Short report

Intramedullary abscess — a rare complication of spinal dysraphism

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SUMMARY Two cases are reported of patients with spinal dysraphism who developed abscesses within the spinal cord. In one case the infection had spread to the cord through a dermal sinus, as in the six cases previously recorded in the literature. In the other patient the sepsis developed within an intramedullary epidermoid tumour, but the route of infection was not clear. Each patient made a virtually full neurological recovery after open drainage of the abscess.

It is well known that spinal dysraphism may be complicated by meningitis, as a result of infection travelling from the skin along a patent dermal sinus. However, spread of infection to within the spinal cord is exceedingly rare, and only six cases have been reported previously of spinal dysraphism leading to an intramedullary abscess. Two further cases are described here. Each made an excellent recovery after the open drainage of pus from inside the cord.

Case 1 (B.H. 33386)

This 21 year old man had a right talipes at birth, but the deformity had not progressed and he was able to work as a lorry driver. Six weeks prior to his admission to the Neurosurgical Unit he gradually developed low back pain which spread down the outside of both legs to the ankles, and paraesthesiae on the dorsum of the left foot. He was admitted to hospital with a provisional diagnosis of a lumbar disc protrusion. Despite bedrest and traction his pain became increasingly severe, and when he developed a feeling of generalised weakness of both legs he was referred for neurosurgical opinion. He had not been febrile and had had no sphincter disturbance.

On examination he was a sallow and ill-looking man in considerable pain. His temperature was 36°C and his pulse was 80 per minute. He had no signs of meningeal irritation but his lumbar spine was immobile with bilateral muscle spasm and tenderness to percussion. At the top of the natal cleft was a small blind pit. Straight leg raising was 50° on the

right, 30° on the left, the stretch test being positive on both sides. There was slight wasting of the right calf and right anterior tibial compartment and the right foot was slightly clawed. There was weakness of hip extension, knee flexion and ankle dorsiflexion on both sides. The ankle jerks and plantar responses were absent. Pinprick sensation was dulled in the L5 dermatomes on both sides and in the right S1 dermatome. Anal tone was normal.

Investigations Plain spinal radiographs showed spina bifida deformities of lumbar vertebrae three to five and the first sacral segment. Myelography showed a wide band running from a low conus at the L2/3 level to the lower end of the subarachnoid space. At the L2 level there was an obstruction apparently due to an expanded conus together with an intradural mass to the right of it. The lumbar cerebrospinal fluid had a protein level of 2.030 g/l and contained 78 white cells per cmm. (76 lymphocytes and 2 polymorphs). No organisms could be grown on culture.

A laminectomy was performed from D12 to L2 inclusive. The conus extended to the L2/3 level. The lower four cm of the spinal cord was swollen, soft and pale yellow. At the L2 level there was a thin rounded projection from the conus which resembled a blister. This was gently torn and thick creamy non-offensive pus escaped. About five ml of pus was gently milked from within the conus which collapsed to a normal size. The area was irrigated with 20,000 units of penicillin and 5 mgm gentamycin dissolved in normal saline and was sprayed with polymyxin powder. The wound was closed without drainage. Post-operatively the patient was treated with parenteral penicillin, flucloxacillin and gentamycin. Pain relief was immediate and within hours he had passed urine without difficulty and had no neurological deficit other than absent ankle jerks. He showed no

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features of meningitis. Culture of the pus grew staphylococcus albus. Histological examination showed large numbers of polymorphs together with squames, suggestive of an inflammatory process within an epidermoid tumour. One month later the pit at the top of the natal cleft was excised. It was a simple dimple which did not lead to a deep sinus. Four months later, the patient had no residual symptoms and was back at work as a lorry driver.

Case 2 (B.H. 32977)

A 20 year old cashier was admitted to the neuro-surgical unit. Three weeks previously she had developed low back pain after lifting. Over the next week the pain worsened and she felt unwell. She then slowly developed weakness of the legs and a numbness of the legs and buttocks. By the time of admission she was unable to stand and had been unable to pass urine for 24 hours. She had been born with a lumbar meningocele which had been excised three days after birth. At the age of two years the scar discharged pus and within a few days she developed meningitis. Antibiotic treatment led to a complete recovery but from time to time thereafter the scar would become inflamed and then discharge a small amount of serous fluid. This had last happened about two years before the illness which precipitated her admission. Otherwise she had remained well with no neurological disability.

On examination her temperature was 36.8°C and her pulse 116 per minute. She was drowsy, photophobic and irritable, with marked neck stiffness and a positive Kernig's sign. Her bladder was enlarged and she had a severe flaccid paraparesis. The leg tendon reflexes and the plantar responses were absent. There was hypalgesia from L3 to S3 on the right, and L4 to S1 on the left, with loss of joint position sense in the right foot. In the lower lumbar region was a transverse scar containing a central sinus. Although the area around this sinus was tender there was no discharge from it.

Investigations included haemoglobin 9.8 g/dL, white cell count $13.9 \times 10^9/l$, sedimentation rate 53 mm in the first hour (Westergren), lumbar cerebrospinal fluid contained protein 14 g/l, 1100 polymorphs and 90 lymphocytes per cmm, glucose 0.8 m.mol/l. Plain radiographs of the lumbar spine showed spina bifida from L4 to S1, with a pinhole defect in the L5 neural arch. Myelography showed a low conus which was tethered at L5 level, and expanded to cause a partial block at L3 level.

At operation the sinus was dissected to the point where it passed between the neural arches of L4 and L5. A laminectomy from L2-L5 inclusive showed that the sinus track entered the dura. The dura was opened and the sinus was seen to enter a swollen conus, which was tethered in a fibrofatty intradural mass at L4/5 level. A large quantity of pus discharged from the conus through the divided sinus. The conus was not incised. The dura was left open and the wound closed in layers.

Culture of the pus grew anaerobic Streptococci. The patient was treated with parenteral gentamycin, flucloxacillin and metronidazole and made a steady and uncomplicated recovery. Five months after operation she was walking almost normally without a stick and had normal sphincter control. She had absent knee and ankle jerks in the right leg and moderate weakness of dorsiflexion at the right ankle. There was patchy hypalgesia in the L3 to S3 dermatomes on the right side.

Discussion

In spinal dysraphism a common feature is the maldevelopment of part of the neural tube, usually at its lower end, where differentiation of the neurectoderm occurs last. Normally the caudal end of the neural canal is obliterated during the fifth month of foetal life to become the filum terminale. Failure of this process may leave an epithelialised "dermal sinus" leading from the central nervous system to the exterior.¹ The superficial entrance to the sinus, often marked by a tuft of hair or naevus, is usually in the lumbosacral area, although such sinuses may occur at any point along the spinal axis and also in the occipital region.² In its minimal and most common form the abnormality consists of a blind dimple of skin tethered to the deeper structures.³ This was the state of affairs in Case 1. When the skin deformity leads into a persistent sinus the track may lead to within the dura and can end in a dermoid or epidermoid cyst,² but sometimes it is in continuity with a dilated central canal of the spinal cord extending down into the filum terminale.¹ A dermal sinus may permit infection to spread from the skin to within the body. If the sinus stops short of the dura, then subcutaneous abscesses may occur. Deeper extension of the sinus may lead to infection of the central nervous system. This usually takes the form of meningitis, generally in early childhood and often recurrent. Matson and Jerva⁴ pointed out that if a midline skin pit is associated with neurological abnormalities in the legs, then it probably connects with a sinus extending to within the dura, and prophylactic excision is advisable.

Although meningitis is a not uncommon consequence of a dermal sinus, and extradural and subdural abscesses occur occasionally,² the development of an abscess within the spinal cord itself is exceedingly rare. Six cases have been reported previously and details of these are shown in the table. In all these cases and in Case 2 reported here there was a dermal sinus present. In Case 1 there was no apparent connection between the skin and the abscess, and

Table Cases of spinal intramedullary abscess associated with spinal dysraphism

	Age	Length of symptoms	Level	Organism	Spinal abnormality	Operation	Result
Dubreuilh (1877)	2½ yr	4 mth	mid cervical	—	Dermal sinus and dermoid tumour C3/4	—	Died
Walker and Dykes (1936)	3 yr	1 mth	conus	pneumococcus staphylococcus coliforms	Lumbosacral dermal sinus	Myelotomy External drainage	No functional disability
Shenkin <i>et al</i> (1944)	22 mth	2 days	conus	diphtheroids bacteroides streptococci E coli	Sacral Dermal sinus	Myelotomy External drainage Dura left open	Died
Mount (1949)	10 yr	7 mth	C7–D7	—	Dermal sinus between D4/5	Myelotomy Dura left open	No functional disability
El Gindi and Fairburn (1969)	16 mth	Several days	D1 downwards	B-proteus streptococcus	Lumbosacral dermal sinus	Myelotomy Dura left open External drainage	Paraplegia below D3 level
Di Tullio (1977)	1 mth	6 days	conus	Proteus mirabilis	Low conus sacral dermal sinus	Myelotomy Dura left open	Severe distal weakness of 1 leg
Bean <i>et al</i> (1979)	15 yr	5 days	C2–D2	Proteus mirabilis	Dermal sinus C1/2	Myelotomy Dura left open	No functional disability
Maurice-Williams <i>et al</i> (1980) Case 1	21 yr	6 weeks	conus	Staph albus	Sacroccygeal dimple Low conus intraspinal epidermoid	Expression of pus under direct vision Dura closed	No functional disability
Maurice-Williams <i>et al</i> (1980) Case 2	20 yr	3 weeks	conus	anaerobic streptococcus	Meningocele Lumbar dermal sinus	Expression of pus under direct vision Dura left open	No functional disability

histological examination of the excised skin pit showed no sinus extending from it. The infection appeared to have occurred within an intramedullary epidermoid tumour, and the only possible explanation for it is that it was blood-borne from some minor septic focus elsewhere in the body.

In Case 2 a meningocele had been excised in infancy. At the age of two an infection in the scar had been followed by an attack of meningitis. This was evidence for the existence of a dermal sinus and in retrospect it is clear that excision of the sinus should have been carried out prophylactically.

Abscesses within the spinal cord are rare whatever the cause. Two recent papers have reviewed the features of over 50 cases collected from the literature.^{5,6} About 60% were metastatic from infection elsewhere while 15% resulted from compound injuries to the spine. The remainder, including those associated with spinal dysraphism, were caused by the spread of sepsis from adjacent structures. Intramedullary abscesses often evolve surprisingly slowly, and it may be a matter of months before the diagnosis is made. The onset of symptoms may be accompanied by the clinical and cerebrospinal fluid (CSF) features of bacterial meningitis or the picture may mimic a spinal tumour. In the latter case the CSF changes may consist of an elevated protein level and a modestly elevated white cell count.

Although the abscess frequently tracks a considerable way along the cord,⁷ definitive surgical treatment may be followed by substantial neurological recovery. Di Tullio⁸ found that 39% of the reported cases of intramedullary abscess had made a full recovery and only 15% had been left severely disabled. Before operation 72% of cases had been judged to be in the latter category. The pus should be drained from the cord either through a midline myelotomy or through any area of obvious "pointing". Post-operative drainage of the cord is not necessary.⁶ Recurrence does not appear to be a problem if adequate primary clearance of pus has been combined with appropriate antibiotic therapy.

We thank Mr JR Bartlett for permission to publish details of Case 2, which was treated under his care.

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