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Subdural Empyema Caused by Brucellosis: A Case Report and Review of the Literature

Bruselloza Bağlı Subdural Ampiyem: Bir Olgu Sunumu ve Literatür Taraması

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Abstract

Neurobrucellosis is a rare and challenging complication of brucellosis. Its non-specific clinical presentation makes it challenging to diagnose. The coexistence of neurobrucellosis with subdural empyema is extremely rare, with only a few cases reported in the literature. Herein, we present the case of a 27-year-old male with a ventriculoperitoneal shunt in-situ and a history of exposure to *Brucella* via animal care and consumption of raw milk. The patient presented with a left frontal headache and vertigo following a head trauma. The initial cranial imaging demonstrated a left-sided subdural hematoma and slit ventricles. The patient underwent drainage by craniotomy. *Brucella* spp. was detected in the drained sample, and the patient was treated with a multi-modal approach, which included antibiotics and surgical drainage. The follow-up plan involved a repeat cranial computed tomography and shunt adjustment for recurrent subdural effusions. This case adds to the few reports of neurobrucellosis associated with subdural empyema. Furthermore, it highlights the importance of considering neurobrucellosis in patients with neurological symptoms and a subdural collection in brucellosis-endemic areas.

Keywords: Subdural hematoma, *Brucella*, ventriculoperitoneal shunt, case report

Öz

Nörobruselloz, brusellozun nadir görülen bir prezentasyonudur ve özgül olmayan klinik sunumu tanısını zorlaştırır. Nörobrusellozun subdural ampiyem ile birlikteliği son derece nadirdir ve literatürde sadece birkaç olgu bulunmaktadır. Hayvan bakımı ile uğraşan ve çiğ süt tüketim öyküsü olan, ventriküloperitoneal şant takılı 27 yaşında bir erkek hasta, kafa travmasını takiben sol frontal baş ağrısı ve baş dönmesi şikayetleriyle başvurdu. İlk kraniyal görüntülemelerde sol taraflı subdural hematoma ve slit ventriküller görüldü. Hastaya kraniyotomi ile subdural hematoma drenajı yapıldı, drenaj örneğinde *Brucella* spp. saptandı. Hastaya antibiyotikler ve cerrahi drenaj ile çok yönlü bir yaklaşımla tedavi uygulandı. Takip sürecinde tekrarlayan subdural efüzyonlar için hastaya kraniyal bilgisayarlı tomografi çekimleri, şant ayar değişiklikleri ve tekrarlayan cerrahi müdahaleler yapıldı. Bu olgu, nörobrusellozun subdural ampiyem ile birlikteliğine az sayıda örnek katkıda bulunmakta, nörolojik semptomları olan ve bruselloz-endemik bölgelerde subdural koleksiyonu olan hastalarda bu tanının göz önünde bulundurulmasının önemini vurgulamaktadır.

Anahtar Kelimeler: Subdural hematoma, *Brucella*, ventriküloperitoneal şant, olgu sunumu

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Introduction

Brucellosis, a zoonotic infection caused by a bacteria of the *Brucella* genus, is a significant public health problem worldwide. It is predominantly encountered in the Mediterranean basin, Arabian Peninsula, India, Mexico, and South America^[1]. The disease is transmitted to humans via the consumption of unpasteurized dairy products or a direct contact with infected animals. Brucellosis is an infectious disease that presents with fever, malaise, night sweats, and arthralgias. It can involve multiple organs and is a major diagnostic and therapeutic issue.

Neurobrucellosis is one of the complications of brucellosis. It is an uncommon but severe form of brucellosis, which affects the central nervous system and manifests as various non-specific neurological symptoms. The frequency of neurobrucellosis has been reported as 2-10% of all brucellosis cases^[2]. Therefore, it is an important condition for clinicians to recognize, especially in endemic areas.

Subdural empyema is a rare presentation of neurobrucellosis, with only a few reported cases in the literature. Subdural hematoma, a common outcome of head trauma, can increase morbidity and mortality if it is not recognized and treated in time^[3]. Although chronic subdural hematoma is a more common entity than subdural empyema, they have a similar appearance on computed tomography (CT). Herein, we present a case that highlights the need for a high index of suspicion for neurobrucellosis in patients with a subdural hematoma in brucellosis-endemic regions, especially if there is a history of exposure to risk factors for brucellosis.

Case Report

A 27-year-old male with a ventriculoperitoneal shunt in-situ for aqueduct stenosis was referred to our department from the emergency department for headache and vertigo. The patient admitted to having a simple fall one month ago and regular contact with goats and sheep. He had a Glasgow coma scale score of 15 and slight hemiparesis on the right side. His white blood cell count was $8.99 \times 10^3/\mu\text{l}$, and the C-reactive protein level was 15.02 mg/L.

A CT was obtained, which revealed a left-sided subdural collection with a hyperdense rim. This finding was suggestive of a chronic subdural hematoma, which may have developed due to over-drainage by the shunt (Figure 1). Because the hyperdense rim had an atypical appearance, magnetic resonance imaging (MRI) was obtained. The MRI revealed a hypodense collection with a hyperintense rim in T1 gadolinium-enhanced sequences (Figure 2). Our preliminary diagnosis was a chronic subdural hematoma because the level of the acute phase reactant was low.

After obtaining an informed consent, a left-sided wide craniotomy was performed to drain the hematoma. Intraoperatively, the collection was found to be surrounded by a thick capsule, which when punctured revealed a yellow purulent fluid (Figure 3). Samples from the capsule and the purulent fluid were sent for microbiological and pathological examination. Additionally, the existing shunt's fixed pressure valve (Delta 1.5; Medtronic) was replaced with an adjustable pressure valve (Strata II; Medtronic) set to high pressure.

Postoperatively, the patient's hemiparesis improved, and a *Brucella* spp. was isolated in the sample sent for microbiological

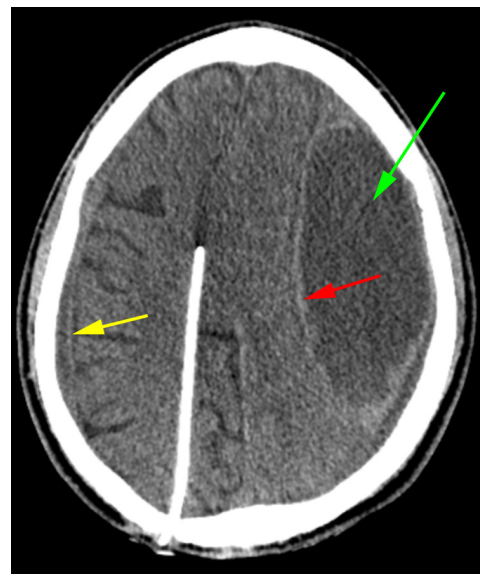


Figure 1. Preoperative CT of the patient demonstrating a left-sided subdural collection (green arrow) with a hyperdense rim (red arrow). A small effusion on the right side can also be seen (yellow arrow)

CT: Computed tomography

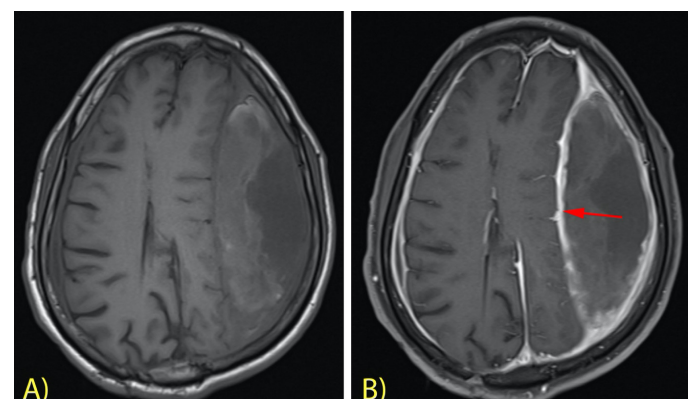


Figure 2. Preoperative MRI of the patient. The (A) T1 axial (B) and T1 gadolinium-enhanced sequences reveal contrast enhancement of the rim (red arrow). This may also be indicative of reactive changes

MRI: Magnetic resonance imaging

analysis. *Staphylococcus epidermidis* was also isolated in the patient's old shunt valve. Although this was mainly considered to be contamination, appropriate antibiotic coverage was planned. The infectious diseases department was consulted, and an antibiotic regimen of ceftriaxone, linezolid, rifampicin, and doxycycline was initiated. *Brucella* serology testing was performed using blood samples, and the Rose Bengal test yielded a positive result. A cerebrospinal fluid (CSF) sample was obtained via lumbar puncture at the fifth postoperative day to rule out meningitis and contamination with *Staphylococcus epidermidis*. The CSF analysis revealed an inconclusive cell count due to a traumatic lumbar puncture, a glucose level of 64 mg/dl, and a protein level of 145.8 mg/dl. The follow-up CSF sample did not yield any bacterial growth.

An echocardiography and ophthalmoscopy were performed to rule out infective endocarditis and endophthalmitis, respectively. Both examinations yielded negative results.

After 1 month in the inpatient unit, the patient was discharged. However, the patient returned after two weeks with slight hemiparesis and focal seizures. A CT was obtained, which revealed a recurrence of the subdural effusion (Figure 4). The craniotomy was extended to perform a total excision of the organized abscess. Because we suspected that an underlying intracranial hypotension was responsible for the juxtaposed chronic subdural hematoma, the middle meningeal arteries were embolized by the interventional radiology department. This is our institutional protocol to prevent the recurrence of a subdural hematoma. The patient was followed up in the inpatient unit for one month and administered parenteral antibiotics. The patient was discharged after a month. At the four-month follow-up the patient has been symptom-free and on an antibiotic regimen of sulfamethoxazole/trimethoprim, rifampicin, and doxycycline.

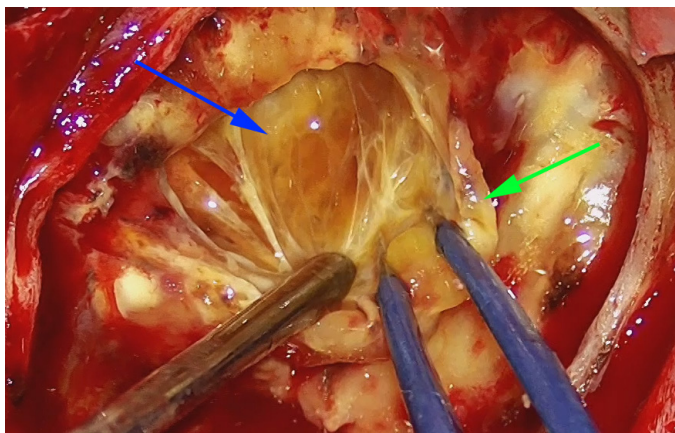


Figure 3. Intraoperative image demonstrating a purulent collection encapsulated within a thick yellow capsule (green arrow), overlaying the brain parenchyma (blue arrow)

Review of Literature

A PubMed-National Center for Biotechnology Information database was searched using the keywords "(subdural) and (brucel*)". The Turkish national database for journals, "TR-Dizin," was also searched using the keywords "*brucella*," "*brusella*," "*brucellosis*" and "*bruselozis*" followed by the phrase "subdural" (Figure 5). A total of 12 studies were found that had been published between January 1990 and February 2024. Of the 12 studies, three were in English and Turkish and their full texts were available (Figure 5). A summary of the study findings is given in Table 1.

Discussion

Chronic subdural hematoma is one of the most commonly treated issues in neurosurgical practice^[3,7]. Thus, according to Occam's razor, we tend to overlook some of the features that might be indicative of more uncommon pathologies. Similarly, in our patient, the hyperintense appearance of the hematoma rim should have made us suspect a more organized abscess formation, rather than a fluid pathology that could be evacuated with a small craniotomy. Our preliminary surgery of internal decompression via a small craniotomy, followed by anti-biotherapy, was not curative. Similarly, an attempt of only drilling burr holes would have allowed for a biopsy alone, warranting another surgery to completely evacuate the organized abscess formation. Both Shoshan et al.^[4] and Zhang et al.^[6] used a large craniotomy to evacuate the empyema. Due to the lack of a mass occupying lesion, no surgery was performed by Tuncer Ertem et al.^[5]. This indicates that surgery may only be warranted in cases with a midline shift or a considerably large lesion.



Figure 4. CT obtained at the time of symptom recurrence demonstrates a large subdural mass (red arrow)

CT: Computed tomography

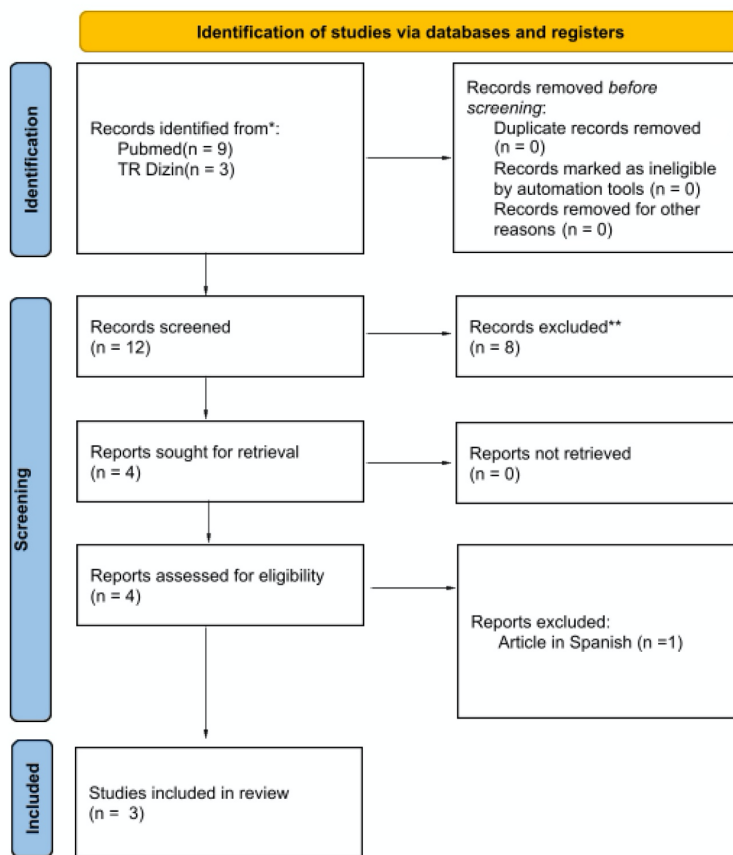


Figure 5. PRISMA flowchart of the literature search

Table 1. Review of the neurobrucellosis cases reported in the literature

Case no.	Author	Date	Age	Sex	Presentation	Location of SDH	History of trauma	Growth in the CSF sample	Diagnosis	Treatment
1	Shoshan et al. ^[4]	1996	8	F	Headache and vomiting	Left frontoparietal region	1	N/A	Empyema culture	Antibiotherapy + Surgery
2	Tuncer Ertem et al. ^[5]	2004	49	F	Fever and blurred vision	Left frontoparietal region	0	<i>Brucella</i> spp.+	Blood and CSF culture and agglutination test	Antibiotherapy
3	Zhang et al. ^[6]	2017	55	M	Fever, aphasia and hemiparesis	Right frontal region	1	N/A	Blood culture	Antibiotherapy + Surgery
4	Present case	2024	27	M	Headache and vertigo	Left parietal region	1	<i>Brucella</i> spp.+	Empyema culture and blood serology	Antibiotherapy + Surgery

CSF: Cerebrospinal fluid, SDH: Subdural hematoma, N/A: Not available, F: Female, M: Male

Conclusion

Although rare, neurobrucellosis may present with a subdural empyema, which warrants evacuation. A hyperdense rim around a collection on CT and contrast-enhanced MRI is a warning sign of neurobrucellosis. Thus, a preoperative MRI should be obtained if there is a clinical suspicion of neurobrucellosis and the patient's status is suitable.

Ethics

Informed Consent: Consent form was filled out by all participants.

Authorship Contributions

Surgical and Medical Practices: B.B.A., A.E., H.B., Concept: M.S.B., T.Y., C.E., B.A., H.B., Design: B.B.A., T.Y., C.E., B.A., H.B., Data Collection or Processing: B.B.A., A.E., H.B., Analysis or

Interpretation: B.B.A., T.Y., C.E., B.A., H.B., Literature Search: B.B.A., A.E., Writing: B.B.A., A.E., M.S.B., H.B.

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