Generalised hypotonia following erosive vertebral osteomyelitis in an infant

Neonatal cervical osteomyelitis is an extremely rare condition. It is a potentially dangerous condition with high fatality and disastrous consequences. Hence, it is important to recognise it early to prevent long-term morbidity and sensory-motor disabilities. It can have an indolent course conspicuous with the absence of pyrexia. We report a four-week-old infant with erosive osteomyelitis involving C1 and C2 and retropharyngeal abscess resulting in neurological abnormalities.

Keywords
hypotonia; osteomyelitis; neonate; cervical spine

Key points
Banerjee J., Bhojani S., Nerminathan V.
1. Neonatal cervical osteomyelitis is a rare condition, which can have high morbidity and mortality.
2. Osteomyelitis should be considered in any infant presenting with neurological signs, even in the absence of pyrexia.
3. Early clinical suspicion, prompt radiological investigations and broad-spectrum antibiotic cover can prevent disastrous consequences, while delayed diagnosis can lead to permanent paralysis and death.

Case report
A four-week-old male infant with no significant antenatal or peri-natal problems presented with ‘floppiness’ and difficulty in feeding. He was born by normal delivery with Apgar scores of 9 at 1 minute and 10 at 5 minutes. Pregnancy was uneventful and there were no risk factors or clinical features of maternal or neonatal sepsis.

According to parents he had been well since birth, establishing feeds and had normal movement of all four limbs until two to three weeks of age. In the following week, he was noted to feed poorly and there were reduced spontaneous movements in the right arm, prompting parents to seek medical attention. The paediatrician who saw the infant at that stage diagnosed it to be birth-related right-sided Erb’s palsy. When reviewed a week later, he was unable to suck or swallow feeds from a bottle, prompting admission to the paediatric ward.

On admission, he was afebrile and pale. There was a small soft lump (1.5 cm x 1.5 cm) localised over the right side of his rib cage (7th-8th rib). Neurological examination revealed reduced tone in all four limbs with normal trunkal tone. Bilaterally, his arms were held in an internally rotated position with flexion of the wrists and his lower limbs were abducted in a frog-like position. There was an absence of spontaneous movements and reflexes in all four limbs.

Initial blood tests showed: white cell count 33 x 10^9/L (neutrophils 26 x 10^9/L and lymphocytes 4 x 10^9/L), platelets 737 x 10^9/L and C reactive protein of 301 mg/L. Urgent cranial and spinal MRI scans revealed a large retropharyngeal multicystic lesion encasing the anterior arch of the C1, consistent with multi-loculated abscess with phlegmon (FIGURE 1). C1 was subluxated with marked narrowing at the cranio-vertebral junction and effacement of the CSF showing evidence of cord compression. In addition there was a high intensity linear structure in the vicinity of the straight sinus suggesting straight sinus thrombosis.

Subsequently, the infant was transferred to the tertiary neurosurgical unit for further management. The retropharyngeal abscess was incised and drained via pharyngeal route by the ENT and spinal surgeons.

Blood cultures showed no growth after five days. Pus culture grew Staphylococcus...
Infants can present with a localised swelling, retropharyngeal abscess or cellulitis. The presenting complaint can include respiratory distress, stridor, irritability and persistent crying. There have been four cases of cervical osteomyelitis in neonates reported in the literature who presented with unilateral/bilateral upper limb paralysis or Erb’s palsy around three weeks of age. The clinical course can be completely indolent as in our case where there was no fever; faltering demonstrated that there were two potential routes for the organisms to reach the spine via haematogenous route: the nutrient arteries and the paravertebral venous system. In 1979, Ogden provided definite histological support for an initial metaphyseal focus in human neonatal osteomyelitis.

Pathophysiology and microbiology
Vertebral plexus of the veins and the inter-metaphyseal communicating artery has an important role in bacterial colonisation and subsequent vertebral osteomyelitis. Osteomyelitis of the cervical spine can lead to neurological deficits due to collapse, fracture dislocation or subluxation of the vertebrae and development of extradural abscess involving the spinal cord or nerve root.

Staphylococcus aureus is the most commonly isolated organism of osteomyelitis. Another organism commonly implicated in neonatal osteomyelitis is Group B beta-haemolytic Streptococcus. Before the introduction of antibiotics, acute haematogenous osteomyelitis had a mortality of approximately 20%, while long-term morbidity occurred in 50% of the cases.

Vertebral osteomyelitis can be the result of spread from the adjoining structures such as the pharynx, or may be due to haematogenous spread which can result from a variety of infections including arthritis, osteomyelitis of long bones, sinusitis, endocarditis, respiratory tract infections and skin infections, eg paronychia. However, in up to 70% of the cases no primary infective focus is identified. The main sites of vertebral osteomyelitis are the lower thoracic and lumbar spine. Vertebral spine involvement is rare, more so in the neonatal age group, where only seven cases have been reported in the literature (TABLE 1).

Clinical presentation
Infants can present with a localised swelling, retropharyngeal abscess or cellulitis. The presenting complaint can include respiratory distress, stridor, irritability and persistent crying. There have been four cases of cervical osteomyelitis in neonates reported in the literature who presented with unilateral/bilateral upper limb paralysis or Erb’s palsy around three weeks of age. The clinical course can be completely indolent as in our case where there was no fever; faltering...
growth, poor feeding and neurological signs were the presenting features, which pointed towards a diagnosis of infection of the spine.

Radiology
Plain lateral radiographs of the cervical spine can be a useful initial investigation, which may reveal vertebral destruction, collapse of vertebrae, loss of bony trabeculation or a paraspinal soft tissue mass\(^1\). CT scans are more sensitive than plain X-rays and can offer useful additional information regarding the extent of the infective lesion. Gadolinium-enhanced MRI scans are particularly useful for detecting spinal cord involvement and extradural abscesses\(^2\). Radionuclide bone scans are sensitive and have been reported to detect up to 90% of pyogenic vertebral osteomyelitis. However, initial bone scans can be unremarkable as illustrated by the case described by Martijn et al\(^8\).

Management
The mainstay of management of cervical spine osteomyelitis is broad spectrum antibiotics. The majority of cases in the literature have preferred a 10-12 week course which we feel could be decided individually in discussion with the microbiologist.

Surgical intervention in vertebral osteomyelitis is warranted in:
- wide-spread osseous involvement
- clinical sepsis which does not respond to antibiotics
- progressive neurological deficit due to spinal cord compression\(^2\), as seen in our patient.

One crucial aspect in the management of cervical osteomyelitis is the stabilisation of the spine, which can be achieved by a brace. This is followed by gentle mobilisation with external bracing using halo braces, moulded spinal jackets or spinal intervention\(^7\). Physiotherapist and occupational therapist should be involved to aid with mobilisation.

The long-term neurological prognosis depends on the degree and duration of spinal cord compression and hence remains largely unpredictable.

### Table 1

<table>
<thead>
<tr>
<th>Author</th>
<th>Age at presentation</th>
<th>Organism isolated and probable primary site of infection</th>
<th>Presenting complaints and MRI scan findings</th>
<th>Intervention</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Ram et al(^1) 2002</td>
<td>4 weeks</td>
<td>Methicillin resistant <em>Staph aureus</em>, Paronychia of right fifth toe</td>
<td>Stridor and bilateral upper limb weakness. MRI – retropharyngeal abscess C3-C4 region, extending posteriorly into epidural space and spinal cord moderately displaced, partial destruction of the body of C3</td>
<td>Neck stabilisation, 48 hours of ventilation, 12 weeks antibiotics with vancomycin and cefotaxime</td>
<td>Follow up MRI – gradual healing of infected area; after 12 weeks of antibiotics, patient was able to lift both arms above shoulders</td>
</tr>
<tr>
<td>Sharma et al(^2) 2000</td>
<td>6 weeks</td>
<td><em>Staph aureus</em>, No obvious infective focus</td>
<td>Weakness of left shoulder and elbow, 5 x 5 cm swelling in posterior triangle of the neck. CT showed destruction of C6, mild compression of thecal sac at C6 level</td>
<td>11 weeks of antibiotics, Incision and drainage of abscess</td>
<td>6 months follow-up – full neurological recovery and new bone formation at the site of vertebral destruction on CT scan</td>
</tr>
<tr>
<td>Barton et al(^3) 1996</td>
<td>1 month</td>
<td><em>Group B Streptococcus</em>, Primary focus unknown</td>
<td>Feeding difficulty, posterior thoracic mass. MRI showed a 4 x 4 cm left paraspinal mass at T8-9, eroded vertebral bodies with wedge compression</td>
<td>Broad spectrum antibiotics, surgical debridement at 3 months of age</td>
<td>One year later the child remained on external body brace with good lower extremity motion and learning to stand</td>
</tr>
<tr>
<td>Ein et al(^4) 1988</td>
<td>3 weeks</td>
<td><em>Staph aureus</em>, Septic arthritis</td>
<td>Cyanotic spell requiring ventilation. CT scan showed a mediastinal mass extending from C7 to the carina, possible neuroenteric cyst</td>
<td>Antibiotics, incision and drainage of abscess</td>
<td>Walking since 2 years of age</td>
</tr>
<tr>
<td>Martijn et al(^5) 1992</td>
<td>3 weeks</td>
<td><em>Staph aureus</em>, Paronychia of both hands</td>
<td>1 week history of decreased movement of arms and legs, absent reflexes. MRI showed involvement of C2-4, prevertebral mass compressing and displacing the spinal cord posteriorly</td>
<td>Intravenous antibiotics</td>
<td>Recovery of neurological symptoms and MRI showing decreasing abscess size and residual changes in the body of C2-4</td>
</tr>
<tr>
<td>Ammari et al(^6) 1992</td>
<td>3 weeks</td>
<td><em>Group B Streptococcus</em> Probable maternal spread</td>
<td>Floppiness at birth progressing to reduced movement of upper extremities. MRI – mass at C5-6 with anterior collapse, kyphosis C6</td>
<td>Antibiotics for 28 days, surgical debridement</td>
<td>Discharged at 7 weeks of age with normal Moro reflex, upper limb power 4/5 bilaterally, improved grasp</td>
</tr>
<tr>
<td>Colville et al(^7) 1975</td>
<td>3 weeks</td>
<td><em>Staph aureus</em> Osteomyelitis of left femur</td>
<td>Bilateral shoulder and upper limb paralysis, swelling of left lower thigh and femur. X-ray – widening of the paravertebral space with destruction of C5</td>
<td>Antibiotics, immobilisation of the cervical spine and lower limbs, blood transfusion</td>
<td>Severe sepsis, bacterial endocarditis, cardiac failure leading to death</td>
</tr>
</tbody>
</table>

Published cases of vertebral osteomyelitis in the neonatal age group (includes cases published in English only).
Conclusion
In any infant presenting with neurological signs viz unilateral or bilateral upper limb paralysis, stridor, irritability and difficulty in feeding, cervical spine lesions including osteomyelitis should be considered, even in the absence of pyrexia. Early clinical suspicion, prompt radiological investigations and broad-spectrum antibiotic cover can prevent disastrous consequences while delayed diagnosis can lead to permanent paralysis and death.

Acknowledgement: The authors would like to thank Dr Marc T ebruegge, Honorary Clinical Research Fellow, Infectious Diseases Unit, The Royal Children’s Hospital Melbourne and Department of Paediatrics, The University of Melbourne Postgraduate Scholar, Murdoch Children’s Research Institute Fellow, for his help and encouragement while writing this paper.

References

SAFESET™ is a closed, needle-free system that protects both you and your patients
• Protects against exposure to bloodborne pathogens and IV line contamination
• Reduces blood waste and therefore may help reduce the risk of iatrogenic anaemia
• May increase the efficiency and safety of the blood-sampling process
• Available integrated with the Transpac® IV Pressure Monitoring Kit or as a stand-alone system
• Adult and neonatal/paediatric
• Various single-, double-, and triple-line options

Advancing safety for patients and providers

SAFESET™
BLOOD SAMPLING SYSTEM

Contact us for information or ordering
Fannin UK Ltd.
42-46 Booth Drive
Park Farm South
Wellingborough
NN8 6G7
Tel: +44 (0) 118 930 5333
www.fanninuk.com
enquiries@fanninuk.com

Fannin Ltd.
Fannin House
South County Business Park
Leopardstown
Dublin 18
Tel: +353 (0)1290 7000
www.fannin.ie
info@fannin.ie

Fannin (NI) Ltd.
Unit 2, Heron View
Airport Road West
Belfast
BT3 9LN
Tel: +44 (0) 2890 735 588
www.fanninni.com
info@fanninni.com

Caring for life
Advancing safety for patients and providers