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Case Report

Lumbar dermoid cyst causing aseptic meningitis

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Abstract. Dermoid cysts are rarely occurring intramedullary spinal cord tumors and they develop due to the inclusion of epithelial elements during closure of the neural tube. They may be detected in the medulla spinalis region extending from cervicomedullary junction to conus medullaris. Spontaneous and iatrogenic rupture of the dermoids causes recurrent meningitis by discharging their contents into subarachnoid space. The purpose of this report is to consider spinal dermoid cysts in the differential diagnosis of young patients with the findings of aseptic meningitis to avoid misdiagnosis and inadequate treatment.

Key words: Abscess, dermoid cyst, meningeal irritation

1. Introduction

Spinal dermoid/epidermoid tumors are congenital tumors developing from inclusion of epithelial cells at the time of neural tube closure. They may rarely cause recurring meningitis (1). Inclusion tumors occurring due to the migration of dermal cells after lumbar puncture have also been reported (2).

Mollaret meningitis is an aseptic meningitis characterized by recurrent attacks without definitive identification of the infectious agent (3). Although numerous causes have been identified in its etiology (4-6), in most published cases, viral infection is the most common cause of the disease (3).

2. Case report

A 12-year-old girl was admitted to our clinic with the complaint of pain in both legs and the waist and urination difficulty. The patient's history revealed that she had been admitted to a hospital two years earlier with complaints of high temperature, headache, waist pain and radicular leg pain bilaterally. She was thought to have meningitis. No microorganisms, except for pleocytosis and a minimum increase in cerebrospinal fluid protein, leading to infection were identified after lumbar puncture. She received a total of 15 days of antibiotic therapy and significant improvement was seen clinically. Following her treatment with the diagnosis of aseptic meningitis, she was discharged from hospital. When her complaints of radicular pain in both legs, waist pain and urination difficulty increased two years later, she was admitted to our clinic. Neurological exam was within normal limits. No cutaneous findings associated with inclusion tumors in the thoraco-lumbar and sacral region were recorded in the physical examination. Routine blood and biochemical tests were within normal limits. Brucella agglutination test was negative. Magnetic resonance imaging (MRI) of the lumbar spine revealed a semisolid cystic lesion measuring 5.5x1.8 cm, with extramedullary extension from conus medullaris inferiorly at the level of lumbar vertebrae 1-2. On T1-weighted MR imaging the well circumscribed semisolid cystic lesion was slightly hypointense compared with the cord, and appeared with hyperintense nodules in the upper and lower pole of the lesion after intravenous injection of gadolinium. A dermal sinus tract was not detected (Figure 1-2). Total laminectomy of L1-L2 was performed. Dura was opened and tethering of the spinal cord was observed. A median puncture was performed. 3 mL of cyst fluid was aspirated. After aspiration of the cyst, intracapsular evacuation of the mass was performed. The mass

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Fig. 1. Magnetic resonance imaging (MRI) of the lesion displays an intra-spinal extension from conus medullaris to the inferior at the level of lumbar 1-2. On T2-weighted sequence the well circumscribed semisolid cystic lesion is slightly hypointense compared with the cord.

Fig. 2. Axial section of the lesion passes through at the level of L2 vertebra corpus.

contained material in dirty grayish yellow color, hair and other neuroepithelial elements. Intramedullary and intradural extramedullary parts of the mass were totally removed together with the capsule. The capsule was not harmed except at the puncture site. No bacteria were detected by the gram stain and no proliferation was observed in the cultured medium. A remarkable improvement was noted in the complaints of leg and waist pain, in addition to improvement of urination difficulty. Control examination performed one and a half years after treatment revealed that radicular complaints and sphincter defects disappeared completely.

2. Discussion

Dermoid, epidermoid, lipoma, teratoma and neuroenteric cysts are inclusion tumors caused by abnormal dysembryogenesis (7). These tumors constitute about 4% of intramedullary tumors in childhood (8). 50% of the spinal inclusion tumors are intradural and extramedullary in location as in our case; the other half is intramedullary in location (9,10). Generally 60% of these tumors are located in the lumbosacral region involving the cauda equina and conus medullaris (11). They may be accompanied by cutaneous lesions, sinus tracts, occult ventral or dorsal raphism or split cord malformation (12). They most frequently lead to leg pain, sensorimotor deficit and sphincter dysfunction. Radicular pain is generally observed in extramedullary tumors (13,14). As in our case, bowel and bladder dysfunction is commonly characteristic of late-stage symptoms of these tumors (15). Spontaneous rupture of the epidermoids causes recurrent meningitis by discharging their contents into subarachnoid space (16). The ruptured wall heals and then re-ruptures, leading to recurrent symptoms (3). Epidermoid tumors may also be observed iatrogenically following lumbar punction. The interval varies between 2 and 6 years in most reported cases (2). Absence of cutaneous signs, as in this case, may result in non-recognition of the dermoid tumor as the cause of aseptic meningitis. In our case, the undamaged tumor capsule suggests that the previous aseptic meningitis attack was caused by rupture of the tumor capsule that subsequently healed. When rupture occurs, cystic contents spread through subarachnoid space and ventricular system. Clinical symptoms of acute rupture vary, and may include headache, nausea, vomiting, dizziness, vision impairment, aseptic chemical meningitis, hemiparesis, mental changes and coma.

Mollaret's meningitis was first reported in 1944 to define recurrent aseptic meningitis with no obvious infectious agent (3). Most published case reports have suggested viral infections in the etiology (3). Vogt-Koyanagi syndrome, Harada syndrome, Behçet's disease (4), allergy, systemic lupus erythematosis, Familial Mediterranean

Fever (5), Whipple's disease, intracranial hydatid cyst, sarcoidosis and epidermoid cyst are causes rarely encountered in the etiology of Mollaret's meningitis (6). Even though Mollaret's meningitis has been suggested as a self-limited disease (4), this may not be the case when an underlying pathological cause is identified. Microorganisms from skin flora in cases associated with a dermal sinus are the most prevalent causes of abscess formation. However, occurrence of abscess formation in cases without a dermal sinus is rare. Previous studies have demonstrated that spinal intramedullary abscess may occur during the course of systemic infections such as Brucellosis (17). Epidermoids causing recurrent meningitis by discharging their contents into subarachnoid space also have been reported (7). Aseptic meningitis is inflammation of the meninges with CSF lymphocytic pleocytosis and no cause apparent after routine CSF stains and cultures. Viruses are the most common cause. Other causes may be infectious or noninfectious. Symptoms include fever, headache, and meningeal signs (18). Altough in the first time our patient admitted to the hospital with complaints of high temperature, headache, waist pain and radicular leg pain bilaterally and tought as a bacterial menengitis, there was no microorganisms, except for pleocytosis and a minimum increase in cerebrospinal fluid protein. Hence, in our case instead of bacterial meningitis we tought that the cause of the meningitis is associated with the rupture of dermoid cyst.

Content of dermoid cysts, containing hair and sebaceous material, causes heterogeneous signal variation on MRI (15). Similar to our case, high lipid content of most spinal dermoids causes hyperintense appearance on T1 and T2 weighted image sections (19).

Ideal treatment of spinal dermoid tumors differs according to the intramedullary or extramedullary location of the tumor. Total excision is not suggested in intramedullary dermoid cysts since the capsule is adherent to the cord and may cause additional neurological deficits (20).

Complete excision was performed to our patient since the dermoid cyst located in the conus medullaris and was not in an intramedullary location. Possibility for the presence of spinal dermoid cyst in the differential diagnosis of patients demonstrating meningeal irritation accompanied by radicular leg pain should not be disregarded. The correlation between radicular leg pain and history of childhood meningitis is important with respect to the differential diagnosis.

References

1. Messori A, Polonara G, Serio A, Gambelli E, Salvolini U. Expanding experience with spontaneous dermoid rupture in the MRI era: diagnosis and follow-up. Eur J Radiol 2002; 43: 19-27.
2. Ziv ET, Gordon McComb J, Krieger MD, Skaggs DL. Iatrogenic intraspinal epidermoid tumor: two cases and a review of the literature. Spine 2004; 29: 15-18.
3. Gao B, Yang J, Zhuang S, et al. Mollaret meningitis associated with an intraspinal epidermoid cyst. Pediatrics 2007; 120: 220-224.
4. Bruyn GW, Straathof LJ, Raymakers GM. Mollaret's meningitis: differential diagnosis and diagnostic pitfalls. Neurology 1962; 12: 745-753.
5. Vilaseca J, Tor J, Guardia J, Bacardi R. Periodic meningitis and familial Mediaterranean fever. Arch Intern Med 1982; 142: 378-379.
6. Schwartz JF, Balentine JD. Recurrent meningitis due to an intracranial epidermoid. Neurology 1978; 28: 124-129.
7. Agnoli AL, Laun A, Schönmayr R. Enterogenous intraspinal cysts. J Neurosurg 1984; 61: 834-840.
8. Steinbok P, Cochrane DD, Poskitt K. Intramedullary spinal cord tumors in children. Neurosurg Clin N Am 1992; 3: 931-945.
9. Barth A, Pizzolato GP, Berney J. Melanocytome meinge intramedulaire. Neurochirurgie 1993; 39: 188-194.
10. Mark Mumenthaler, Otto Appenzeller (eds). Neurologic differential diagnosis, New York, Georg Thieme Verlag 1992: 156-159.
11. Graham DV, Tampieri D, Villemure JG. Intramedullary dermoid tumor diagnosed with the assistance of magnetic resonance imaging. Neurosurgery 1988; 23: 765-767.
12. Gregorios JB, Gree B, Page L. Spinal cord tumors presenting with neural tube defects. Neurosurgery 1986; 19: 962-966.
13. Stein BM. Surgery of intramedullary spinal cord tumors. Clin Neurosurg 1979; 26: 529-542.
14. Froment A, Celi P, Palma L. Oligodendrogliomas of the spinal cord. Acta Neurochir 1980; 52: 305-329.
15. David G. Mclone. Anomalies of the spine and spinal cord. In Bermans J Iskandar and W Jerry Oakes, eds. Chicago. Pediatric Neurosurgery 2001; 307-317.
16. Aristegui FJ, Delgado RA, Oleaga ZL, Hermosa CC. Mollaret's recurrent aseptic meningitis and cerebral epidermoid cyst. Pediatr Neurol 1998; 18: 156-159.
17. Helvaci M, Kasirga E, Cetin N, Yaprak I. Intramedullary spinal cord abcess suspected of Brucella infection. Pediatrics International 2002; 44: 446-448.
18. Rotbart HA. Viral meningitis. Semin Neurol 2000; 20: 277-292.
19. Peter C. Burger, Bernd W Scheithauer. Benign Cystic Lesions; Tumors of the Central Nervous System. Washington 1994; 362-365.
20. Froment A, Celi P, Palma L. Oligodendrogliomas of the spinal cord. Acta Neurochir 1980; 52: 305-329.