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Case Report

An unusual presentation of cystic hydatid disease: Left temporal bone involvement with transverse sinus obliteration

Abdulkerim Gökoğlu [a,*], Hüseyin Yiğit [b], Bülent Tucer [c]

[a] Department of Neurosurgery, Private Dunyam Hospital, Kayseri, Türkiye
[b] Department of Medical Services and Techniques, Cappadocia University, Nevşehir, Türkiye
[c] Department of Neurosurgery, Private Kartal Hospital, Kayseri, Türkiye

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ABSTRACT

Cystic hydatid disease (CHD), caused by Echinococcus granulosus, remains a significant global health concern. While commonly affecting the liver and lungs, rare localizations can pose diagnostic and therapeutic challenges. This report presents a case of CHD affecting the left temporal bone, originating from the mastoid cells, resulting in left transverse sinus obliteration and cerebellar hemisphere compression. This unusual presentation highlights the importance of considering CHD in the differential diagnosis of neurological symptoms, including vertigo, even in the absence of more typical clinical signs. We aim to raise awareness among neurosurgeons, neurologists, and otorhinolaryngologists regarding this atypical manifestation of CHD.

1. Introduction

Cystic hydatid disease (CHD), caused by the larval stage of the tapeworm Echinococcus granulosus, is a significant medical and public health problem worldwide [1], particularly in the Mediterranean region, including Turkey, the Middle East, South America, New Zealand, Australia, South Africa, and Southeast Asia [2]. The most commonly affected organs are the liver (63 %) and lungs (25 %), followed by muscles (5 %), bones (3 %), kidneys (2 %), brain (1 %), and spleen (1 %) [3]. This report describes an unusual localization of CHD in the left temporal bone, originating from the mastoid cells, which led to the obliteration of the left transverse sinus and compression of the left cerebellar hemisphere due to mass effect. Its rare localization in bone tissues and potential for erosion make it challenging for both diagnosis and treatment when it is situated in an anatomically complex area like the temporal bone. The fact that this condition originated in the mastoid cells and caused obliteration of the left transverse sinus and compression of the left cerebellar hemisphere explains the emergence of atypical neurological symptoms such as vertigo. This case study is the first report of temporal bone CHD leading to the obliteration of the left transverse sinus.

2. Case report

A 51-year-old male patient presented to the neurosurgery clinic with a left post-auricular mass and vertigo, exacerbated by cold weather (Fig. 1). Although vertigo attacks initially occurred 1–2 times a day, they had been occurring 3–4 times a day in the last two weeks. Each attack lasted at least 2–3 min. The patient stated that the vertigo worsened in cold weather. Neurological examination of cranial nerve functions and cerebellar functions were normal. The mass had been present for two years, and the associated pain and vertigo had developed one month prior to admission. Physical examination revealed a 3 × 2 cm, tender mass in the left post-auricular region. The patient was admitted for further evaluation.

Cranial CT and MRI revealed CHD of the left temporal bone, originating from the mastoid cells, resulting in obliteration of the left transverse sinus and compression of the left cerebellar hemisphere (Fig. 2a). The cyst appeared as a homogenous, hypodense lesion eroding the mastoid bone. The cystic lesion measured 6 cm in diameter. On MRI, it was hypointense on T1-weighted images and hyperintense on T2-weighted images, with hyperintense septations visible on both T1- and T2-weighted sequences (Fig. 2b, c). The lesion had destroyed the mastoid process and extended to the squamous part of the left occipital bone. Serological tests for hydatid disease, including the Casoni test and IHAT, were positive. The erythrocyte sedimentation rate was 20 mm at one

* Corresponding author.
E-mail address: akerimg@hotmail.com (A. Gökoğlu).

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Fig. 1. Painful lump with palpation, 3 × 2 cm in sizes, located in left post-auricular region.

hour, and no other abnormalities were detected in the laboratory work-up. Possible differential diagnoses, such as cystic neoplasms or other inflammatory lesions, were considered but ruled out based on serological tests and imaging findings. Doppler ultrasound revealed no vascularity or flow within the lesion. Additionally, MRI venography confirmed that the left transverse sinus was completely occluded and that the left sigmoid sinus and left internal jugular bulb were not visible (Fig. 2d).

Abdominal ultrasound revealed three cystic lesions of varying sizes in the right lobe of the liver, the largest measuring 3.5 × 2.5 cm. Abdominal CT confirmed these findings, demonstrating two hypodense cystic lesions in liver segment 6 (the largest 2.5 cm), two hypodense cystic lesions in segment 5 (1 and 1.5 cm), and another cystic hypodense lesion in the lower pole of the spleen.

Surgical intervention was performed via a 7 cm left post-auricular incision. Multiple cystic lesions were observed invading the left transverse sinus and mastoid region (Fig. 2e). The cystic masses were carefully removed en bloc without rupture (Fig. 2f). The surgical field was irrigated with hypertonic saline solution. The resulting space was filled with abdominal subcutaneous fat tissue. Pathological examination showed numerous scolices located beneath the laminated cuticular membrane that forms the cyst wall, along with the presence of foreign body reaction-related multinucleated giant cells.

The patient's general condition stabilized rapidly in the post-operative period. On the first postoperative day, a physical examination revealed that the patient's complaints of vertigo and post-auricular pain had resolved. The patient was consulted by the infectious diseases and clinical microbiology department and started on oral albendazole (400 mg twice daily). The treatment was administered in three-month cycles for a total of six months.

To evaluate the effectiveness of the treatment and the course of the cysts, the patient was followed up regularly. A control brain CT scan performed at the end of the six-month follow-up showed that the cystic lesion in the left temporal bone was resorbed and the transverse sinus was re-canalized (Fig. 3). Additionally, an abdominal ultrasound revealed a significant reduction in the size of the cystic lesions in the liver and spleen. Repeat serological tests (Casoni test and IHAT) at the 6th postoperative month showed that the initially positive results had become negative. The patient's clinical and serological follow-up continued for 3 years.

3. Discussion

CHD is endemic in various regions, including the Mediterranean countries (such as Turkey), the Middle East, South America, New Zealand, Australia, South Africa, and Southeast Asia [4]. Its prevalence and incidence in Turkey are 50–400/100,000 and 3.4/100,000, respectively [5]. The patient's history of coming from a region with a high CHD

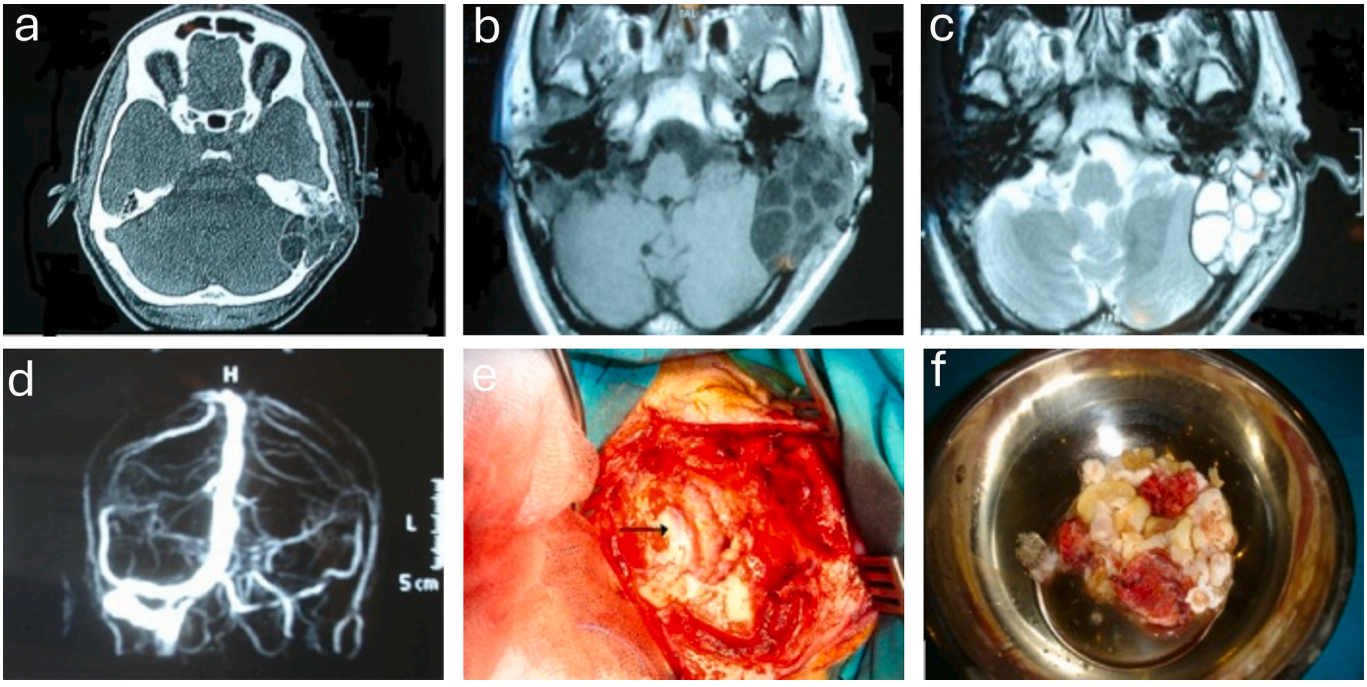

Fig. 2. a: Cranial computed tomography axial sections revealed eroded mastoid bone due to smooth outlining, hypodens cystic lesion 6 × 4 cm in sizes, b, c: T1and T2 weighted magnetic resonance imaging studies revealed T1-W hypointense, T2-W hyperintense multicystic lesion 6 × 4 cm in sizes originating from mastoid cells and eroded mastoid bone and left cerebellar compression due to mass effect, d: MR venography revealed significant decrease left transverse sinus calibration due to mass effect, e: Intraoperative photograph of eroded mastoid bone occluded left transverse sinus multiloculated cystic lesion, f: Operation material consisting of unruptured cysts of hydatid disease.

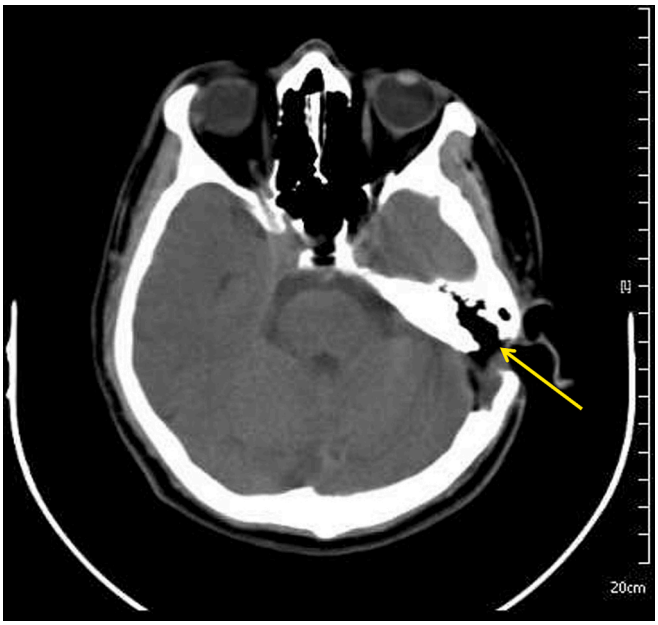

Fig. 3. Postoperative CT image of cystic hydatid disease involving the mastoid cells. Mastoidectomy was successfully performed without damaging the facial nerve, and the left sinus obliteration was treated.

endemicity, such as Kayseri (Türkiye), supports this epidemiological context [6]. The World Health Organization (WHO) reports a substantial global burden of cystic echinococcosis (CE), with an incidence exceeding 50 cases per 100,000 person-years in some regions [7]. The prevalence of cystic echinococcosis varies considerably across endemic areas, ranging from 5 % to 10 %. Furthermore, the prevalence of cystic echinococcosis in slaughtered livestock is estimated to be between 20 % and 95 %, highlighting the significant animal health implications of this zoonosis [2]. Current estimates suggest that approximately 1 million people are affected by cystic echinococcosis worldwide, underscoring the urgent need for effective prevention and control measures [8].

Echinococcosis is a zoonotic disease caused by the tapeworm Echinococcus granulosus [9]. The parasite's lifecycle involves dogs as the definitive host and sheep as the intermediate host, with humans serving as accidental intermediate hosts. Adult tapeworms reside in the intestines of dogs and eggs are shed in their feces [10,11]. Human infection occurs through ingestion of these eggs, which hatch in the intestines, releasing oncospheres that penetrate the intestinal wall and migrate through the bloodstream to various organs, primarily the liver and lung [12,13]. Echinococcus cysts exert their effects on organs through mechanical compression and the release of toxic substances. As cysts enlarge, they compress surrounding tissues and organs. Additionally, the release of cytokines and enzymes from the cyst can lead to bone erosion [14]. Hydatid disease can affect any part of the body. The liver and lungs are the most frequently involved organs in humans, while central nervous system involvement occurs in 1 % to 3 % of all cases [15]. In the presented case, the cyst compressed and invaded the left transverse sinus, compressed the left cerebellar hemisphere, and eroded the mastoid bone. Clinical presentation of central nervous system hydatid cysts is nonspecific and often mimics other space-occupying lesions [16,17]. There are no pathognomonic signs or symptoms. Symptoms typically arise from the mass effect of the slowly growing cyst, leading to neurological deficits depending on the location and size of the cyst. The most common locations for intracranial hydatid cysts are the parietal, frontal, parieto-occipital, and occipital lobes, especially in the watershed areas of the middle cerebral artery [18]. Unusual locations for hydatid cysts have also been reported, including the sellar and parasellar regions, cerebellopontine angle, cavernous sinus, thalamus, ventricles, aqueduct of Sylvius, pons, skull, posterior fossa, and extradural spaces [19]. Several case reports have described hydatid cysts in unusual locations. Ennouri et al. reported the first case of a temporal bone hydatid cyst in a 24-year-old male patient who presented with recurrent left-sided swelling, otalgia, and vertigo [20]. Taghipour et al. described a 25-year-old male with a hydatid cyst in the foramen magnum, presenting with progressive spastic quadriparesis and sensory deficits [19]. Ameli and Abbassioun reported a series of 69 cases with unusual locations, including the cerebellar hemispheres, cerebellopontine angle, fourth ventricle, and pons, but none involving the foramen magnum [21]. Other case series have documented intraventricular, cerebellar, and meningeal locations, as well as involvement of the pons, intrasellar region, cerebellopontine angle, thalamus, petroclival region, and Sylvian aqueduct [22–25]. Hamza et al. conducted a study of 55 hydatid cyst cases examined by CT scan. The mean patient age was 10.5 years, with a range of 3 to 41 years. Most cases involved intracerebral locations, with only one case affecting the cerebellum and two cases involving the temporal bone. The authors emphasized the importance of CT imaging in the diagnosis of hydatid cysts and noted that angiography was not helpful in their series [26]. Llanes et al. reported a case of a 28-year-old male with a temporal lobe hydatid cyst mimicking a brain abscess. The patient presented with seizures and a history of tinnitus [27]. Raynham et al. described a similar case in a 25-year-old HIV-positive female with a multicystic lesion involving the left temporal bone and posterior cranial fossa [28]. A case of a male patient with soft tissue attenuation of the middle ear cleft and multiple cystic lesions showing intracranial spread of CHD in the middle and posterior cranial fossae was recently presented by Kiran et al. [29].

WHO recommends antigen detection using various techniques, including enzyme-linked immunosorbent assay, indirect hemagglutination, latex agglutination test, immunoblot, Western blot, and species-specific polymerase chain reaction. The WHO also recommends regular follow-up for patients with hydatid disease. Treatment typically involves chemotherapy with albendazole for at least 6 months, and surgical intervention may be necessary for cyst removal. Careful surgical technique is essential to prevent cyst rupture, which can lead to anaphylactic reactions and dissemination of the parasite [30].

4. Conclusion

Hydatid cysts can present with diverse clinical manifestations and should be considered in the differential diagnosis of cystic lesions, particularly in endemic areas. A high index of suspicion is crucial for early diagnosis and appropriate management. This case report highlights the importance of considering hydatid disease in patients with unusual presentations, such as temporal bone involvement with transverse sinus obliteration. This case is highly unique because the CHD led to the obliteration of the left transverse sinus, which made its surgical management even more complex. Accurate preoperative diagnosis and meticulous surgical planning are essential to prevent complications and ensure optimal outcomes.

CRediT authorship contribution statement

Abdulkerim Gökoğlu: Writing – original draft, Methodology, Investigation, Conceptualization. **Hüseyin Yiğit:** Writing – review & editing, Writing – original draft, Visualization. **Bülent Tucer:** Supervision, Investigation, Conceptualization.

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Declaration of competing interest

The authors have no relevant financial or non-financial interests to

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Data availability

All data relevant to this case report are presented within the manuscript. For any further inquiries, please contact the corresponding author..

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