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Unusual association of diseases/symptoms

Septic arthritis presenting as brachial plexus neurophaty

A Mascarenhas,[1] C Almeida,[2] C Constantino,[1] A P Soudo,[3] E Calado,[4] J P Vieira[4]

[1]Department of General Pediatrics, Hospital Dona Estefânia, Rua Jacinta Marta, Lisboa, Portugal;
[2]Department of General Pediatrics, Hospital do Divino Espirito Santo, Rua da Grotinha – Arrifes, Ponta Delgada, Portugal;
[3]Physical Medicine and Rehabilitation Department, Hospital Dona Estefânia, Rua Jacinta Marta, Lisboa, Portugal;
[4]Pediatric Neurology Department, Hospital Dona Estefânia, Rua Jacinta Marta, Lisboa, Portugal

Correspondence to Dr A Mascarenhas, amascarenhas22@yahoo.com

Summary

Bone and joint infections are rare in the neonatal period. They often present with pseudo paralysis of the affected limb due to pain and discomfort caused by movement. The existence of a concomitant neuropathy is a rare and insufficiently understood phenomenon with few cases described. The authors report the case of a 7-week infant, born prematurely and with *Staphylococcus aureus* neonatal sepsis, who presented to the emergency room with a paretic right upper limb. Osteoarticular infection complicated with brachial plexus neuropathy was considered and MRI and electromyography the confirmed diagnosis. There was a good outcome after antibiotic treatment and functional rehabilitation.

BACKGROUND

Osteomyelitis and septic arthritis are rare in the newborn and usually result from bacterial haematogeneous dissemination. *Staphylococcus aureus* is the most common aetiologic agent although other bacteria, and more rarely viruses and fungi may be implicated.[1–3] A pseudoparalysis of the involved limb is frequently found, around the infection focus.[4] The association between these infections and true paralysis is either rare or under-recognised; only a few cases were reported with electromyographic (EMG) studies that document the nerve lesion.[5–9]

CASE PRESENTATION

A 7-week male infant, the second twin of a trigemelar gestation, was delivered by caesarean section after spontaneous rupture of membranes at 32 weeks of gestation. His birth weight was 2.004 g and Apgar scores 8 and 9 at the first and fifth minutes. He had a transitory respiratory distress syndrome and a late onset *S aureus* septicaemia treated with flucloxacillin and gentamicin for 21 days. Cardiologic evaluation for a heart murmur disclosed a patent foramen ovale and ductus arteriosus. He was clinically well and discharged home at the age of 5 weeks.

He was brought to the Emergency Department 10 days later, because his parents noticed that his right arm was kept in a peculiar position, had decreased mobility and seemed painful with attempted movement. These symptoms became gradually apparent for about 1 week. There was no history of trauma, immunisation, fever, refusal of feeding or lethargy.

On examination, the child was afebrile and alert and looked generally well. The right upper limb was flaccid and held in a position of extension at the elbow and slight pronation of the forearm with flexion of the first and second fingers. Any attempt to passively move the paretic limb evoked a cry. The baby's irritability precluded any meaningful testing of sensation. The general and neurological examinations were otherwise normal, and neither palpebral ptosis nor myosis was present.

INVESTIGATIONS

In the peripheral blood, there were 11.820/mm^3 white blood cells, with 41.2% neutrophils. C reactive protein was 5.21 mg/dl and erythrocyte sedimentation rate was 21 mm (first hour). X-rays and ultrasonography of the shoulder were normal. The MRI of the shoulder and upper arm revealed distension of the shoulder capsule by increased interarticular fluid suggesting septic arthritis. In the axilla, inferomedially to the joint, there was also a triangular zone with abnormal T2 hypersignal consistent with a periarticular inflammatory lesion (figure 1).

The neurophysiologic study had normal motor and sensory nerve conduction studies but demonstrated signs of acute neurogenic damage to the brachial biceps muscle consistent with a lesion of the musculocutaneous nerve or lateral cord of the brachial plexus. Bone scintigraphy was normal and blood cultures were negative.

TREATMENT

The patient was treated with intravenous cefotaxime and gentamicin and had a full recovery within 1 month.

DISCUSSION

Hypomobility of one limb in infancy leads to the suspicion, among other causes, of bone or joint infection.[4] Rare cases of septic arthritis of the shoulder or osteomyelitis of the humerus were associated with brachial plexus neuropathy.[5–12] Early detection and treatment may be important in minimising lesions of the shoulder joint or humerus and reduce the chance for neurologic deficits related to brachial plexus neuropathy.

Osteomyelitis and septic arthritis in newborns and in young infants may not manifest fever or systemic symptoms and the laboratory parameters of infection may be

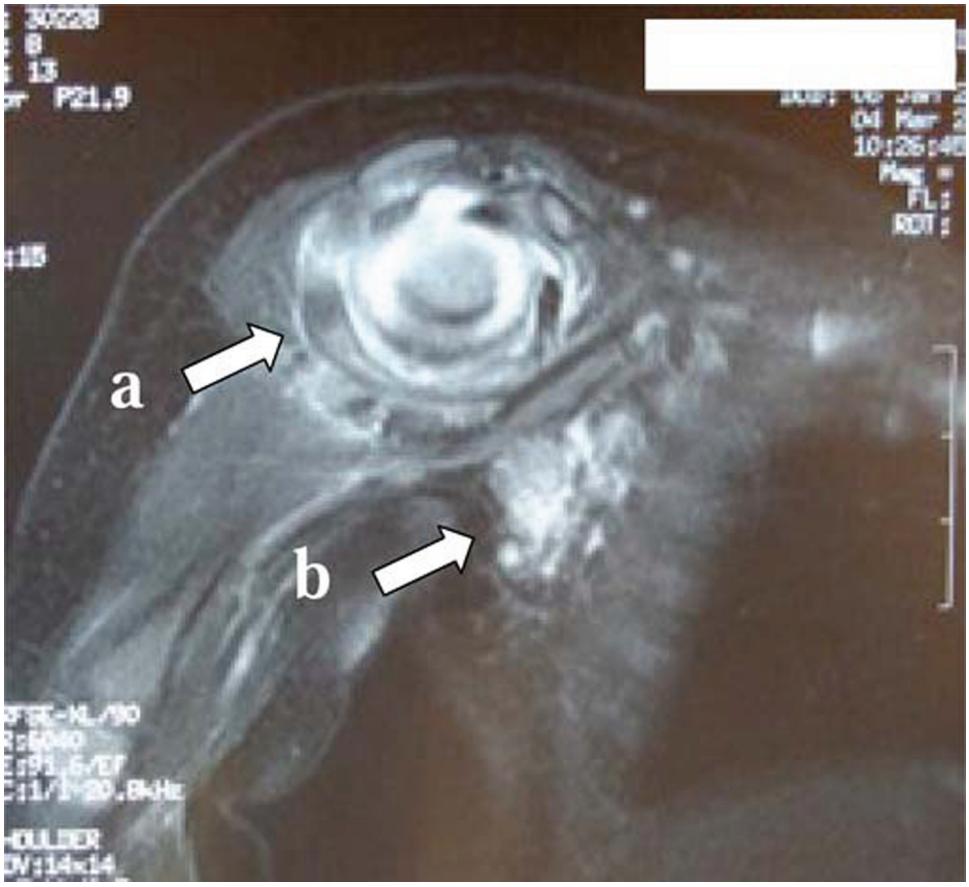

Figure 1 MRI of the right shoulder and upper arm revealing distension of the shoulder capsule by increased interarticular fluid (A) and T2 hypersignal in the axilla (B).

only slightly abnormal.[1 3] Moreover in the immediate postpartum the most common cause for upper limb paralysis is trauma, not infection.[1]

Our patient's symptoms did not appear immediately postpartum and the presence of multiple risk factors (prematurity, multiple invasive procedures such as venipuncture and umbilical catheterisation) for *S aureus* bacteriemia evoked the diagnosis of subsequent septic arthritis or osteomyelitis.[3] Although we could not isolate the agent for the patient's arthritis (this may occur in up to 50% of cases), the infectious aetiology was more likely, considering the global clinical picture.[1 3]

Brachial plexus neuropathy may in general have multiple aetiologies, trauma being one of the most common. Radiotherapy applied to the axillary field, brachial plexus invasion by tumours of the thoracic outlet and apical lung, inflammatory plexitis secondary to immunisations or idiopathic plexitis are other possible causes. The leading cause of neonatal brachial plexus neuropathy is trauma although several other cases were reported in association with arthritis of the shoulder or osteomyelitis of the proximal humerus.[5–12]

This has been reported only in neonates and its pathophysiology is not clear.

Comparing our patient's findings with those reported previously we would like to emphasise some points: (1) The absence of fever and/or systemic symptoms is striking in many cases; paralysis associated with local pain is probably the most helpful signs. (2) Not all patients had MRI; the patients reported by Gabriel *et al* had a MRI abnormality (T2 hypersignal extending inferomedially to the shoulder joint) similar to our patient's. This finding seems helpful in the investigation of these patients (as bone scans with Tcm99 do not provide information on the state of structures adjacent to the shoulder/humerus) and suggests that extension of the inflammatory lesion to nearby structures may lead to the brachial plexus lesion. Occlusion of the vasa nervorum due to vasculitis may cause ischaemic neuropathy or alternatively nerve compression may occur. (3) Neurophysiologic findings were reported in several cases.[5–9] EMG signs of denervation (decreased interference pattern and spontaneous activity) in more proximal muscles were more frequently reported, although distal muscles also revealed lesser abnormalities. Nerve conduction studies (motor and sensory amplitudes and nerve conduction velocity in the median and ulnar nerves) were normal in our case and other's but were abnormal in the patients reported by Gabriel *et al* and Sadleir and Connolly. (4) The treatment strategy was different from one case to another (from antibiotics to antibiotics plus steroids to surgical decompression). Although we believe that treatment in each case must be individualised, the fact that not all patients had a full recovery should lead the physician to consider that this entity may have a guarded prognosis regarding recovery of neurologic function.

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In conclusion, paralysis of an extremity in the neonatal period should raise the suspicion of septic arthritis or osteomyelitis. Early recognition and treatment may be imperative measures to minimise not only joint destruction but also diminish the potential for neurological damage.

The patient was not involved in any clinical trial.

Learning points

- Paralysis of an extremity in the neonatal period should raise the suspicion of septic arthritis or osteomyelitis.
- The association between these infections and true paralysis is either rare or under-recognised.
- Osteomyelitis and septic arthritis in newborns and in young infants may not manifest fever or systemic symptoms.
- Early detection and treatment may be important in minimising joint destruction and neurological damage.

Competing interests None.

Patient consent Obtained.

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