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The cause of persistent fever in a case of meningoencephalitis: infected splenic cyst

Meningoensefalitli bir olguda düşmeyen ateş nedeni: enfekte dalak kisti

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Summary

In this paper, we reported a 15 years old case of acute meningoencephalitis who had persistent fever despite improvement in other clinical and cerebrospinal fluid findings with treatment. In the investigation of fever etiology, abdominal ultrasonography revealed splenic cysts of varying sizes. After percutaneous drainage of the largest cyst, fever decreased dramatically, so it was concluded that the infected splenic cyst was the cause of the persistent fever. Splenic cysts are rare in childhood which are often asymptomatic and detected accidentally. In this case report, the symptoms and treatment modalities of splenic cysts were reviewed.

Key words: *Meningoencephalitis, fever, spleen, cyst*

Özet

Bu makalede, akut meningoensefalit tanısı alıp, tedavi ile diğer klinik ve beyin omurilik sıvısı bulguları düzelmesine rağmen ateşi devam eden 15 yaşında bir olgu sunuldu. Ateş etiyolojisi araştırıldığında, batın ultrasonografisinde değişik boyutlarda dalak kistleri saptandı. En büyük kistin perkutan drenajı ile ateş dramatik olarak düştü ve inatçı ateşin nedeninin enfekte dalak kisti olduğu sonucuna varıldı. Dalak kistleri çocukluk çağında nadir görülen, çoğunlukla asemptomatik seyreden ve tesadüfen saptanan oluşumlardır. Bu olgu sunumunda, dalak kistlerinin semptomları ve tedavi şekilleri gözden geçirildi.

Anahtar kelimeler: *Meningoensefalit, ateş, dalak, kist*

Introduction

Acute meningoencephalitis is a central nervous system (CNS) infection caused most commonly by viruses, but some bacteria can also be causative agent. Upon clinical suspicion, the definitive diagnosis of CNS infection is achieved by cerebrospinal fluid (CSF) analysis and imaging technics. By appropriate treatment, the infection generally improves without any sequelae within 10-14 days (1,2). If the fever does not fall despite appropriate treatment, the pathogen may be resistant to the antibiotics or secondary bacterial, viral, nosocomial infections, as well as localized, undrained infections may have complicated meningoencephalitis (1-3). Among rare causes of persistent fever, spleen cysts generally present with pressure effects on adjacent tissues, or, as a result of rupture, hemoperitoneum, peritonitis and sepsis (4,5).

In this paper, we present a case with meningoencephalitis whose persistent fever disappeared after drainage of the infected splenic cyst.

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Case Report

The patient, 15-year-old girl, was admitted to the state hospital with the symptoms of fever, headache, vomiting, ataxia, and inability to urinate. Upon positive meningeal irritation and cranial magnetic resonance imaging (MRI) findings consistent with meningitis, lumbar puncture was performed revealing 300 leukocyte /mm3 in the CSF. The ceftriaxone therapy was commenced against probable bacterial agents without CSF culture.

Due to persistence of the patient's complaints despite the treatment, the patient was referred to our hospital on 8th day of the disease. The physical and neurological examination of the patient with unremarkable history showed the following results: fever 38º C, increased deep tendon reflexes (DTR), positive clonus, lack of superficial abdominal skin reflexes, and neck stiffness, and ataxic gait. Meningoencephalitis, tuberculosis meningitis, and acute disseminated encephalomyelitis were considered in the differential diagnosis.

The laboratory examination results were as follows: leukocyte 12300/mm³ (74% PNL), CRP 36.8 mg/dl, sedimentation 78 mm/hour. Routine urine, and

biochemical analysis were normal (Table 1). Subarachnoid diffuse contrast agent uptake was present in cranial MRI. Spinal MRI was normal. Meningoencephalitis was considered as the most probable diagnosis, thus cephtriaxone and acyclovir therapies were continued. Spontaneous urination was occurred on the 2nd day of the hospitalization. Due to persistent fever (temperature above 38.5ºC) on the 3rd day, spinal tap was performed. The results of cerebrospinal fluid analysis were as follows: 90 cell/mm^3 (60% PNL), glucose 61 mg/dl with a serum glucose of 118 mg/dl, protein 58,4 mg/dl. In one area of the CSF Gram staining one coccus was seen. The antibacterial therapy was shifted to vancomycin and cefotaxime due to probability of resistant S. pneumonia. Acyclovir was continued. PPD, salmonellosis, brucellosis, cerebrospinal fluid brucella agglutination tests and aerobic cultures were negative. During the 1st week of this treatment ataxia, headache, and other pathological symptoms were resolved, except for fever (39ºC) and elevated acute phase reactants. Thus, the lumbar puncture was repeated with following results in the CSF: glucose 78 mg/dl with serum glucose of 125 mg/dl, protein 39,3 mg/dl, leukocyte 20/mm^3 (Table 1).

On the 13th day, because of persistent high fever despite improvement in clinical and laboratory findings of meningoencephalitis, another infectious focus was searched. On the abdominal ultrasonography (USG) and subsequent abdominal computed tomography (CT) revealed splenic fluid-filled structures, the largest of which is 12 cm in diameter with probably infected appearance (Figure 1).

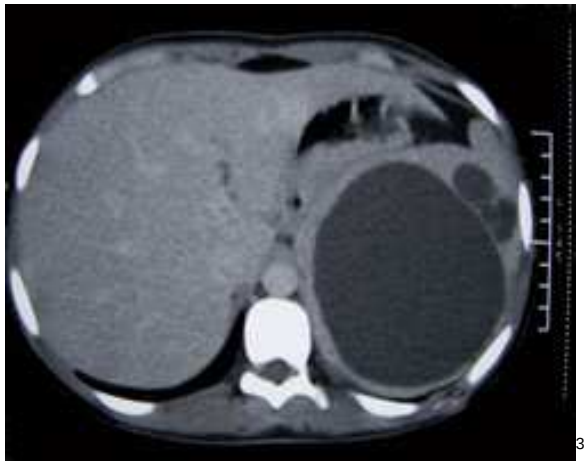

Figure 1. The image of large spleen cyst and satellite cysts in axial CT.

Metronidazole was added to the regimen. Hydatid cyst, a parasitic cyst of spleen, was serologically negative. Upon pediatric surgery consultation CT-guided percutaneous splenic cyst drainage was performed by interventional radiology department for both diagnostic and therapeutic purposes. The aspirated cyst fluid was of yellowish orange color with following findings: 1380 erythtrocyte/mm3, 80 leukocyte/mm^3 (65% PNL), protein 5.6 g/dl, no bacteria on Gram staining. In the evaluation of the aspirated fluid in terms of hydatid cyst, no daughter cyst was determined. Pathology department interpreted the cyst content as benign cytology and no growth were found in aerobic and anaerobic cultures. After drainage of 700-cc cyst content, the fever disappeared with considerable decrease in acute phase reactant levels (Table 1).

Table 1. The results of the laboratory examination before and on 3rd day after the drainage of the cyst

	SH		**ADÜTF Hospital**				**After Drainage of The Cyst**
	First	1th day	3rd day	10th day	13th day	18th day	3rd day
WBC mm^3	12000	12300	16400	17500	16600	32000	7200
Sedimentation (mm/hour)	61	78	---	---	90	98	66
CRP (mg/L)	19,5	36,8	48,6	15	43	31	6
CSF/Blood Glukose (mg/dl)	63/112		58/118	78/125			
CSF protein (mg/dl)			58,4	39,3			
CSF cell/mm^3 (PNL%)	300		90 (60 %)	20			
CSF culture	Absent		no growth	no growth			

SH: State hospital, ADÜTF: Adnan Menderes University Faculty of Medicine

In-depth history of the case revealed an in-vehicle traffic accident 5 years ago and a 1 day monitoring in a hospital due to head and blunt abdominal trauma. Control abdominal USG, was performed 3 weeks after discharge showed multiple splenic cysts, the largest of which was of 8.5cm in size. The patient refused rehospitalization and follow-up.

Discussion

Meningoencephalitis is one of the most important diseases which require early diagnosis and prompt intervention due to its high morbidity and mortality rates. While clinical symptoms vary by age; fever, headache, vomiting, stiffness of neck, and other meningeal irritation findings are commonly seen. Symptoms, although rare, such as ataxia, athetosis, chorea, difficulty with urination due to sphincter dysfunction, or urine or stool incontinence, may be encountered as well. Definitive diagnosis is established through CSF analysis. The treatment is started empirically against the most probable agents depending on the age. In bacterial infections, fever usually falls within 5-7 days by appropriate antibiotic treatment. If the fever does not fall, the pathogen may be resistant to the antibiotics or secondary bacterial, viral, nosocomial infections or localized, undrained infections may have complicated meningoencephalitis (1-3). In our case, the diagnosis of meningoencephalitis was achieved through clinical findings, confirmed by CSF analysis and cranial MRI. While continuing the empirical antibiotic and antiviral therapy during the hospitalization salmonellosis, brucellosis, tuberculosis meningitis, Herpes encephalitis, and CNS demyelinating diseases, were ruled out through further analysis.

The repeated lumbar puncture performed due to persistent fever on the 10th day of the treatment, suggested that there might be a resistance to antibiotics, therefore treatment was rearranged. One week later, despite improving CSF results, leukocytosis, and persistence of elevated CRP levels and high sedimentation rates suggested that there might be a localized, closed infection. Imaging methods revealed spleen cysts. Spleen cysts are rare lesions, and non-parasitic spleen cysts have been reported in few cases in the literature (4). These cysts are often asymptomatic and detected accidentally. According to their etiology and pathophysiology, they are classified as primary/real and secondary/pseudocysts. Primary cysts of the spleen which contain secretory cell layer are of epithelial (dermoid and epidermoid cysts), or endothelial (lymphangioma, hemangioma, polycystic disease, certain serous cysts, neoplastic cysts) in origin. Secondary or pseudocysts of the spleen without secretory cells can be hemorrhagic, serous, inflammatory (acute necrosis-related, chronic tuberculosis), and degenerative cysts (arterial embolism and thrombosis) (4-6). Congenital nonparasitic cysts of the spleen comprise 10% of all spleen cysts (7). Pseudocysts comprise 70-80% of all nonparasitic spleen cysts (5, 8). Spleen cysts, as in our case, are commonly encountered in females and young people between 20-40 years of age (8). Because spleen is the most commonly injured organ in blunt abdominal traumas, as a result of the increase in traffic and sportive accidents, posttraumatic pseudocysts have been reported to occur more frequently. Those cysts are mostly formed following the resorption of subcapsular or intraparenchymal hemorrhage following the trauma, and the subsequent formation of pigment is reabsorbed; thus making the cyst serous (4,5,8). Posttraumatic cysts display an asymptomatic course. Thus, cysts are detected by routine examinations, laparotomies performed for other reasons, or casual USG. Cases mostly present with pain due to pressure effect of the cyst on adjacent tissues leading to pain, and its infection or rupture causing hemoperitoneum, peritonitis, and sepsis (4, 5). Many cases have been reported in the literature which presented with palpable mass in the left upper quadrant; sense of distension; pain in abdomen, chest, left shoulder, epigastrium, back, umbilical area, pelvis, left costovertebral angle, or dyspnea, cough, nausea, vomiting, constipation, dysuria, albuminuria, high temperature, weight loss, or shock profile, along with an unclear history of a trauma. Moreover, a posttraumatic spleen cyst which was ruptured 34 years later, has been reported in the literature (5, 9). When the spleen cyst had been determined, our case recalled having had a traffic accident. Furthermore, despite a 12 cm spleen cyst, she had no symptoms until infected by bacteremia during the course of meningoencephalitis. In the literature, E. coli grew in cyst fluid of a 16 year old patient with a posttraumatic spleen cyst, and the protein content has been found as 7% g. The leukocyte count (65% PNL) and protein content of the cyst fluid of our case were compatible with infection, the absence of growth in culture was attributed to the contunuing antibiotic treatment.

Spleen cysts are diagnosed through imaging methods (USG, CT, MRI). Whether it is primary or secondary cyst can only be determined through histological analysis.

The treatment may differ by age and clinical findings of the patient, and the structure, size, and position of the cyst. Small cysts may remain asymptomatic or may be reabsorbed by the organism. Cysts larger than 4 cm should be treated with surgery because of its potential complications. Approximately 25% of cysts larger than 5 cm are ruptured, leading to intraabdominal hemorrhage and peritonitis (10). Splenectomy has been the preferred treatment modality since 1979.

However, because of risk for developing pneumococcal sepsis after splenectomy, particularly in children; conservative methods have recently been preferred in order to preserve the immunologic function of the spleen (4-8). Conservative treatment methods are percutaneous drainage with or without sclerosing agent, marsupialization, laparoscopic cyst fenestration, and partial splenectomy with the complete excision of cyst. Optimal treatment has been still discussed; however, particularly in children, importance of spleen conservation has been underlined due to the short- and long-term complications of splenectomy. Thus, there has been an increasing tendency to use treatment modalities such as percutaneous drainage or partial splenectomy. Percutaneous drainage, as it is minimally invasive, has been more commonly preferred method, with up to 90% success rate in unilocular spleen abscess. The most significant drawback is cyst recurrence. In a study by Wu et al., cyst recurrence has been observed in 3 of 8 cases after percutaneous drainage (one case has been drained two times). Moreover, individual recurrences have also been reported in the literature (4). Cyst recurrence has not been reported following total excision of cyst and partial splenectomy. Partial splenectomy is often recommended if the size and location of the cyst are convenient (4,11). Pneumococcus, meningococcus, H. influenza type B vaccine before partial or total splenectomy, and a 3- month prophylactic penicillin administration are recommended (10). The infected spleen cyst in our patient was drained percutaneously and no complication was encountered during immediate postoperative period. However, although it was not clear whether the cyst is primary or secondary, the fact that the patient experienced a blunt abdominal trauma 5 years ago, suggested that there might be a posttraumatic secondary cyst. The cyst was recurred 3 weeks later.

In the present case report, the symptomatology and the treatment modalities of rare spleen cysts, which is detected during the course of meningoencephalitis, are reviewed.

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