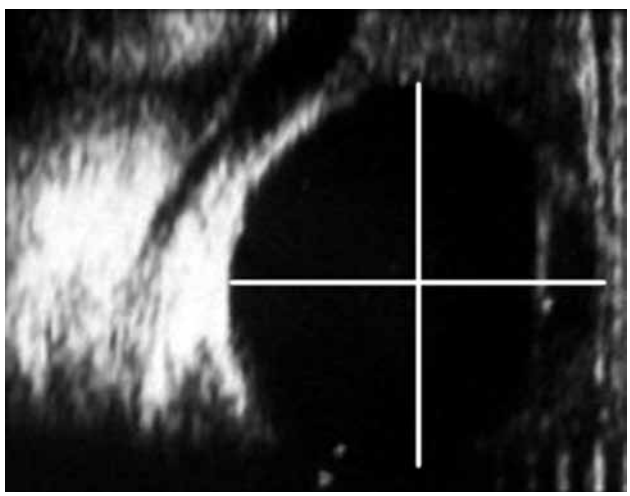

Figure 1. *Hydatid cyst 25x20 mm in size (white lines).*

DISCUSSION

Hydatid cyst disease is a parasitic infestation caused by *Echinococcus spp.* The term "Hydatid Cyst" was first used by Rudolphi in 1808. Two *Echinococcus spp.* cause parasitic infection in humans: *Echinococcus granulosus (E. granulosus)* (%99) and *Echinococcus multilocularis (E. multilocularis)* (%1).[3] The former is the agent in single hydatid cyst, while the latter causes multiple hydatid cysts. *E. multilocularis* has a more rapid course, invasive character and poorer prognosis but it is rarely encountered compared to *E. granulosus*.

It is transmitted via oral ingestion of eggs excreted by dog stool. Larvae released by eggs in the small intestine move through the blood circulation into liver and lungs. If it fails to attach liver or lungs, it passes into the systemic circulation (kidneys, bone, CNS). The disease is termed as hydatid cyst.[3]

Primary cysts most commonly involve liver and lungs.[1] Secondary cysts occur via rupture of cyst wall or spillage of cyst fluid. In the light of these data, the presence of primary or secondary hydatid cyst in the submandibular region is extremely rare and diagnosis is incidental in most cases.[1,2]

Today, the Casoni skin test is not preferred due to cross reactions and potential allergic reactions,

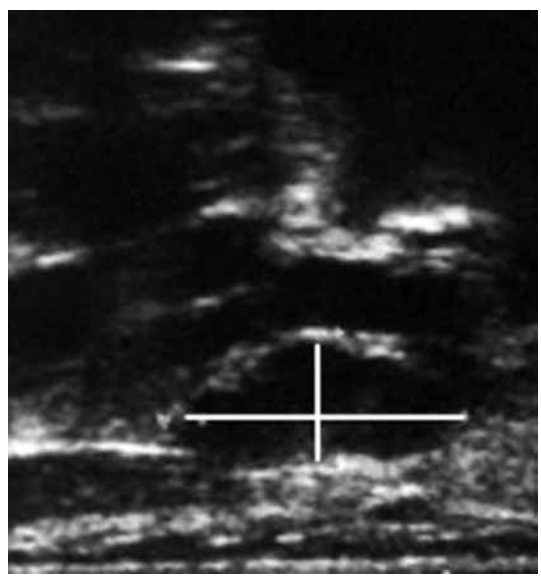

Figure 2. *Lymph node 15x10 mm in size (white lines); a few small lymph nodes adjacent to index lymphe node.*

Figure 3. *Cystic mass and adjacent solid mass; cystic mass is beside 2.5 mL syringe.*

Figure 4. *Cystic mass and adjacent solid mass; solid mass is beside 2.5 mL syringe.*

while complement fixation test (Weinberg test) is not preferred either due to the difficult assay procedure and low sensitivity.[4] Other serological tests are insufficient in the diagnosis of hydatid cyst as they generally have low specificity and sensitivity.[5] In particular, serologic tests have a sensitivity of 50% in extra-hepatic localizations of hydatid cyst.[6] Increased eosinophil count is observed in only 25% of all cases. Hypogammaglobulinemia is detected in 30% of the cases.[7]

Accurate diagnosis is made by detection of characteristic protoscolices or hooklets of *E. granulosus* in cyst fluid samples obtained by aspiration.

On direct radiography, it appears as a round or oval homogenous opacity with well defined margins from parenchyma. On computed tomography (CT), it appears as a well-defined, smooth, thin-walled, homogenous fluid density. As sonography is not sufficient in the diagnosis of pulmonary hydatid cyst, direct radiography or CT is generally used for diagnosis.

The treatment of hydatid cyst is either surgical or medical. Medical treatment indications include cysts smaller than 2 cm, multiple cysts, suspicion of inoculation during surgery or refusal of surgery by the patient. Mebendazole (40-50 mg/kg/day) or albendazole (10 mg/kg/day) can be used in the treatment. In our patient, we did not prescribe medical therapy in the postoperative period because the cyst was >2 cm and there was no rupture or contamination.

Figure 5. *Laminar layer of cyst (H-E x 400).*

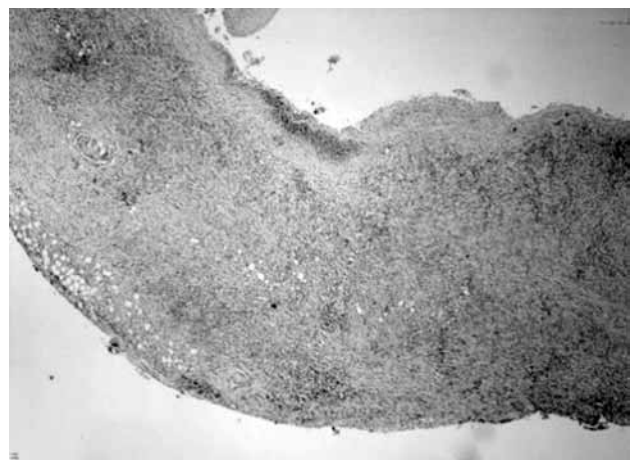

Figure 6. *Pericystic granulation tissue (H-E x 400).*

Our history, physical examination, laboratory evaluations and surgical findings did not suggest hydatid cyst in our patient. We aimed to present this patient who underwent surgery with an initial diagnosis of branchial cleft cyst, as an unexpected diagnosis reported after surgery.

Conclusion

In masses in the submandibular region, hydatid cyst disease can be diagnosed by advanced laboratory analysis. That Echinococcus can involve any organ should not be forgotten and clinicians must be aware of this. Physical examination, radiological evaluations and laboratory tests are only complementary, and definitive diagnosis can only be made by histopathological evaluation.

Declaration of conflicting interests

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