
CASE REPORT

Anterior Thoracic Meningocele Presenting As Recurrent Chest Infections

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SUMMARY

Intrathoracic meningocele is rare and is usually associated with neurofibromatosis type I¹. Most of the reported thoracic meningoceles are not strictly anterior in location, but also lateral or anterolateral. We report a case of true anterior thoracic meningocele with no associated generalized mesenchymal dysplasia.

INTRODUCTION

A spinal meningocele is a herniation of the meninges through a foramina or a defect in the vertebral column and is frequently located posteriorly in the lumbosacral area. An anterior spinal meningocele is rare and is generally described in the thoracic or sacral region. These frequently occur as a manifestation of generalized mesenchymal dysplasia such as neurofibromatosis type 1 (NF-1) or Marfan syndrome and rarely as an isolated defect².

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A 4-year-old boy who was brought to neurosurgical OPD by his mother with history of recurrent chest infections since birth and was being managed at primary care level. At the age of 4 her she sought the opinion of pediatrician who advised X-ray chest (never done before) which showed a hyper dense mass along the left border of heart along with findings consistent with left basal atelectasis, most likely due to obstruction of the left main bronchus. Lateral view of the chest revealed a post mediastinal mass on the left side. This patient was then referred to thoracic surgeon who advised CT scan thoracic region which revealed a non enhancing hypo dense lesion in left post mediastinum compressing the left main bronchus and heart. This lesion was continuous with the spinal cord at the level of D5-D6 through a split corpus deformity. This child was then referred to neurosurgical department. On examination, he was found to have diminished breath sounds in the left middle and lower chest. No focal neurologic signs or clinical evidence of NF-1 or Marfan syndrome were

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Found. Laboratory findings were within normal limits. MRI thoracic region plain and with contrast was done which confirmed a cystic lesion hypo intense on T1 and hyper intense on T2 weighted sequences. There was no enhancement of the lesion on contrast administration. A provisional diagnosis of anterior thoracic meningocele was made. Surgery was planned. 200 RCC of blood was arranged. Pre anesthesia assessment was carried out. The lesion was approached through left antero-lateral thoracotomy via the 5th rib. Pleura were opened. Lungs retracted. The lesion was identified. Its neck approached and opened through a longitudinal incision in order to confirm if there was some neural tissue or not. There was no neural tissue. The cystic fluid was brownish in color. It was drained, saved for cytology and C/S. The sac wall was separated from left main bronchus and pericardium and was removed in toto. The neck was repaired in two layers. Valsalva maneuver was done at the end of repair, which confirmed no CSF leak. 24 Fr chest drain was placed via a separate incision, and kept in place for 24 hours. This child had a smooth post operative recovery, and was discharged on 8th post operative day after removal of sutures. At follow up on 4th postoperative week, the basal atelectasis on the left side had resolved on chest radiograph. The patient thereafter remained asymptomatic in regard to repeated chest infections.

DISCUSSION

A spinal meningocele is a herniation of meninges through bone defects or foramina to form a CSF-filled sac. An acquired meningocele is a comparatively common laminectomy complication, whereas congenital meningocele is a relatively rare developmental anomaly³. Although there are several theories, their embryologic origin is still unclear. More than 80% of spinal meningoceles are located posteriorly in the lumbosacral area. Anterior

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meningoceles are rarer and are described generally in the thoracic and sacral region. Most of the reported thoracic meningoceles are not strictly anterior in location, but lateral in origin, with a secondary forward projection of a dural herniation usually passing through a previously enlarged intervertebral foramen⁴. The thoracic meningocele described in our report does not pass through an enlarged foramen, but rather through a wide midline bony defect of the vertebral bodies and is therefore a true anterior thoracic meningocele.

In their review of the literature, Andrade et al found 134 cases of intrathoracic meningoceles reported before 1992.⁵ They stated that 69% of these thoracic meningoceles were in association with NF-1, whereas only 22% were isolated cases. In the same article, a total of 16 patients with thoracic meningocele between 1992 and 2003 were reported. Twelve of these 16 patients were together with NF-1 and the remaining four being isolated cases. The patient presented in this case report had no association with NF-1 or other generalized mesenchymal dysplasia and therefore is classified as being an isolated case of true anterior thoracic meningocele.

The clinical manifestations of a thoracic meningocele are closely related with its size and its relationship with surrounding structures. It may include back pain, paraparesis from insult to the spinal cord, or shortness of breath, coughing, and palpitation by compression of the lung and mediastinal structures, as was the case for the patient presented here. Even progressive hydrothorax caused by rupture of meningoceles has been reported in the literature⁶ On the other hand, in the setting of a small meningocele, no symptoms can be recorded, and the lesion may be incidentally diagnosed on a routine chest radiograph.

Although there are still conflicts concerning their treatment, it is widely accepted that excision of the lesion is indicated in the presence of progressive neurologic deficit, respiratory distress or rapid progress in the size of the meningocele.^{7,8} The decision for the surgical approach is closely related with the lesion size. For small and medium-sized meningoceles, the most common approach is through a laminectomy and an intradural repair of the cyst, with the advantage of avoiding a thoracotomy. The posterior approach is inadequate for larger lesions, as in the case presented here, and a transthoracic access is indicated because it offers a larger operating field with a small chance to damage the spinal cord.⁹ Regardless of the treatment decision, cross-sectional imaging techniques, such as CT and MR imaging are essential, not only for the diagnosis, but also for the depiction of its relationships to

surrounding structures and the exclusion of other possible accompanying lesion such as neuroma in the setting of NF-1. In the case presented here, segmentation anomalies and widening of the spinal canal caused by the thoracic meningocele was first noticed on a chest radiograph, but the final diagnosis, with its anatomic relationships, was made possible by CT and MR imaging. Other modalities of treatment like cystopleural shunts have also been described for cervicothoracic meningoceles.¹⁰

CONCLUSION

Anterior thoracic meningocele is extremely rare and even rarer if not associated with neurofibromatosis. The presentation in our child was unusual so any child with repeated chest infections needs to be thoroughly evaluated in order to identify and treat such pathologies.

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