Hydatid Cyst of the Parotid Gland: Case Report

Parotis Bezinin Hidatid Kisti: Olgu Sunumu

***Cüneyd ÜNERİ, MD, *Alev ÜNERİ, MD, *Özmen ÖZTÜRK, MD, **Çiğdem ATAİZİ ÇELİKEL, MD**

* Acıbadem Kozyatağı Hospital, Department of Otorhinolaryngology Head and Neck Surgery,
** Marmara University Medical Faculty, Department of Pathology Department, İstanbul

ABSTRACT

Hydatid cyst disease, an infection of the larval form of *Echinococcus granulosus*, is a serious public health problem. Considering the rare manifestation in the head and neck region, we report a 23-year-old female patient with hydatid cyst of the parotid gland. After routine investigations, superficial parotidectomy was performed, but the patient presented with facial palsy due to a recurrent cyst one year later. The recurrent hydatid cyst was treated surgically, and utmost care was taken to eliminate cyst membrane completely and to sterilize the operative site. The follow-up period is thus far 3-years with no evidence of recurrence. When dealing with cystic masses of the head and neck region, otolaryngologists should be suspicious for hydatid cyst disease especially in endemic areas where widespread infestation is known to occur.

Keywords
Echinococcosis; parotid diseases; cysts; head; neck; surgery

ÖZET

Ekinokokus granulosus'un larva formunun bir infeksiyonu olan hidatid kist hastalığı önemli bir toplum sağlığı problemidir. Baş boyun bölgesinde nadiren görülmesini göz önüne alarak parotis bezi hidatid kisti olan 23 yaşındaki kadın hastayı sunmaktayız. Rutin değerlendirmeler sonrası süperfisyal parotidektomi yapılan hasta bir yıl sonra parotis bezi nüks kistine bağlı gelişen fasiyal palsi ile başvurdu. Nüks hidatid kist cerrahi olarak tedavi edilirken, kist membranının tamamen çıkarılmasına ve operasyon sahasının sterilizasyonuna büyük itina gösterildi. Takip süresi 3 yıl olan hastada nüks saptanmadı. Baş ve boyun bölgesi kistik kitlelerine yaklaşırken KBB hekiminin özellikle parazitik hastalığın yaygın olarak görüldüğü endemik sahalarda hidatid kist hastalığından şüphelenmesi gerekmektedir.

Anahtar Kelimler
Ekinokokkozis; parotis hastalıkları; kistler; baş; boyun; cerrahi

Çalışmanın Dergiye Ulaştığı Tarih: **16.12.2009** Çalışmanın Basıma Kabul Edildiği Tarih: **04.05.2010**

≈

Correspondence
zmen ZT RK, MD
Acıbadem Kozyatağı Hospital,
Department of Otorhinolaryngology, Head and Neck Surgery,
İnönü cad. Okur sok. No: 20
34742, Kozyatağı, İstanbul
E-mail: ozmenozturk@hotmail.com

INTRODUCTION

Hydatid cyst disease (HCD) is a serious public health problem in the sheep raising communities.[1-3] *Echinococcus granulosus* (*EG*) (canine tape worm) is the most common cause of unilocular HCD in human beings.[1]

After ingesting egg contaminated plants, the outer layer of the egg dissolves in the host intestinal tract, liberating an embryo.[1,4] An embryo, having a size of about 30μm, penetrates the mucosa of the host intestine, enters the portal venous system and settles in the liver; or enters systemic circulation to spread throughout the body.[1,4,5] The embryo vacuolates into a large fluid-filled hydatid cyst (HC), 6-10 days after ingestion.[1] The parasite may settle in any site of the body and may rarely manifest in the head and neck region.[2,3,5,6] Considering this rare involvement, we report a case of HC localized to the parotid gland.

CASE REPORT

A 23-year-old female patient presented with a 3-year history of a progressively increasing right preauricular swelling. On physical examination, she had a 30 x 30 mm, immobile, firm, and non-tender mass over the right preauricular region. The computed tomography (CT) revealed a well-demarcated, heterogeneously intense, trabeculated cystic lesion with a measurement of 30 x 30 x 20 mm (Figure 1). Fine-needle aspiration biopsy under ultrasonographic guidance revealed only acellular clear fluid. A differential diagnosis of Warthin's tumor or mucocele was made.

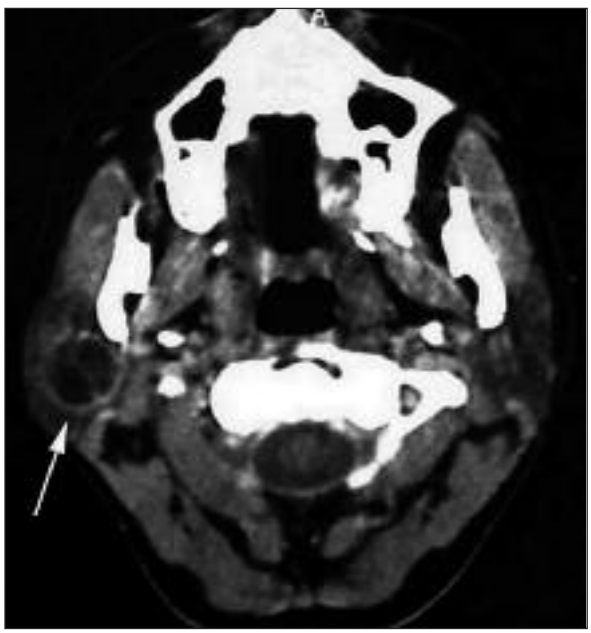

Figure 1. Computed tomography of the right parotid gland revealing a well-demarcated, heterogeneously intense, trabeculated cystic lesion with septations (arrow).

A superficial parotidectomy was performed. No surgical difficulties were encountered, and the postoperative period was uneventful. The histopathologic examination revealed a 20 x 20 x 15 mm cyst, containing clear serous fluid. An easily detachable inner germinal membrane with a thickness of 1 mm showed numerous vesicular projections. An outer acellular laminated cyst membrane with a peripheral mononuclear cellular infiltration was detected. The diagnosis was made of HC. Whole body investigation to rule out multiorgan involvement was negative.

After a follow- up period of 1 year, the patient presented with a facial paralysis of a one month duration, and a preauricular mass over the same region operated on previously. The physical examination showed a 20 x 20 mm, immobile, painless, and firm right preauricular mass and a facial nerve dysfunction with a "House & Brackmann facial nerve grading score" (HBS)[7] of 3/6. Laboratory findings were normal except positive indirect hemagglutination assay for *EG* serology with the titres of 1/1024. Acoustic immitancemetry revealed bilaterally negative acoustic reflexes. The CT scan showed a 15 x 20 x 25 mm, unilocular, well-demarcated, cyctic parotid mass.

After using albendazole (800mg/ day p.o.) for 10 days, a revision surgery was performed. Intraoperatively, a cystic mass measuring 20x20x10 mm in diameter was found to be located deep in the parotid remnant, medial to pes anserinus of the facial nerve. After puncture with a Seldinger needle, injection of the cyst with 20% hypertonic saline and aspiration were repeated until complete removal of the cyst membrane had been achieved. The operation field was thorougly irrigated with hypertonic saline. The histopathologic examination showed a collapsed laminated membrane which was avascular, eosinophilic, refractile, and chitinous, with a germinal membrane. No daughter cysts were detected (Figure 2).

Postoperatively, albendazole was administered for 2 weeks. After a 1-month of follow-up, the facial paralysis recovered completely. Follow-up ultrasonographic evaluation 3 months postoperatively revealed an

Figure 2. Histopathology showing a collapsed laminated membrane which is avascular, eosinophilic, refractile, and chitinous, with a small portion of germinal membrane (arrowhead) (hematoxylin & eosin, original magnification x100).

operation field with a solid appereance. The control serologic tests for *EG* was within normal limits. Follow-up period is thus far 3-years with no evidence of recurrence.

Consent has been taken from the patient and this report was approved by the local ethics committee of our department in accordance with the Declaration of Helsinki.

DISCUSSION

HCD of the head and region is rare, and even in geographic areas where *EG* infestation is endemic, a few cases are reported.[2-4,6,8-10] The diagnosis is based on the history (e.g., close contact with infected dogs, the practice of feeding viscera of home-butchered animals to dogs, domestic and farming duties of rural workers), physical examination, imaging studies (ultrasonography, computed tomography, and magnetic resonance imaging), fine-needle aspiration cytology, and serologic tests (indirect hemagglutination, latex agglutination, immunoelectrophoresis, ELISA, and skin tests).[2,3,9-12] The serologic tests are often used in the follow-up after treatment.[11]

The diagnosis of HC localized to head and neck region is quite challenging for the clinician, especially if no prior HCs in other sites of the body have been found. Imaging modalities are more sensitive than serological tests.[13] The CT examination may show detachment of a laminated membrane as linear areas of increased attenuation within a well-defined cyst (water lilly sign).[13] As in our case, a well-defined cystic mass with trabeculation and septations also suggest the diagnosis.[13]

Although long-term medical therapy with mebendazole or albendazole has been used, the results are still unpredictable.[1] It is generally agreed that medical treatment should be administered to inoperable patients.[1] These drugs also play an important role in conjunction with surgery, both preoperatively for sterilization of HC and postoperatively in case of spillage.

Surgery is the most effective and accepted way to treat HC.[1,2] The removal of the HC without causing any spillage is important, because fatal anaphylaxis due to the high antigenicity of the cystic content occur, and the locally released fluid may result in recurrence.[1,4-6,9,10] Before manipulation of the HC, inactivation of the protoscolices can be achieved by injecting 20% hypertrophic saline solution or 0.5% silver nitrate into the cyst.[9] The irrigation of the operation field should also be performed if there is suspicious spillage of the cyst content.[9] In our case, reasons for the recurrence after the primary operation were due to a suspicious spillage of the cyst content, leaving the endocyst in the field, or the presence of a satellite neighboring cyst. In the secondary operation, care has been given to decrease the risk of recurrence. After puncturing the HC with a Seldinger needle, aspiration and injection with 20% hypertonic saline solution were repeated to provide complete removal of the endocyst, and to minimize the surgical trauma to the facial nerve adherent to a solid and fibrotic parotid gland remnant.

CONCLUSION

HCD should be considered in the differential diagnosis, while interpreting a cystic mass affecting the parotid gland. In endemic areas where HCD is known to occur, physicians should be highly suspicious, and patients with HCD must undergo a systemic investigation to rule out multiorgan involvement with a long-term mandatory follow-up.

Ack now led ge ment

The authors wish to thank Asist. Prof. Jeffrey Howlett from "Kadir Has University, Department of American Culture and Literature" for language editing of the manuscript.

REFERENCES

1. Levinson WE, Jawetz E. Cestodes. In: Levinson WE, Jawetz E, eds. Medical Microbiology and Immunology, 3rd ed. Norwalk, Connecticut: Appleton & Lange;1994. p.266-71.
2. Turgut S, Ensari S, Katirci H, Celikkanat S. Rare otolaryngologic presentation of hydatid cyst. Otolaryngol Head Neck Surg 1997;117(4):418-21.
3. Akyildiz AN, Ozbilen MS, Goksu N. Hydatid cyst of the pterygopalatine fossa. J Oral Maxillofac Surg 1991;49(1):87-8.
4. Shuker S. Hydatid cyst in the maxillofacial region. J Oral Maxillofac Surg 1982;40(3):171-4.
5. Sayek I, Tirnaksiz MB, Dogan R. Cystic hydatid disease: current trends in diagnosis and management. Surg Today. 2004;34(12):987-96.
6. Saxena SK, Chaudhary SK, Saxena GR, Rao S. Hydatid cyst of the parotid gland (a case report). J Postgrad Med 1983;29(2):105-6.
7. House JW, Brackmann DE. Facial nerve grading system. Otolaryngol Head Neck Surg 1985;93(2):146-7.
8. Ennouri A, Rhouma S, Makni S, Bouzouita KA, Marrekchi H, Atallah M. Cervicofacial hydatidosis. Rev Laryngol Otol Rhinol 1989;110(2):169-71.
9. Sennaroglu L, Onerci M, Turan E, Sungur A. Infratemporal hydatid cyst- unusual location of echinococcosis. J Laryngol Otol 1994;108(7):601-3.
10. Hotz MA, Gottstein B. Cystic echinococcosis of the parapharyngeal space: case report with a 20-year follow-up. J Oral Maxillofac Surg 1999;57(1):80-3.
11. Akhan O, Ensari S, Ozmen M. Percutaneous treatment of a parotid gland hydatid cyst: a possible alternative to surgery. Eur Radiol 2002;12(3):597-99.
12. Amice J, Sparfel A, Petillon F, Amice V, Jezequel J, Riviere MR. Hydatid cyst of the neck: diagnosis by fine needle aspiration. Acta Cytol 1992;36(3):454-5.
13. Adaletli I, Yigiter R, Selcuk D, Sirikci A, Senyuz OF. Primary hydatid cyst of the head and neck diagnosed with ultrasound and computed tomography: a report of two cases. South Med J 2005;98(8):830-2.