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Case Report

CASE REPORT

Pyomyelia presenting as acute flaccid paralysis

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Abstract

We report an unusual case of a 10-month-old girl with intramedullary spinal cord abscess who presented with fever and acute flaccid paraplegia. Nerve conduction study showed demyelinating neuropathy after which she received intravenous immunoglobulin therapy. This was followed by ascending paralysis and left-sided ptosis. Lumbar puncture revealed purulent cerebrospinal fluid and magnetic resonance imaging (MRI) with gadolinium showed intramedullary holocord abscess with a dermal sinus tract extending from skin to intramedullary canal in the lumbosacral region. This tract was excised completely along with drainage of pus. She was treated with broad-spectrum antibiotics for 6 weeks and underwent neurorehabilitation after which there was significant neurological improvement. Follow-up MRI shows good resolution of the intramedullary abscess.

INTRODUCTION

Intramedullary spinal cord abscess (ISCA) is a well-recognized and rare entity. It was first described by Hart [1]. Limited data are available in the literature on intramedullary abscess involving the whole length of the spinal cord (holocord abscess) in children [2–5]. It is very important to remember this entity as it is a curable condition if detected and treated early. We report a case of a 10-month-old girl presenting with acute flaccid paralysis who was later diagnosed to have holocord abscess.

CASE REPORT

A 10-month-old girl was referred to our service with fever for 1 month and bilateral lower limb weakness with urinary retention since the previous day. The referring hospital had treated her with intravenous antibiotics (ceftriaxone) for 5 days. She had normal developmental milestones. Examination showed flaccid paraplegia with Grade '0' power and absent deep tendon reflexes. Nerve conduction velocity (NCV) test showed absence of F waves and low NCV in the tibial nerve, suggesting demyelinating neuropathy. With a provisional diagnosis of Guillain–Barre syndrome (GBS), she was treated with intravenous immunoglobulin (IVIG). In spite of the treatment, there was rapid progressive weakness with involvement of bilateral upper limbs, shallow respiration, weak gag reflex and left-sided ptosis. Lumbar puncture was done, which shows purulent cerebrospinal fluid (CSF) on gross examination, and gram stain smear showed uncountable pus cells per high power field (precise measurement of pus cell per high power field is not available) along with gram positive cocci. Biochemical analysis of CSF showed protein of 317 mg/dl and sugar of 3 mg/dl with blood glucose of 86 mg/dl. CSF Latex agglutination test was negative for *Escherichia coli*, *Streptococcus agalactiae*, *Haemophilus influenzae*, *Neisseria meningitidis* and *Streptococcus pneumonia*. Blood and CSF culture did not show any growth of organisms as the child had received empirical intravenous ceftriaxone before lumbar puncture and CSF examination. Magnetic resonance imaging (MRI) with gadolinium revealed features of myelitis with hydromyelia, meningitis and empyema involving whole cord (holocord abscess) with arachnoiditis. A dermal sinus tract was seen extending from skin to intramedullary canal in lumbosacral region (Fig. 1). Careful examination of back showed a small opening of the dermal sinus in lower lumbar region without any pus discharge. The child underwent L4–L5

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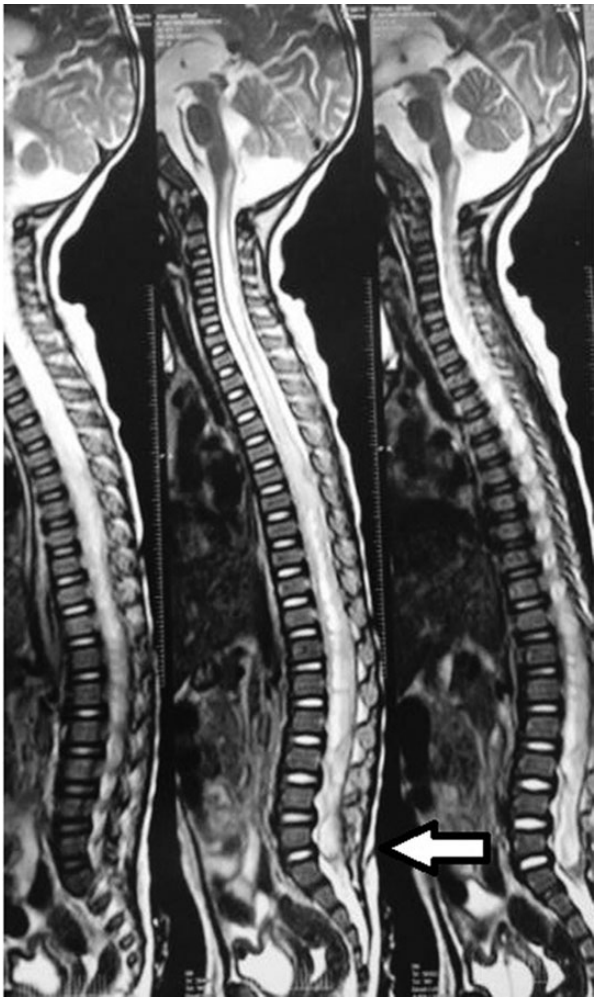

Figure 1: Preoperative MRI with gadolinium (T2-weighted sagittal section) showing holocord abscess and dermal sinus tract (shown with arrow).

laminotomy and complete excision of dermal sinus along with drainage of pyomyelia (Fig. 2). She was treated with vancomycin and meropenem for 6 weeks and underwent neuro-rehabilitation. She started showing neurological improvement within a few days in the form of some movement in both the legs and regained normal tone, power and reflexes by the end of antibiotic treatment. Follow-up MRI shows good resolution of the intramedullary abscess.

DISCUSSION

Children who present with acute flaccid paralysis are often commonly labeled as poliomyelitis, GBS, transverse myelities or traumatic neuritis. We had sent the patient's stool sample as part of acute flaccid paralysis surveillance which did not reveal polio virus. NCV was indicative of GBS after which we did lumbar puncture to look for albumino-cytological dissociation. The presence of pus in CSF was a surprise as the child did not have signs of meningeal irritation nor bulging fontanel. MRI revealed the intramedullary holocord abscess and a dermal sinus tract. The opening of the tract on the skin was very small and found only after seeing with a magnifying glass and hence was missed on routine clinical examination initially.

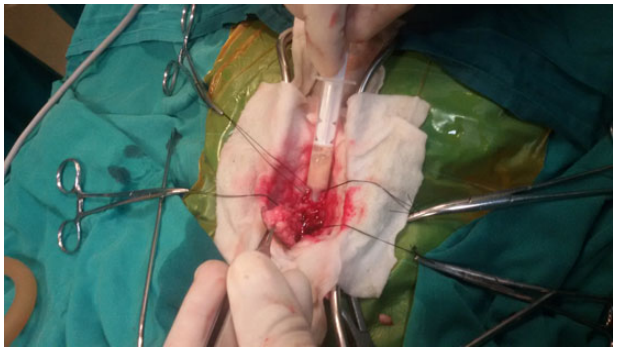

Figure 2: Intraoperative photograph showing aspiration of abscess from spinal canal.

Congenital dermal sinus (CDS) belongs to a group of disorder called occult spinal dysraphism in which skin covers most of the defect of the neuraxis and overlying tissues, except for a small skin ostium of the dermal sinus tract. It is due to imperfect separation between cutaneous and neural ectoderm in the early fetal life [6]. Some patients with the anomaly can develop severe infection of the central nervous system, such as bacterial meningitis and spinal cord abscesses, through the dermal sinus tract [7]. If the sinus stops short of the dura, then subcutaneous abscesses may occur. Meningitis generally occurs in early childhood and is often recurrent. Thoraco-lumbar segments and mid-thoracic segments of the spinal cord are the most common areas involved in ISCA. The mean range of longitudinal spread of the infection in the spinal cord is usually three to six levels. Thus, involvement of the entire cord as seen in our case is a rare and severe complication of CDS [7]. CDS is found to be the origin of infection in approximately half of affected children with ISCA [8]. Other sources of ISCA are hematogenous from infections of the lungs, genitourinary tract or endocarditis or through penetrating injuries. *Staphylococcus* and *Streptococcus* are the common infecting organisms; however, a causative organism may not be found in 38% cases. Even with a marked reduction in the mortality rate in the post antimicrobial era, this infection may result in a considerable devastating mechanico-vascular insult of the spinal cord from formation of the abscess and an expansion of the spinal cord within the limited intraspinal space with a resultant disappointing neurological outcome. Still, early diagnosis and appropriate surgical intervention with administration of wide-spectrum antibiotics are the main stay to prevent neurological disability and to improve the functional outcome [8, 9].

CDS has to be identified in all newborn to prevent contiguous spread of infection to the spinal cord. This has to be differentiated from a benign clinical entity called sacrococcygeal dimple which are innocuous small lesions (<5 mm in width) located just above the coccygeal area within 2.5 cm from anus. All other atypical dimples located higher on the back warrant further evaluation. Ultrasonography is the screening modality of choice for the infant with atypical sacral dimples. The presence of an intraspinal mass, abnormal position or shape of the conus, or a thick filum necessitates MRI study. MRI has become the reference study technique because of its ability to accurately depict the extent of the sinus tract and associated lesions [10].

In conclusion, lumbar puncture and a careful back examination are essential in all children presenting with acute flaccid paralysis and fever so as to differentiate ISCA from other causes of acute flaccid paralysis since a timely surgical and medical management will prevent devastating neurological deficits.

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AUTHORS' CONTRIBUTION

S.S. and R.J. were involved in conceptualization of the manuscript, collecting patient data, conducting literature search and drafting the manuscript. This was critically reviewed and approved by R.J. Both authors were involved in clinical management of the patient.

CONFLICT OF INTEREST STATEMENT

None declared.

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ETHICAL APPROVAL

Ethical approval was taken from Hospital Ethics Committee.

CONSENT

Written informed consent was obtained from the parents of the patient for publication of this case report and accompanying images.

GUARANTOR

R.J. will act as the guarantor of the manuscript.

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