Case Reports and Technical Notes

Intramedullary Spinal Abscess as a Complication of a Congenital Dermal Sinus

Case Report

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Congenital dermal sinuses result from a failure of separation of the cutaneous epithelial ectoderm from the neuroepithelial ectoderm during the first month of intra-uterine life. Moise\textsuperscript{10} was the first to report a case of meningitis in an 18-year-old boy with a discharging sacral sinus. Since that time, many other authors\textsuperscript{8,9,11-16} have reported cases of infection of the central nervous system in which the pathway of infection was along the track of a dermal sinus. Most of the infections which have arisen in this way have taken the form of meningitis or subdural abscess. In only three previous cases\textsuperscript{11,14,16} has the infection resulted in the formation of an intramedullary spinal abscess. We report here a fourth case of this unusual complication.

Case Report

A 16-month-old boy was admitted to the Infectious Diseases Unit at Rush Green Hospital on August 8, 1966, with a 7-day history of fever, restlessness, and irritability. He showed signs of meningism, and a clinical diagnosis of pyogenic meningitis was confirmed by lumbar puncture. The cerebrospinal fluid showed 1,300 white blood cells per cu mm (90\% polymorphs), and \textit{B. proteus} was grown on culture. The child's previous medical history had been clear, but it had been noted at birth that he had a small dimple with an underlying sinus in the skin in the midline over the lumbosacral region. He was treated with penicillin, sulphadiazine, and chloramphenicol, and made a rapid and full recovery from his meningitis.

On September 28, 1966, he was seen in the Neurosurgical Out-Patient Clinic at Oldchurch Hospital at the request of his physi-cian who suggested that the earlier meningitis had probably occurred as a result of infection spreading along the track of the dermal sinus. The child was now generally quite well. There were no abnormal neurological signs in the legs.

The dimple in the lumbosacral region was covered by a dry scab, and there was slight redness of the surrounding skin over a diameter of about 5 mm. It was agreed that the recent meningitis had almost certainly been due to infection entering along the track of the dermal sinus and prophylactic excision of the sinus was advised. However, in view of the presence of an apparently trivial local infection around the mouth of the sinus, it was advised that operation should be deferred for a short time. He was listed for admission in 3 weeks and the parents were advised as to the appropriate local treatment to be applied to the superficial infection in the meantime.

Within a few days of his visit to the Out-Patient Clinic, the child became febrile, restless, and irritable, and was admitted as an emergency case on October 10, 1966.

Examination. The patient had a temperature of 102° F. He showed no signs of meningism, and the legs were neurologically normal. There was an area of brawny red swelling 5 cm in diameter around the mouth of the lumbosacral sinus. Aspiration of this swelling produced a few drops of pus from which \textit{B. proteus} and non-hemolytic Streptococci were grown. He was treated by erythromycin, ampicillin, and sulphonamides. Over the next few days, his temperature came down to almost normal, his general condition improved, and there was some regression of the cellulitis. On the morning of October 13, however, he was found to have developed a complete flaccid paralysis of the legs.

Received for publication September 11, 1968.
Operation. The afternoon of October 13, vertical incision was made over the lumbo-sacral swelling. A ragged abscess cavity was found in the subcutaneous tissues containing 5 ml of pus. This abscess did not appear to penetrate the lumbo-sacral aponeurosis. However, a dermal tube was found passing deeply from the sinus and entering a defect about 5 mm in diameter in one of the vertebral arches to become continuous with the dura mater. There was no epidural abscess. The neck of the dermal tube was ligated close to the dura and its superficial part excised. The wound was closed with drainage.

There was no change in his clinical condition over the next 24 hours and a cisternal myelogram was carried out under general anesthesia to gain more information as to the cause of the paraplegia. The contrast medium descended freely to C-8 (Fig. 1.) and after prolonged tipping a small amount trickled down as a thin layer on each side of the spinal canal as far as D-12.

Second Operation. On October 15, 1966, a laminectomy from D-12 to L-2 was carried out. There was no epidural pus. The dura was tense and showed no pulsation; when it was opened, the cord appeared swollen and uniformly pink throughout the exposure. There was no subdural pus. A midline incision 1.5 cm in length was made over the dorsum of the cord centered at D-11. At a depth of 5 mm a large abscess cavity containing thick yellow pus was entered. Some 5 ml of pus drained from this cavity. A fine rubber catheter was introduced cranially into the abscess cavity and passed upward for a distance of 7.5 cm. A similar myelotomy was carried out at L1-2, and an identical abscess cavity was found. Fine rubber catheters were left in the abscess cavity for drainage and instillation of antibiotics. The dura was left open, and the superficial tissues closed in layers.

Over the next 3 days, there was an improvement in the child's general condition but no recovery of the paraplegia. Systemic antibiotics were given in large doses, and local antibiotics were instilled into the abscess cavity after aspiration of all available pus. On October 19, 1966, 4 days after myelotomy, the child was found to have developed a severe flaccid paresis of both arms, more marked on the left, with a left Horner's syndrome.

Third Operation. Later the same day, a laminectomy from D-1 to D-9 was carried out. The intramedullary abscess was found to have extended throughout this portion of the cord. Further dorsal myelotomies were carried out at D-2 and D-7 with findings identical to those at the D11-L1 level. A catheter was found to pass upward in the abscess cavity to the mid-cervical level. Drainage was established as after the previous operation.

Following this operation, there was a slow but steady improvement in the child's general condition. There was a rapid and full recovery in the function of the right arm in the course of 2 weeks. Recovery in the left arm was much slower and took place over a period of many months. The child has been left with a mild weakness of the intrinsic muscles of the left hand. There has been a return of a flicker of movement in the right leg but no recovery in the left leg. At 18 months after the onset of his illness, the child remains for all practicable purposes completely paraplegic and anesthetic up to the D-3 level.

Discussion

Intramedullary spinal abscess is a rare condition. The disease was first described in 1830 by Hart. In 1944, Arzt reviewed the 40 cases previously described in the litera-
ture and added three cases of his own. Since that time, Seven further cases\(^1,3,5,6,14,18\) have been added, making a total of 50. The first case of successful treatment was that described by Cavazzani\(^6\) in 1899. The abscess occurred as a complication of traumatic paraplegia and following laminectomy and drainage of the abscess, the patient survived but without any improvement in his paraplegia. In all, 15 cases have been operated on. Nine of these survived, and seven of the survivors made a good functional recovery. There were no survivors among the non-operated cases.

Successful surgical treatment has nearly always taken the form of laminectomy and dorsal myelotomy. In a few cases, aspiration of the abscess with or without the instillation of antibiotics has been successful.

Many different etiological causes have been described for these abscesses. Most of them have been metastatic in origin, occurring in association with such diseases as bacterial endocarditis, pulmonary infection, septic abortion and infection of the urogenital tract. Some have arisen by direct spread from local infective lesions such as diseases of the vertebrae, penetrating injuries and myelomeningocele. In a few cases, no primary focus of infection has been found. Only three cases have previously been described in association with congenital dermal sinuses.\(^1,14-16\)

A striking pathological finding in many of the cases described has been the length of the cord involved in the abscess. Having become established in the substance of the cord, the abscess is able to extend longitudinally, presumably by separating the fiber tracts. In three cases, the whole length of the cord was found to be involved at autopsy. In 10 cases, half the cord was affected. This tendency to longitudinal spread is well illustrated in our case, with the abscess extending from the mid-lumbar to the upper cervical region.

The disease very often runs an acute course with death supervening in a few days or weeks. Some cases, however, have been relatively chronic, and have shown a surprisingly good neurological recovery after surgical drainage of their abscess. Wolman and Adson's\(^17\) case had been almost completely paraplegic for 3 months before operation yet subsequently made an almost full functional recovery. Walker and Dyke's\(^16\) patient had been paraplegic for a month before operation but again made an almost complete recovery after operation. The return of function in these cases is in marked contrast to that in cases of epidural spinal abscess in which a good functional recovery is almost unknown once conduction in the spinal cord has become seriously affected. Presumably the centrally placed abscess is less likely than the epidural abscess to produce irreversible vascular changes in the cord. In our case there was good return of function in the arms following drainage of the cervical part of the abscess while the legs remained completely paralyzed.

The potential danger of a congenital dermal sinus acting as a pathway for infection to enter the nervous system has been stressed by many authors.\(^6,16\) Matson and Jerva\(^6\) recommend total excision of every congenital dermal sinus tract as soon as the condition is recognized. They warn that the surgeon undertaking such an operation must be prepared to carry out an extensive dissection since the tract may extend for some distance within the spinal canal and may be associated with an intraspinal dermoid cyst.

Our experience in this case further underlines the need for early prophylactic surgery in these cases. This child was first seen 1 month after his first attack of meningitis. At that time there was an apparently trivial infection in the soft tissues surrounding the opening of the sinus. Operation was deferred to allow this infection to subside and to permit definitive surgery to be undertaken with safety. However, the child actually developed a more serious local infection within 10 days, followed by an intramedullary spinal abscess. In retrospect it would have been wiser to have hospitalized the child at once and to have applied intensive local and systemic treatment to overcome the infection as a preliminary to early surgery.

**Summary**

A case of intramedullary spinal abscess arising in association with a congenital dermal sinus has been described, and the importance of early excision of such dermal sinuses before serious infection can enter the nervous system has been stressed.
References
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