Double meningocele Case Report

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SUMMARY: Durmaz R, Arslantaş A, Özön YH, Tel E. Double meningocele: case report. Turk J Pediatr 2000; 42: 331-333.

The coexistence of two distinct meningoceles of the spine is a very unusual event. We report a three-day-old boy with double meningoceles at the thoracic and lumbar levels. The connection between the stalk of the thoracic meningocele and the spinal cord, as seen on magnetic resonance imaging, showed a neurological involvement in this lesion. Our case is only the third without association of congenital anomalies or neurofibromatosis to be reported to date.

Key words: double meningocele, tethered cord.

The occurrence of double meningocele and/or myelomeningocele in different regions of the spine is an exceedingly rare event. Apart from cases with an association of congenital anomalies or neurofibromatosis, only two cases are reported in the literature[1,2]. We present a case with double meningoceles, of which one was connected to the spinal cord.

Case Report

The patient, a three-day-old male, was admitted to the Neurosurgical Department of Osmangazi University Hospital with two cysts located on the spine. He was born by cesarean section to a 30-year-old mother at 40 weeks of gestation. He weighed 4000 g at birth (90th percentile) and was 51 cm in length. He was the third child of non-consanguineous parents. His mother's second pregnancy was terminated due to congenital hydrocephalus at 28 weeks of gestation.

Physical examination revealed two celes in the midline, one in the thoracic and the other in the lumbar area (Fig. 1). The one overlying the upper thoracic region was 6x7 cm and appeared more tense than the lumbar cele, which was 3x2 cm in diameter. Both lesions were widely covered by skin but only partly by an opaque membrane at the top of the fundus. The patient's head circumference was 37 cm (over 90th percentile) and no congenital anomaly was found. There was no sign of neurofibromatosis, and the neurological examination was normal. There were no numerical or structural anomalies in the GTG-banding chromosome karyotype analysis of blood samples taken from the patient or from his parents.

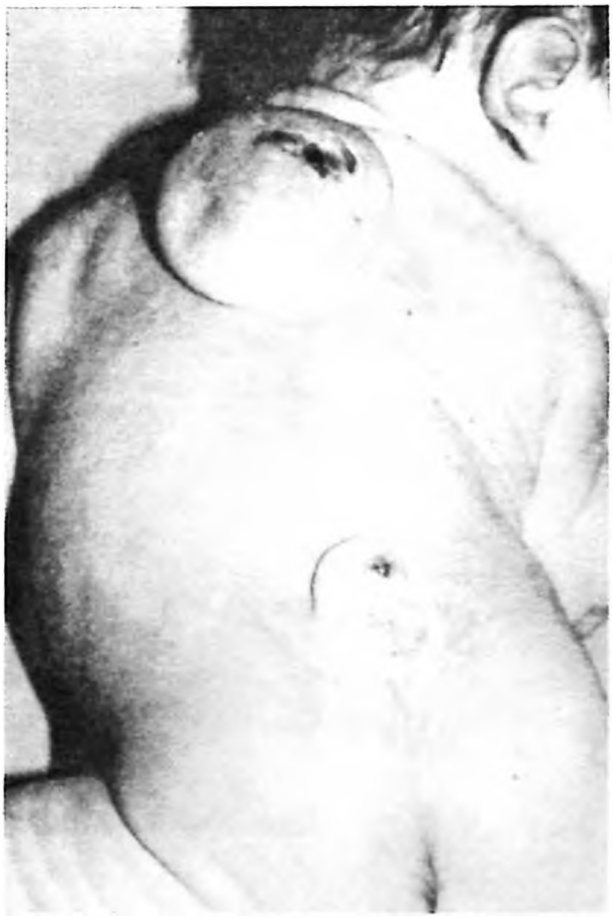
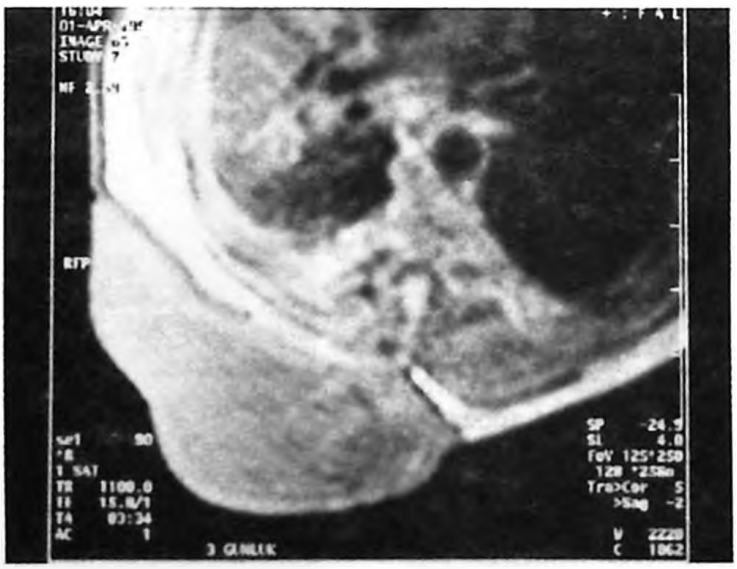


Fig. 1. The patient with double meningomyelocele at the dorsal and lumbar areas of the spine

An X-ray radiogram of the spine was normal. On magnetic resonance imaging (MRI) the thoracic sac was shown to be covered by skin and connective tissue, and there was a connection between the spinal cord and the neck of the lesion (Fig. 2).

Both lesions were explored at the same time through a circumferential incision. Both sacs contained clear cerebrospinal fluid but no neural tissue; however, neural elements were seen attached to the stalk of the thoracic sac. A partial laminectomy was performed on the bony

(a)



(b)

Fig. 2. a) The sagittal T1-weighted MRI shows two distinct sacs and also a neural connection between spinal cord and the stalk of the thoracic meningocele. b) A neural attachment to the neck of the lesion on the axial T1-weighted MRI of the thoracic cele.

structure surrounding the neck of the lesion and the neural elements were released as far as possible. Both defects were repaired with subcutaneous fascia and normal skin.

The patient developed no neurological deficits nor hydrocephalus over the duration of a one-year follow-up.

Discussion

The incidence of meningoceles in Turkey is reported to be high[3]: seven cases were found among a total of 41,785 deliveries[4]. The coexistence of meningocele and/or myelomeningocele in different regions of the spine is very unusual; to the best of our knowledge, only six cases of such an occurrence have been reported to date. Unfortunately, two cases with thoracic and lumbar meningoceles were associated with neurofibromatosis type 1[5]. Potter[2] noted two such occurrences, one of which was associated with hydrocephalus and Arnold-Chiari malformation. Bertan and Wilson[6] also reported a similar case of a patient who underwent operation for double myelomeningocele and died from bronchopneumonia as a result of a ventriculo-pleural shunt complication. Bailey[1] reported a case of double meningocele of which one contained neural elements. Following the first case of Potter and the second case of Bailey, our case is only the third without congenital anomalies or neurofibromatosis to be published to date.

Etiological factors responsible for spinal dysraphism are not known. Dietary causes and vitamin deficiency, such as of folic acid, especially during the critical period of neurolation between 18 and 21 days of gestation, may play a role in its development[7]. In addition, karyotype analysis of chromosomes was performed on blood samples from our patient and from his parents, but no abnormality was found.

The MRI findings of dysraphic thoracic cele may disclose cord involvement in our case, which showed a connection between the stalk of the cele and spinal cord. The term "meningocele with tethered cord" has been coined for this type of lesion, and found in nearly 50 percent of patients with meningocele[8].

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