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Exceptional case

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A lateral meningocele in a 48 years lady revealed by a CSF fistula. Exceptional case

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ABSTRACT

Background: Lateral meningocele is defined by the presence of protrusions of the arachnoid and the dura matter extending laterally through inter- or intravertebral foramina. It is an extremely rare condition; to the best of our knowledge, only a few cases are reported in the literature and most of them in childhood.

Case presentation: Authors reported a case of a 48 years old lady who consulted for a lombo-sacral mass right-sided with a CSF fistula. The Spinal MRI objectified a meningocele lateralized in the right side associated with multiples malformations. The patient underwent surgery and the meningocele was closed after excision of the associated subcutaneous lipoma. The long-term outcome was favourable and the follow up was assured by clinical examination monthly in the first year.

Conclusion: Lateral meningocele is very rarely reported, it is usually associated with multiples malformations. Surgical treatment is a good option for treatment for avoiding complications. The prognostic depends on the preoperative status and the associated malformations.

INTRODUCTION

Lateral meningocele is defined by the presence of protrusions of the arachnoid and the dura matter extending laterally through inter- or intravertebral foramina.

These often occur in the setting of Marfan syndrome, neurofibromatosis type 1 or lateral meningocele syndrome but may also be seen as isolated anomalies.

It is extremely rare condition, and only few cases are reported in the literature and most of them in childhood. The most reported cases are in thoracic and cervical regions. The localization at the sacral spine is very infrequent [1].

Here we report the management of a fistulized lumbo sacral lateral meningocele in a 48 years old lady in the department of neurosurgery of Bab El Oued teaching hospital of Algiers Algeria.
CASE PRESENTATION

Madame B N aged of 48 years with past medical history of surgical intervention for a right foot deformation consulted in the emergency unit of our department for Cerebro Spinal Fluid (CSF) leaking in the lumbosacral region through a small mass. The patient reported that she was born with a small mass in the lumbo sacral region but any investigation was performed and she never consulted for. One year before, she was victim of a traffic accident and since date the volume of the mass grows progressively with the extension toward the right side.

At the admission the clinical exam found conscious patient in good general health, temperature 37 degree celcius, complaining of back pain and a paraparesia coted 4/5, a right clubfoot with stigmas of previous surgical interventions. There was no sensory or sphincter disturbance and no <<café au lait>> lesions. In the lumbo sacral region right sided there was a mass of 15 cm of size, well epidermised with large implantation base (Figure 1). Through the masse there was a fistula of Cerebro Spinal Fluid (CSF).

The biological investigations are normal; Spinal MRI (Figure 2) objectified a spinal cord extension up to S1 and a subcutaneous liquid mass extending to the right sacral region as a meningocele. This investigation revealed other malformations; a tethered spinal cord at S1 level, a subcutaneous lipoma, a malformative L2-L3 fusion.

We operated the patient under general anesthesia, on prone position. We performed an “ogive fashion” skin incision, after the excision of the subcutaneous lipoma, we dissected the meningocele and found a fibrous stalk attached in the wall of the meningocele, this fibrous element was detached before the closure of the meningocele using silk 3/0. (Figure 3)

The post-operative outcome was marqued by the relief of back pain and improvement of paraparesia. The patient exited from the hospital seven days after surgery with appointment in three days for total removal of sutures. After 3 days the patient came back for the appointment but we discover a liquid collection, we use a needle gauge to carry out a puncture of 125 cc of serum bloody. The bacteriological study revealed a staphylococcus a coagulase negative. We put her under Acetazolamide 250mg twice daily, oxacillin 1g bid daily. The evolution was uneventful and the patient was discharged from the hospital after 7 days of treatment with appointment in one month. She was followed up monthly and after 2 years there is no complaint. She recovered the motor disturbance with the help of physiotherapy.

Figure 1. Pre-operative image of the Mass in the lumbo sacral region right sided.

Figure 2. Spine MRI showed a spinal cord extension up to S1 and a subcutaneous liquid mass extending to the right sacral region as a meningocele.

Figure 3. Per operative image showing the walls of the meningocele.
**DISCUSSION**

Meningomyelocele are the most common forms of neural tube defects. Lateral presentations are extremely rare [2, 5], the incidence of lateral meningoceles was reported to be 0.3% [4].

These lateral presentations often occur in the setting of Marfan syndrome or neurofibromatosis type 1 but our patient did not present any café au lait lesions.

Lateral meningocele is also seen in Lateral meningocele syndrome.

This syndrome is a rare disorder, originally described by Lehman et al. in 1977 [3]; Patients with lateral meningocele syndrome present widened spinal canal with scalloping of the posterior surfaces of the vertebral bodies and multiple lateral meningoceles, frequently associated with distinctive craniofacial features such as downslanting palpebral fissures, ptosis, mandibular hypoplasia, a high palate, and skeletal abnormalities such as hypoplasia of the posterior arch of the atlas, short stature, scoliosis, and kyphosis [3], our patient did not present any distinctive craniofacial features.

To the best of our knowledge lateral meningocele in adult not associated with Marfan syndrome or neurofibromatosis type 1 or Lateral meningocele syndrome has not been published earlier. Many meningomyelocele often are associated with tethered cord [2].

Some authors reported that Lateral meningoceles are usually associated with vertebral defects such as hemivertebrae, scoliosis, absence of neural arches on the affected side, and widening of the spinal canal and intervertebral foramina. Scalloping of the pedicles, laminae and vertebral bodies that are adjacent to the meningocele result in an enlarged spinal canal. Butterfly vertebra and segmental anomalies of the vertebral bodies may be found in as many as 43% of affected patients. Sacral anomalies, such as confluent sacral foramina and partial sacral agenesis, occur in up to 50% of reported cases [6, 7].

Our patient presented many associated malformations, a tethered spinal cord at S1 level, a subcutaneous lipoma, and a malformative L2-L3 fusion.

A patient with lateral meningoceles may remain asymptomatic or may suffer from paraparesis or back pain [8]. Chronic symptoms in lateral meningoceles usually arise when meningeal protrusions compress against or deform the adjacent structures such as vertebral bodies, nerves, and viscera.

The index patient presented a mass since birth but the clinical sign were back pain and paraparesia. The consultation was motivated by the CSF fistula upon the right sacral mass. Patient leaving in some regions did not come to consultation early because of ignorance or poverty.

Surgical closure is the treatment of choice, because the lateral meningocele does not have spontaneous regression and generally progresses its enlargement with a corresponding increase in the risk of complications like bladder or bowel dysfunction or neurological deficits. Surgical excision may be indicated in cases where giant and symptomatic cysts are present and they are causing bladder or bowel dysfunction or neurological deficits [9].

**CONCLUSION**

Lateral meningocele is a very rarely reported entity and only few published cases are reported in the literature. Surgical treatment is a good option of treatment avoiding complications. The prognostic depends on pre-operative status and the associated malformations.

**CONFLICT OF INTERESTS**

None

**REFERENCES**