Neglected Lumbosacral Meningocele in an Adult: A Rare Case Report

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ABSTRACT

Background: Meningocele is a neural tube defect that is usually seen in infants. Adult presentation is extremely rare, with few cases reported in the literature **Case Summary:** We report a case of a 27year old black African lady who presented to our facility with a lumbosacral swelling since birth that gradually increased in size. No associated weakness of the lower limbs, faecal or urinary incontinence. No swelling in other parts of the body. Patient and relatives attributed the late presentation to lack of initial awareness of the condition, and subsequently to lack of accessible neurosurgical care within their reach. Examination of the spine revealed a huge sessile lumbosacral swelling measuring 20 x18 x 8cm, cystic, fluctuant and brilliantly transilluminating. Neurological examination revealed normal lower limbs muscle bulk and tone. Power was 5/5 in all muscle groups with normal reflexes and intact sensations. Other systems were essentially normal. Lumbosacral MRI revealed a huge swelling in the lumbosacral region, containing cerebrospinal fluid that communicates with the spinal canal. Patient had excision of the swelling and repair of the dura. Postoperative period was uneventful. **Conclusion:** The adult presentation of meningocele is often a consequence of limited public awareness and inadequate access to neurosurgical care. Increasing public education and expanding the availability of specialized neurosurgical services could significantly reduce the incidence of such late presentations, along with their associated psychological and social burdens.

Keywords: Neglected, Meningocele, Adult

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Introduction

Meningocele is a congenital abnormality of the neural arch fusion in association with an open neural tube defect, and is characterized by protrusion of the spinal meninges containing cerebrospinal fluid without involvement of the neural tissues.[1] Majority are surgically repaired during the neonatal period, or in childhood, as survival was reported to be low without intervention.[1,2] Consequently, adult presentation is quite rare.

Here, we report the rare presentation of a lumbosacral meningocele in a 27-year-old African lady who was not operated on, in childhood.

Case Presentation

A 27-year-old black African lady who presented to the surgical outpatient department of our facility with a lumbosacral swelling that was noticed at birth. It was said to be slowly increasing in size and painless. There were no swellings in other parts of the body, associated lower limbs weakness, faecal or urinary incontinence. However, she complained of difficulty in lying supine, and reported significant psychological distress living with the condition, which led to her divorce twice. She had a miscarriage in her second marriage and was a divorcee at presentation. No family history of similar conditions or other congenital abnormalities. Patient's parents attributed the delayed presentation to ignorance and the lack of accessible neurosurgical care in their environment.

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On examination, her general condition was stable with stable vital signs. Examination of the spine revealed a huge midline sessile lumbosacral mass covered with normal skin, measuring 20 x 18 x 8cm, no differential warmth, non tender, cystic, fluctuant and brilliantly transilluminating. Neurological examination revealed normal lower limbs muscle bulk and tone. Power was 5/5 in all muscle groups with normal reflexes and intact sensations. Other systemic examination was essentially normal. Magnetic resonance imaging (MRI) of her lumbosacral spine revealed a spina bifida defect around S1/S2 with a huge protruding meningeal sac around the lumbosacral region containing cerebrospinal fluid (CSF) without neural tissue involvement (Fig.1) which confirmed the diagnosis of a meningocele. Other baseline investigations were normal. Patient underwent elective excision and repair under general anesthesia. Intraoperative findings revealed a huge meningeal sac containing clear CSF, which was dissected down to its base. Dura was opened and CSF drained. Redundant and thickened dura was excised to prevent recollection of CSF, remnant closed in a watertight fashion using Vicryl 3/0. Wound closed in layers (Fig 2). Patient had an uneventful postoperative period and was discharged home after stich removal. At follow up period, she expressed satisfactory cosmetic appearance, resolution of her psychological distress and can comfortably lie supine.

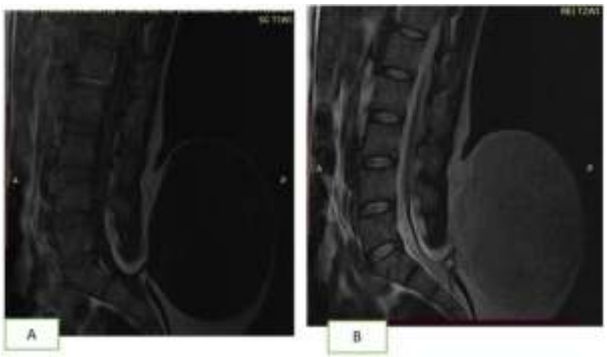
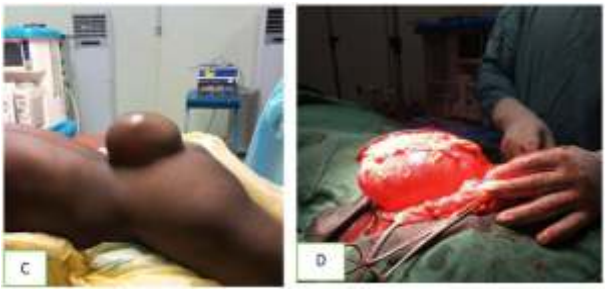


Figure 1: Sagittal T1WI (A) and T2WI (B) showing a spina bifida at S1/S2 and a large hypointense and hyperintense lumbosacral meningeal sac communicating with the spinal canal respectively.



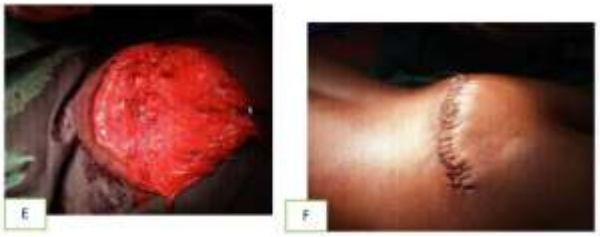


Figure 2: Intraoperative images show a huge sessile lumbosacral swelling (C), a huge dural sac containing cerebrospinal fluid (D), excised redundant dural sac and watertight closure of the residual dura (E), and soft tissue and skin closures (F).

Discussion

Neural tube defects (NTDs) constitute the second most common type of congenital abnormality after congenital heart defects.[2] They occur in the spinal region where they are termed spinal dysraphism, or in the cranium as cranial dysraphism. Spinal dysraphism can be broadly classified into open or closed, depending on whether the lesion is covered by skin.[2] Meningocele is a form of open neural tube defect characterized by a sac of meninges, covered by skin and containing cerebrospinal fluid (CSF) alone, herniates through an anterior or posterior bony defect.[3] These lesions, though skin-covered, are considered open neural tube defects as they result from a failure of the neural tube to close completely during fetal development. The majority of cases of meningoceles are diagnosed and treated perinatally, as such, cases are rarely diagnosed in adulthood.[1,2]

Most spinal meningoceles showed female preponderance and are frequently located posteriorly over the lumbar and sacral regions,[4,5] which is consistent with the findings in the index case. Our patient was a female that presented at the age of 27 years, similar to the findings in the literature.[2,6] However, our findings were contrary to those of other authors who reported a male predominance, characterized by a very late presentation at the ages of 53, 60 and 67 years, respectively.[1,7,8] The gender difference in adult presentation of meningocele could be coincidental, as the condition has generally been reported to be more common in females.[4,5] However, females tend to present early, probably due to their greater concern about cosmesis than male patients, who do not present until when complications have occurred. Our patient was asymptomatic but presented due to cosmetic concerns, as the swelling led to her being divorced twice.

Spinal meningoceles often cause neurological deficits, skeletal deformities, anorectal and urinary bladder dysfunctions, paraplegia, and sensory loss below the cord level of involvement.[1,2] The index case had no neurological deficits, similar to the findings of other authors.[7,8] In meningocele cases, neurological involvement is not seen as often as in myelomeningocele lesions, but the local signs of sacral nerve root involvement present as pain in both legs and bladder dysfunction.[6] As such, somatosensory evoked potentials (SSEP) can be used in these patients as in myelomeningoceles.[9,10] The equipment for SSEP is not available in our centre. Symptoms may be precipitated by direct trauma to the lumbosacral region causing deformation of the marginally functioning neural elements within the stretched cord.[11] Other precipitants include, the bending movements, lithotomy position during childbirth, and movements occurring during motor accidents.[11] Our patient had no previous history of trauma, and remained nulliparous despite two marriages.

Complications of late presentation include local infection, ulceration, CSF leakage and possible squamous metaplasia due to chronic mechanical irritation and bacterial infection.[2,6,7] Our patient presented with a large lumbosacral swelling measuring approximately 20 x18 x 8 cm, covered with normal skin, in contrast to cases reported in the literature, which were smaller in size but presented with complications.[2,7] The greater resilience of the black skin compared to other races could possibly explain why our patient did not present with any of the aforementioned complications despite the large size at presentation.

Magnetic resonance imaging (MRI) of the spine in the sagittal plane is the investigation of choice for evaluating the meningocele sac, and to observe the spinal cord itself, as well as the possible congenital anomalies associated with it.[6] In the index case, it revealed a spina bifida at S1/S2 and a huge hypointense and hyperintense lumbosacral meningeal sac that communicates with the spinal canal on T1WI and T2WI respectively, which agrees with the findings of other authors.[6,7]

Adult presentation of meningocele may occur due to a patient's parent or caregiver refusal of surgery during childhood,[6,7] or may present for the first time in adulthood.[2] Our patient presented for the first time in adulthood, similar to the finding of Wahid *et al*.[2] The patient's parents attributed the delayed presentation to a lack of awareness about the condition and limited access to neurosurgical care in their locality. This emphasizes the need for greater public health awareness about neural tube defects and their prevention, as well as the need to establish more accessible neurosurgical care centres in our community.

Early surgical intervention is recommended in meningocele cases,[2] for cosmesis and to prevent infection and neurological deficits.[12] Our patient had elective excision and repair of the dura. Intraoperative findings were that of a huge meningeal sac (fig 2) containing clear CSF with no neural tissues. The sac was dissected down to its base and the redundant dura was excised to minimize the pouch for good cosmetic result. The remnant dura was closed in a watertight fashion to prevent CSF leakage using Vicryl 3/0. Our technique was similar to that described by Ozdemir *et al*, [6] although no fibrin adhesive tissue sealant was used to reinforce the dural tissues at the site of dural repair, as the sealant is not readily available at our facility. The patient had unremarkable postoperative period with no complications at 6 months follow up period. She also acknowledged good cosmetic outcome at the operative site.

Conclusion

Adult presentations of meningocele may result from a combination of low public awareness and insufficient access to neurosurgical care. Addressing these gaps through targeted health education and the development of additional neurosurgical centres could help prevent delayed diagnoses and mitigate the long-term psychological impact on affected individuals.

Conflict of Interest

None

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