Unusual presentation of more common disease/injury

Tuberculous radiculomyelitis presenting as urinary retention in a child with Down's syndrome

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Correspondence to Dr Arani Sridhar, arani.sridhar@uhl-tr.nhs.uk**Summary**

Tuberculous radiculomyelitis (TBRM) is an uncommon complication of TB meningitis. The authors report the case of a 10-year-old Asian girl with trisomy 21, who presented with acute urinary retention and fever. She was initially treated for a urinary tract infection. After an acute neurological deterioration she was found to have evidence of TB meningitis with TBRM. She developed acute hydrocephalus requiring ventriculo-peritoneal shunt. She was treated with quadruple antituberculous therapy and high dose intravenous dexamethasone. She needed tracheostomy with continuous positive airway pressure (CPAP) support. Although she showed gradual neurological improvement in her cognitive functions, she persisted to have quadriparesis with the need for tracheostomy and CPAP support overnight and gastrostomy feeding. Acute urinary retention in children is uncommon, and should serve as a 'red flag' to consideration of further underlying neurological problems. This presentation and subsequent events should serve as a learning point to clinicians.

BACKGROUND

Tuberculous meningitis (TBM) is the commonest cause of chronic meningitis worldwide. Though the prevalence of TBM is consistently high in developing countries, where the disease continues to have a high death rate among infants and children, it must always remain a consideration in developed countries.¹ Early diagnosis and prompt initiation of treatment are essential to improve the otherwise poor outcome.

Neurological sequelae such as cranial nerve palsy, motor deficit, cognitive impairment and seizures occur frequently in TBM, as a consequence of local infiltration, inflammation and infarction, tuberculoma formation and development of hydrocephalus. TB radiculomyelitis (TBRM) is a relatively rare complication of TBM which causes the neurological sequelae through local infiltration and adhesive arachnoiditis of spinal nerve roots.

CASE PRESENTATION

An 11-year-old Asian girl with trisomy 21 presented with a 3-week history of dysuria, low-grade fever, constipation, abdominal pain and being generally unwell with lethargy and irritability.

Antibiotics for a presumed urinary tract infection were initiated before admission to our hospital. On further questioning, it transpired that she had been frequently visiting the toilet in the last few days, and had been squatting in the bath. She had not opened her bowels for 3 weeks. Her bowel habits were regular before this illness. Her family were also concerned that she had become unsteady on her feet and lacked energy. She had a low-grade fever subsiding with the use of paracetamol. There was no history of sensory symptoms, backache, headache, vomiting, altered sensorium, chronic cough, lymphadenopathy and night sweats. There was no history of contact with a known case of tuberculosis. There was no history of any recent foreign travel. In her medical history she was known to have

trisomy 21, with bilateral cuneiform cataracts, and moderate learning difficulties. She had no cardiac problems. She attended a special school, and could walk unaided, talk and was socially interactive. She had no BCG scar.

On admission, she was miserable, irritable and reluctant to walk. She was afebrile and alert, with Glasgow coma score 15. She had no signs of meningitis. On neurological examination, she had a normal power and slightly exaggerated reflexes in her lower limbs with equivocal plantar response. On abdominal examination, she had a distended palpable bladder with faecal mass in the left iliac fossa. Warm bathing was attempted but proved unsuccessful in relieving the urinary retention, so she was catheterised and antibiotics were started.

Her urinary retention was initially thought to be due to constipation and hence she did not undergo further neurological assessment or neurological imaging.

Initial blood investigations showed a white cell count of $3.5 \times 10^9/l$. The lymphocyte count was low $0.7 \times 10^9/l$. The other routine blood tests including urea, creatinine, electrolytes, liver function tests, bone profile, clotting screen were normal. The C reactive protein was elevated at 64. Urine culture showed a growth of *E. coli*. Lymphocyte phenotyping later showed moderate T cell lymphopenia affecting both CD4+ and CD8+ T cells with normal CD4:CD8 ratio. There was no evidence of immunoglobulin deficiency.

However, 2 days into her inpatient stay she suddenly deteriorated neurologically. Her conscious level deteriorated, she stopped speaking and she was only responsive to painful stimuli with abnormal pupillary responses. She was quadriplegic with hypotonia, absent deep tendon reflexes and bilateral extensor plantar response. A detailed sensory examination was not carried out as she was sedated for ventilation.

Following intubation and ventilation an urgent MRI scan of her brain (figure 1) showed hydrocephalus with meningeal enhancement. The MRI of spine (figure 2) showed an

Figure 1 T1 axial brain postgadolinium image shows hydrocephalus with meningeal enhancement extending deep into the sulci.

extensive enhancing dural thickening with debris in the lumbar dural sac causing lack of definition of the conus. She was transferred to neurosurgical unit where an external ventricular drain was placed. The cerebrospinal fluid (CSF) was seen to be under pressure and showed reduced glucose 0.5, elevated protein 1.5, white blood cell (WBC) 1, red blood cell (RBC) 334. CSF culture was positive for *Mycobacterium tuberculosis*. The T-spot test for tuberculosis was positive. Mantoux test was strongly positive. Chest x-ray showed radiological evidence of pulmonary tuberculosis. Gastric lavage was positive for *M tuberculosis*. Fundal examination showed papilloedema with no choroid tubercles. She was started on quadruple therapy for tuberculosis meningitis along with high-dose intravenous dexamethasone.

Although she has continued to show gradual improvement over time her neurological status remains hugely impaired. She remained quadriparetic with some limited movements in her hands. She needed tracheostomy with continuous positive airway pressure (CPAP) support over night. She showed some improvement in her cognitive functions. She was fed through a nasogastric tube initially followed by gastrostomy feeding.

INVESTIGATIONS

MRI scan of her brain (figure 1) showed hydrocephalus with meningeal enhancement. The MRI of spine (figure 2) showed an extensive enhancing dural thickening with debris in the lumbar dural sac causing lack of definition of the conus. A repeat MRI scan of brain and spine after 12 months of antituberculous therapy showed significant amount of meningeal, basal cistern and dural exudative dis-

Figure 2 T1 sagittal postgadolinium image of the thoraco lumbar spine shows extensive enhancing dural thickening with debris in the lumbar dural sac causing lack of definition of the conus.

ease. The MRI spine showed significant cord compression at C1 with generalise cord atrophy (figure 3).

The CSF was seen to be under pressure and showed reduced glucose 0.5, elevated protein 1.5, WBC 1, RBC 334. CSF culture was positive for *M tuberculosis*. The T-spot test for tuberculosis was positive. Bone marrow examination was normal. Gastric lavage was positive for *M tuberculosis*. Fundal examination showed papilloedema with no choroid tubercles. Mantoux test was strongly positive. Chest x-ray showed radiological evidence of pulmonary tuberculosis. Lymphocyte phenotyping showed moderate T cell lymphopenia affecting both CD4+ and CD8+ T cells with normal CD4:CD8 ratio. There was no evidence of immunoglobulin deficiency.

Figure 3 T1 sagittal postgadolinium image of the brain including cervical spinal cord showing nodular enhancement of basal and spinal meninges compressing the cord at C1 level.

DIFFERENTIAL DIAGNOSIS

Includes a number conditions presenting with neurological manifestations such as partially treated bacterial meningitis, fungal meningitis, transverse myelitis, encephalomyelitis, acute demyelinating encephalomyelitis.

TREATMENT

She was started on quadruple antituberculous therapy including isoniazid, rifampicin, pyrazinamide and ethambutol for 12 months along with 6 weeks of high-dose intravenous dexamethasone followed by gradual weaning over few weeks.

OUTCOME AND FOLLOW-UP

Although she showed gradual neurological improvement in her cognitive functions, she persisted to have quadriplegia with the need for tracheostomy and CPAP support overnight and gastrostomy feeding.

DISCUSSION

TBM is the commonest cause of chronic meningitis worldwide. Though the prevalence of TBM is consistently high in developing countries, where the disease continues to have a high death rate among infants and children, it must always remain a consideration in developed countries.¹ Early diagnosis and prompt initiation of treatment are essential to improve the otherwise poor outcome.

TBM occurs with a variety of non-specific clinical manifestations. In children, apathy, lack of interest in play, irritability, restlessness at night, minor headaches, loss of appetite, nausea, vomiting and abdominal pain are the usual presenting symptoms.

Neurological sequelae such as cranial nerve palsy, motor deficit, cognitive impairment and seizures occur frequently

in TBM, as a consequence of local infiltration, inflammation and infarction, tuberculoma formation and development of hydrocephalus. A retrospective study of TBM reported neurological sequelae in 27% of cases, whereas a prospective study of 65 survivors noted neurological sequelae at in 78.5% cases at 1 year.^{1 2}

TBRM is a relatively rare complication of TBM which contributes to the above neurological sequelae through local infiltration and adhesive arachnoiditis of spinal nerve roots. In a review of 74 previously reported cases of TBRM, the commonest symptoms were subacute paraparesis, radicular pain, bladder disturbance and subsequent paralysis.³⁻⁵ Retrospectively all these features were present in our case. In our reported case initially the urinary retention was thought to be due to constipation and hence detailed neurological assessment and imaging was not carried out at presentation. However, we suspect that the clinical presentation of leg pains, unsteady gait and bladder disturbance is likely to be due to tuberculous lumbo-sacral radiculitis.

Secondary radiculo-myelitis may appear either during the acute stage as in our case or after variable periods following the onset of TBM. Spinal extension of basal meningitis usually develops within few weeks of starting inadequate antituberculous therapy and in some reported cases TBRM can develop during appropriate treatment of intracranial TB.^{3 5}

Diagnosis of TBM in the literature is most often based on clinical, CT head and CSF criteria.¹ More recently, positive PCR for *M tuberculosis* or IgM ELISA are also considered definitive evidence of cranial tuberculosis. In our case, although CSF protein was elevated and glucose was reduced, there was no lymphocytosis. This could be due to the moderate T cell lymphopenia. Children with trisomy 21 are known to have altered immune system with lymphopenia.

The T-spot test, performed in our case, is an invitro T cell measurement assay used for diagnosing tuberculosis disease and latent infection. Unlike the traditional tuberculin skin test, the T-spot incorporates a positive control, allowing the user to distinguish between a genuine negative result and one which is indeterminate through operator error.

MRI and CT are critical for diagnosis of tubercular radiculomyelitis, revealing loculation and obliteration of the subarachnoid space along with linear intradural enhancement. The CT features include irregular subarachnoid contrast medium, extradural and intradural filling defects with the spinal cord of variable dimension. Adherence of the nerve roots to one another and to the meninges is not an uncommon finding. Given the right clinical setting, CT myelographic features are reasonably characteristic to suggest a diagnosis of TBRM.⁶

Patients with active meningeal tuberculosis are offered a treatment regimen initially lasting 12 months, comprising isoniazid, pyrazinamide, rifampicin and a fourth drug (in this case, ethambutol) for the first 2 months, followed by isoniazid and rifampicin for the rest of the treatment period. In addition, glucocorticoids are administered and gradually weaned.⁷ Enhanced resolution of the basal exudate and improved survival rates have been shown to be associated with the use of corticosteroids.⁸

Learning points

- ▶ Acute urinary retention in children is uncommon, and should serve as a 'red flag' to consideration of further underlying neurological problems and appropriate neurological investigations.
- ▶ Though central nervous system tuberculosis infection remains relatively uncommon in the developed world, this presentation and subsequent events should serve as a learning point to clinicians to consider a wider differential diagnosis when faced with acute neurological presentations.
- ▶ This case also demonstrates the atypical CSF cellular findings due to lymphopenia which can be a feature in children with trisomy 21.

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Competing interests None.

Patient consent Obtained.

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