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CASE REPORT

Intractable post dural puncture headache in a patient with undiagnosed intrathoracic meningocele

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SUMMARY

Post dural puncture headache (PDPH) is a troublesome complication of dural puncture. Although its risk factors and management have been extensively discussed in the literature, the underlying cause may sometimes remain unclear, making treatment challenging for anaesthesiologists. We describe a noteworthy case of PDPH following spinal anaesthesia administered for an emergency cesarean section in a patient with known neurofibromatosis. Despite conservative and pharmacological treatment, followed by an autologous epidural blood patch, no symptomatic relief was achieved. Neuroimaging of the brain and spine revealed a large intrathoracic meningocele (IM). Although rare, intrathoracic meningocele should be considered in patients with neurofibromatosis who develop intractable PDPH after dural puncture, with neuroimaging being the most effective diagnostic tool.

Keywords: Dural puncture; Epidural blood patch; Intrathoracic meningocele; Neurofibromatosis; Neuroimaging; PDPH.

Introduction

Intrathoracic meningocele (IM) is a rare entity, and its association with neurofibromatosis was first described by Phol in 1933.[1] The diagnosis of IM is usually incidental and is best visualized by magnetic resonance imaging (MRI), which delineates its anatomy and its connection to the dural sac. The associated bony deformities are better seen in computerized tomography of the spine.[1]

Post dural puncture headache (PDPH) is a positional headache usually presenting within 3 days of dural puncture. In the postpartum period, it interferes with breastfeeding and extends the hospital stay, requiring a more vigorous approach in its management. Unresolved PDPH may warrant the use of neuroimaging studies to rule out any pathological cause.

We are presenting a case of PDPH following spinal anaesthesia for an emergency cesarean section, which did not respond to established treatments and was found to be associated with IM. We obtained written informed consent from the patient before writing this case report, and all attempts were made to ensure anonymity.

Case Report

A 33-year-old, 65 kg full-term primigravida was posted for emergency cesarean section due to fetal distress. Clinical examination revealed multiple nodular lesions throughout the body, and she was a diagnosed case of neurofibromatosis (Fig. 1). Her medical history included asthma for the past 3 years and use of inhalers during winters. Chest auscultation confirmed mild wheeze, especially on the left side. There were multiple neurocutaneous lesions on her back, but the lumbar spine appeared externally normal (Fig. 1). The rest of the clinical examination and blood investigations were within normal limits.

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In the operating room, essential monitoring was started with non-invasive blood pressure, pulse oximetry, and electrocardiogram. An 18 G IV cannula was secured, and lactated Ringer's solution was started. The patient was given spinal anaesthesia with a 25 G Quincke's spinal needle in the sitting position, with 2.2 ml of bupivacaine heavy 0.5%. The surgery was uneventful, and the patient was shifted to the postoperative ward. On the 2nd postoperative day, the patient started complaining of headache upon assuming the sitting position, which was minimized in the supine position. She was advised bed rest, and her oral and intravenous fluid intake was increased to improve hydration. She was also advised to increase tea or coffee intake, and paracetamol 650 mg was started every 6–8 hours. We escalated the treatment by adding non-steroidal anti-inflammatory drugs and tramadol. When there was no relief in 2 days, we aseptically performed an epidural blood patch (EBP) with 20 ml of autologous blood. However, it also failed completely, and her headache worsened. Therefore, we decided to perform magnetic resonance imaging (MRI) of the brain with screening of the whole spine. The findings were astonishing, showing upper dorsal spine scoliosis with left convexity and a large left lateral meningocele (10 cm×12.2 cm×15.9 cm) at D1–D5 vertebral levels, extending into the left paravertebral spaces and left hemithorax (Fig. 2).

The patient was referred to the neurosurgery and cardiothoracic surgery departments in view of these findings for further management. She was advised to stay in follow-up and to undergo cystoperitoneal shunt or total excision only when surgically indicated. Further imaging included a chest X-ray posteroanterior view showing a large cystic opacity occupying most of the upper and mid lung zones (Fig. 3). Contrast-enhanced computed tomography (CECT) of the thorax also showed the lesion occupying the left upper and mid hemithorax, causing significant collapse of the left upper lobe and lingula, along with a mass effect on the left lower lobe and mediastinum (Fig. 4). Spirometric tests showed moderate restrictive ventilatory impairment. Her electrocardiographic and echocardiographic findings were within normal limits. Over a period of 2 weeks, her PDPH resolved with conservative management, and she was discharged home with advice to stay in follow-up with the neurosurgery and thoracic surgery departments.

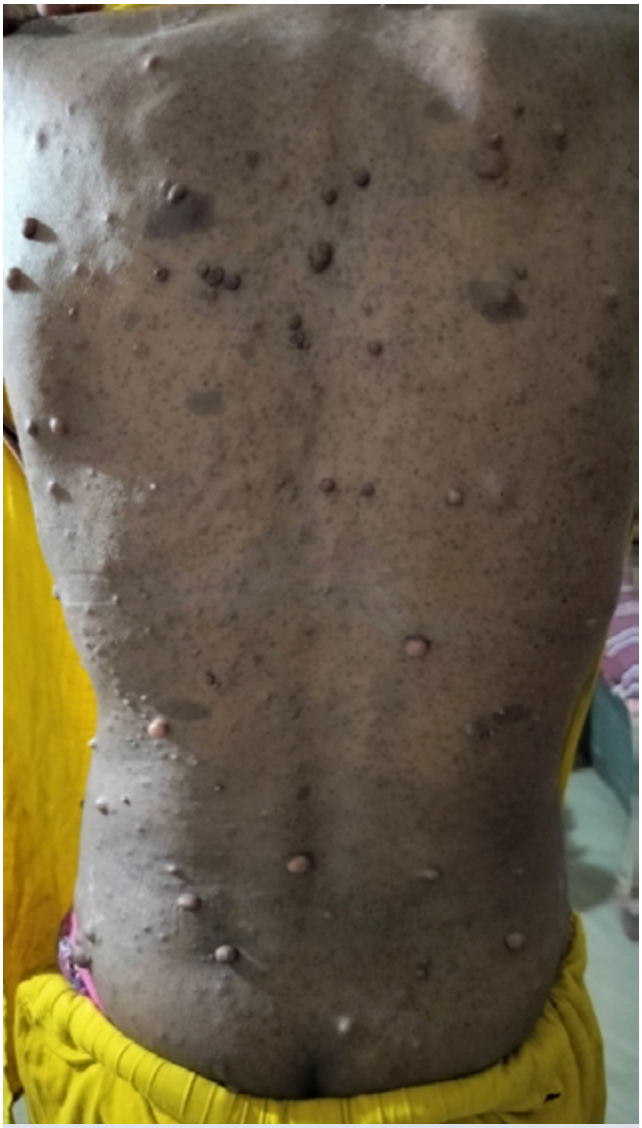

Figure 1. Neurocutaneous lesions confirming neurofibromatosis.

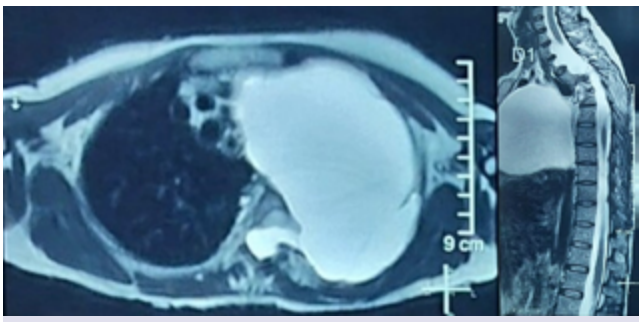

Figure 2. MRI dorsal spine and thorax.

Discussion

Intrathoracic meningoceles are a rare entity, with 60–85% associated with neurofibromatosis type 1.[2] They may remain asymptomatic for many years and are mostly found incidentally when imaging studies are performed for other reasons. Symptoms are re-

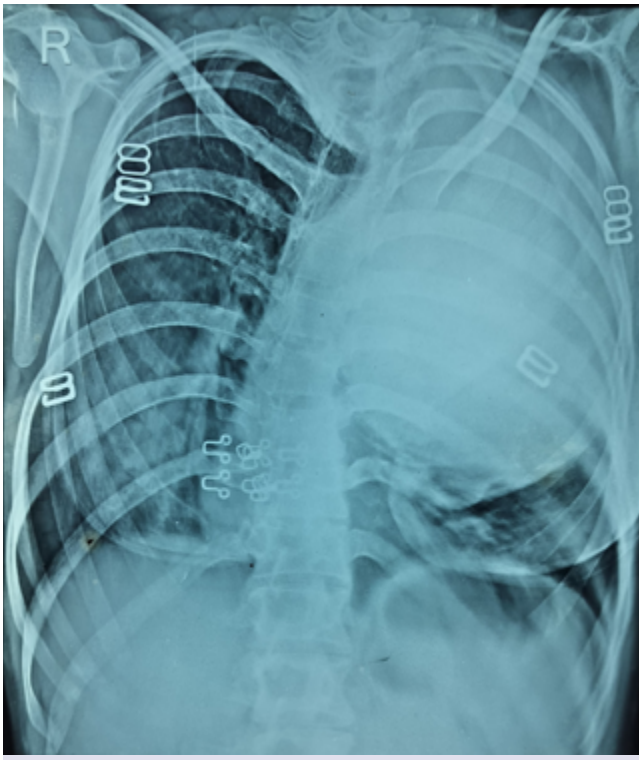

Figure 3. Chest X-ray PA view.

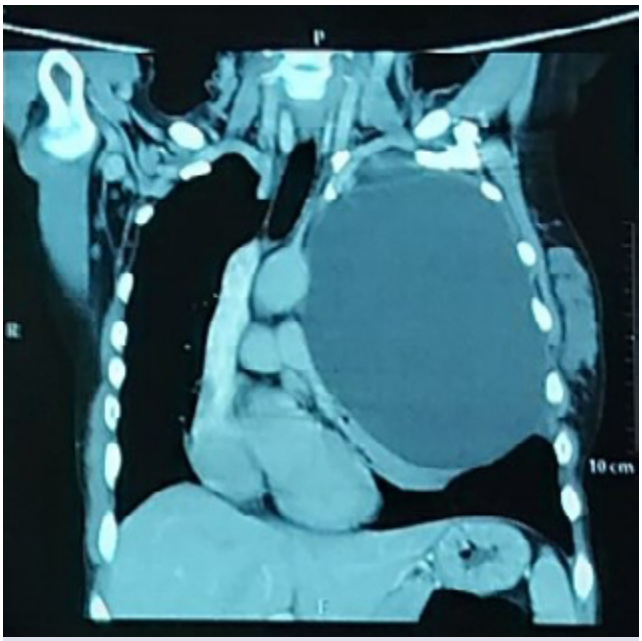

Figure 4. CECT thorax.

lated to the size, location, and rate of growth, and are caused by the mass effect on surrounding structures, such as breathing difficulties, pain, low-pressure headaches, paraparesis due to spinal cord compression, or even rupture causing hemothorax.[1] In our patient, there was a history of asthma for the past 3 years, for which she used inhalers intermittently. The milder symptoms, even in the presence of a large IM, can be explained by its slow growth over the years.

It is proposed that in NF1, the dura mater has dysplastic regions which are unable to withstand CSF pulsatile pressure alterations with respiratory and cardiac movements, leading to saccular protrusion of the meninges. Spinal deformities include scoliosis, enlargement of intervertebral foramina, and bone defects in thoracic vertebrae, and are believed to be secondary to the saccular protrusion.[3] Asymptomatic IM requires regular follow-up with imaging studies to monitor for any increase in size. Surgery should be reserved only for incapacitating symptoms and requires a multidisciplinary approach involving neurosurgery, cardiothoracic surgery, anaesthesiology, critical care, and respiratory medicine. In our case, the patient remained asymptomatic except for intermittent asthmatic complaints that responded to inhalers, and the IM was an incidental finding during the diagnosis and management of PDPH. She was the mother of a newborn baby, so she decided to avoid immediate surgery and remain in follow-up.

Literature search revealed surgical management of IM in various case reports,[4] while only one case report was found describing anaesthetic management of IM.[5] To our knowledge, this is the first case report where IM was incidentally diagnosed in association with PDPH. Though disabling for patients, PDPH is usually self-limiting even if left untreated. Our patient was a new mother whose well-being needed to be ensured early so that she could resume breastfeeding and start taking care of her newborn. Therefore, when both conservative and EBP therapy failed in the management of PDPH, we wanted to rule out any pathological cause, which was likely to be present in our patient, who was a diagnosed case of neurofibromatosis.

Among the various risk factors of PDPH, female sex, pregnancy, and the use of a dura-cutting Quincke's needle are identifiable factors in our patient. The worsening of symptoms and unresponsiveness to established treatments can be attributed to the presence of IM. After spinal blockade, the dural hole created would have caused slow seepage of cerebrospinal fluid, leading to traction over the meninges. This, in turn, would have resulted in traction of the IM, which is essentially an extension of the dura through the intervertebral foramina. Spontaneous resolution of PDPH occurred when the small dural hole closed on its own. Occasionally, persistent PDPH can lead

to complications such as chronic headache, backache, depression, cranial nerve palsies, subdural hematoma, or even seizures.[6] Neuroimaging for PDPH is indicated when it persists beyond 5 days of dural puncture, there is worsening of symptoms despite an EBP, a change in the nature of the headache, or the development of new focal neurologic symptoms.[6] This case report not only adds to the scarce literature on intrathoracic meningoceles but also prompts us to undertake neuroimaging to rule out pathological causes of headache following dural punctures, especially when the patient is a known case of neurofibromatosis.

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