Intramedullary abscess of the spinal cord in children: a case report and review of the literature

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A case report of an intramedullary spinal cord abscess in a 13-month-old boy and a review of relevant existing pediatric literature is presented. Thirty-eight cases of pediatric intramedullary spinal cord abscess are analyzed for presenting signs and symptoms, microbiology of isolated organisms, surgical intervention, antibiotic administration and outcome. The most significant variable on outcome is timely surgical intervention, followed by appropriate antibiotic administration.

Intramedullary abscess of the spinal cord (IASC) is a rare clinical entity described initially in an adult patient by Hart in 1830. Since then fewer than 100 cases have been described in adults in the medical literature and in several reviews, focused largely on the presentation of this disease in adults. The pathogenesis of this disease in adults is chiefly hematogenous spread from a cardiopulmonary source. In children hematogenous spread from a variety of sources does occur, but preexisting congenital defects in spinal cord development are a more common means by which infection occurs. IASC in children is predominantly secondary to direct contiguous spread of infection from the skin.

We report a case of a 13-month-old boy seen in our institution with an IASC and review the medical literature of cases of IASC in children.

CASE REPORT

A 13-month-old Hispanic boy was evaluated at our institution in September 1999 because of a 2-week history of fever and inability to walk. At birth a tuft of hair was noted in the lumbosacral region. He was referred to our institution, and a magnetic resonance imaging (MRI) study revealed a dermal sinus at the level of the fifth lumbar vertebra (L5) with possible tethering of the spinal cord. The patient was followed in the neurosurgery clinic for the first 6 months of life and was subsequently lost to follow-up. During the initial 12 months of life, he developed normally without neurologic deficits. He began to walk at 12 months, but 2 weeks after his first birthday, he developed a fever, increased crying while lying on his back during diaper changes and loss of the ability to walk. A primary care physician diagnosed acute otitis media and prescribed a 10-day course of amoxicillin. The fever initially resolved but resumed several days after the course of amoxicillin was completed, prompting his presentation.
to our institution. Additional information obtained on further questioning of the mother included a report of occasional white discharge from the lesion on the back since birth.

On physical examination the patient was in no distress, afebrile, alert and interactive. Muscle strength was normal in the upper extremities but diminished in the lower extremities. The bilateral lower extremity proximal muscle strength was graded 1/5, and distal muscle strength was graded 0/5. Patellar and Achilles deep tendon reflexes were absent bilaterally. Anal wink reflex was present, and there was no evidence of a neurogenic bladder. On the back was a 1- by 1.2-cm tuft of hair in the L4–L5 region surrounded by a 3- by 4-cm nevus. An MRI of the spine (Figs. 1 and 2) revealed a tethered cord with multiple gadolinium-enhancing intramedullary cystic structures from T12 to L3 vertebral levels and a dermal sinus at L4–L5 disk level. On the ninth hospital day the patient underwent an L1–L3 laminoplasty with L4 laminectomy, drainage of purulent material from the intramedullary cyst and resection of the dermal sinus. The patient was treated with vancomycin, ceftriaxone and ampicillin iv for 2 weeks. An MRI study on Postoperative Day 16 showed a decrease in the size of the intramedullary cyst. Intraoperative cultures grew a mixed growth of anaerobic bacteria: Propionibacterium acnes, Prevotella oralis, Peptostreptococcus magnus, Veillonella parvula, Actinomyces meyeri and Bacteroides ureolyticus. The antibiotic coverage was changed to ampicillin and metronidazole iv for an additional 3 weeks.

He was transferred to a rehabilitation center after 53 days of hospitalization and completed another 9 weeks of iv ampicillin therapy for a total of 3.5 months of iv antibiotic therapy. The patient remained afebrile with minor increase in movement of the lower extremities, but he was unable to walk. One month after the discontinuation of antibiotics, a repeat laminectomy of L1–L3 was performed to repair the tethered cord. No tumor was identified. At 32 months of age he was able

FIG. 1. A midline sagittal T1-weighted gadolinium-enhanced scan obtained when the patient was 13 months of age, immediately before surgery, demonstrates the sinus tract (arrow) extending from the posterior superficial soft tissues to the midlumbar spinal canal. The low signal structure within the canal probably represents a dermoid cyst. 5, fifth lumbar vertebra.

FIG. 2. A parasagittal image demonstrates two enhancing intramedullary lesions (arrows) representing the abscesses. 5, fifth lumbar vertebra.

FIG. 3. A parasagittal image demonstrates the abscesses at the L4–L5 vertebral level (arrow) after the removal of the dermoid sinus. 5, fifth lumbar vertebra.
to ambulate independently with ankle-foot orthotics, with bilateral lower extremity proximal muscle strength 2/5 and distal muscle strength 1/5 without regaining patellar or Achilles deep tendon reflexes.

**METHODS**

**Review of the literature.** A Medline search of the world’s literature was conducted using keywords pediatric, intramedullary, spinal cord and abscess. Patients were included if they were <18 years of age. Thirty-eight cases of pediatric IASC were identified between the years of 1875 and 2001. Case histories were found from many countries and included translation from Spanish and German journals. The case definition of IASC was the presence of bacterial, fungal and parasitic infections involving abscess formation within the spinal cord as identified by radiologic evaluation, surgical examination or autopsy. Outcome was graded as: Grade 1, complete neurologic recovery; Grade 2, mild neurologic impairment including nonfunctional paresis, hypoesthesia, hyperreflexia; Grade 3, moderate neurologic impairment including loss of sphincter tone, gait disturbance, continued ability to ambulate; Grade 4, severe neurologic impairment including paralysis, inability to ambulate; and Grade 5, death (Table 1). Compiled data included demographics, laboratory test results, antibiotic administration, surgical intervention and outcome.

**Statistical assessment.** We used a two tailed Fisher exact test and chi square tests to assess correlation between datasets including time of presentation, surgery and antibiotic administration with relation to outcome. All calculations were performed with and without inclusion of death as an outcome measure, and outcome was assessed in absolute terms as well as in relation to severity of symptoms at presentation, i.e., change in outcome. The statistical programs used were Epistat (CDC) and Sysat.

**RESULTS**

**Patient population.** Of the 38 cases of pediatric IASC, 17 (45%) were female and 21 (55%) were male. Age of presentation ranged from 8 days to 17 years (Fig. 3). The median age of presentation was 36 months. Of the 38 cases 20 (53%) had a prior anatomic defect, whereas 2 of the 13 children (15%) presenting at 5 years or older had a preexisting anatomic defect (not including the decubitus ulcer) was 15.5 months. Five patients had preexisting anatomical defects that were not appreciated until their presentation with IASC. Seventeen of the 25 children <5 years (68%) had a preexisting anatomic defect, whereas 2 of the 13 children (15%) presenting at 5 years or older had a preexisting deficit (P = 0.003).

**Clinical features.** Presenting signs and symptoms were available for review in 36 cases (see Table 2). Thirty-two (89%) of the patients presented with neurologic deficits: paralysis; paresthesia; urinary or fecal incontinence. The diagnosis of IASC is made difficult because of absence of fever in 44% and paralysis in 42%. On admission 12 of 36 (33%) patients presented with documented body temperature ≥100.4°F (mean 101.2°F). The median duration of signs or symptoms was 8 days with a range from 1 day to 3 years.

**Laboratory data.** The peripheral white blood cell (WBC) count was available for 16 cases with ranges from 5,000 to 34,500/mm³ and a mean of 13,100/mm³. Eleven patients (69%) had a total WBC count of >10,000/mm³, but only 2 patients (13%) had counts >2 sd from the normal mean value for age. Ten cases had a record of the neutrophil count, and 9 (90%) had neutrophil predominance with a mean of 70% neutrophils. Cerebrospinal fluid (CSF) was abnormal in 18 (78%) of 23 patients in whom it was obtained. Six CSF cultures were positive. CSF was described as purulent CSF in 4 patients, there was a pleocytosis in 9 patients, the CSF protein was elevated in 5 patients and the CSF glucose was low in 2 patients. A patient who had antischistosome antibodies in his serum and CSF was not included as a case of meningitis because no clinical signs of meningitis were reported.

| Outcome                      | No. | %    |
|------------------------------|-----|------|
| Complete neurologic recovery | 7   | 18   |
| Mild neurologic impairment   | 15  | 39   |
| Moderate neurologic impairment| 4  | 11   |
| Severe neurologic impairment | 4  | 11   |
| Death                        | 8   | 21   |

* See text for definitions of degrees of impairment.

*Fig. 3. Age of presentation for 38 cases of pediatric IASC.*
Radiologic findings. Imaging was available for 35 cases. Plain film radiographs performed in 10 patients revealed spina bifida in 2 patients and spinal dysraphism in 1 patient. Twelve patients received myelograms of which 10 revealed blockage of CSF flow at the site of abscess. Results of the other 2 patients who had undergone myelograms were not reported in the case histories. Computed tomography scans of the spine were performed in 2 patients. One spinal CT scan interpreted as normal had an abscess detected on subsequent MRI. A computed tomography myelogram performed in 1 patient revealed blockage of CSF flow at the site of an abscess. MRI scans performed in 13 patients revealed pathology in all 13 cases.

Microbiology. Abscess culture results were available for 29 cases (Table 3). Sixteen patients (55%) had a single organism isolated, 5 (17%) had mixed bacterial flora and 8 (28%) had sterile fluid. Four of the 8 cases with sterile fluid had frank pus, and 3 of these patients reported prior antibiotic use. Of the cases with sterile fluid, a Gram stain was consistent with Fusobacterium spp. in one patient and Gram-negative bacilli in another. There were equal numbers of cases from whom a single isolate of Gram-positive as well as Gram-negative bacteria was grown in the abscess or CSF fluid, a Gram stain was consistent with Fusobacterium spp., fermentative coliform bacteria. All mixed bacterial flora grew from patients with lumbar dermal sinuses and included aerobic Streptococcus, Staphylococcus spp., fermentative coliform bacteria and anaerobic bacteria. Unusual microorganisms causing IASC in children included Brucella abortus, Mycobacterium tuberculosis and Schistosoma spp.

Source of infection. Twenty-one (55%) patients had an isolated anatomic spinal defect as the likely source of direct spread of infection from contiguous skin via a sinus tract. In these cases no other source was identified. In the remaining 17 patients, the primary spread of infection was presumed to be hematogenous in 12 patients. The largest source of probable hematogenous spread was from a urogenital tract source: 2 cases of vulvovaginitis; 1 unspecified urinary tract infection; 1 pyelonephritis; and 1 perinephric abscess. Other probable hematogenous sources included 2 patients with pneumonia, 1 with endocarditis, 1 with a middle ear infection and a sagittal sinus thrombosis, 2 with schistosomiasis and 1 with brucellosis. One patient had no evidence of a primary source of infection, and 4 had insufficient information to determine the primary source of infection.

Surgical management. Surgical reports were available for review in the case description of 33 patients. Four patients (12%) had undergone excision of a dermal sinus 2 weeks to 7 months before presentation with IASC. They had had previous surgery requiring subsequent laminectomies, and 2 of them required reexcision of a dermoid cyst or sinus. Three additional patients had incomplete surgery at the time of presentation with IASC that included dermal sinus exploration and excision without laminectomy; all three required subsequent laminectomies. A fourth patient that had only dermal sinus explora-

### TABLE 2. Common presenting signs and symptoms of 36 cases of pediatric IASC

| Presenting Signs and Symptoms | No. | %  | Median (Days) | Range (Days) |
|-------------------------------|-----|----|---------------|-------------|
| Paralysis                     | 21  | 58 | 7             | 1–180       |
| Fever                         | 20  | 56 | 7             | 1–60        |
| Urine retention               | 11  | 31 | 8             | 1–690       |
| Paresis                       | 11  | 31 | 20            | 2–690       |
| Back pain                     | 10  | 28 | 125           | 8–1090      |
| Irritability                  | 9   | 25 | 2             | 1–25        |
| Urinary incontinence          | 6   | 17 | 5             | 1–7         |
| Fecal incontinence            | 6   | 17 | 5             | 1–8         |
| Meatingismus                  | 6   | 17 | 5             | 1–14        |
| Paresthesia                   | 5   | 14 | 7             | 3–30        |
| Headache                      | 4   | 11 | 3             | 1–365       |
| Nausea/vomiting               | 4   | 11 | 5             | 1–365       |
| Fecal retention               | 3   | 8  | 5             | 2–7         |
| Abdominal pain                | 2   | 6  | 45            | 3–90        |

### TABLE 3. Organisms identified in bacterial cultures of 29 cases of pediatric IASC

| Organisms                        | No. | %  |
|----------------------------------|-----|----|
| Single isolates (N = 16)          |     |    |
| Staphylococcus spp.              | 5   | 17 |
| Proteus mirabilis                | 3   | 8  |
| Escherichia coli                 | 2   | 7  |
| Schistosoma mansoni              | 2   | 7  |
| Streptococcus haemolyticus       | 1   | 3  |
| Brucella abortus                 | 1   | 3  |
| Enterococcus faecalis            | 1   | 3  |
| Mycobacterium tuberculosis       | 1   | 3  |
| Mixed isolates (N = 5)           |     |    |
| Streptococcus spp.               | 2   | 6  |
| Proteus spp.                     | 2   | 6  |
| E. coli                          | 2   | 6  |
| Bacteroides spp.                 | 2   | 6  |
| Diphtheroids                     | 2   | 6  |
| Streptococcus pneumoniae         | 1   | 3  |
| Staphylococcus spp.              | 1   | 3  |
| Other anaerobes                  | 1   | 3  |

FIG. 4. Frequency of spinal segment involvement in 31 cases of pediatric IASC.
tion died on the ninth postoperative day and autopsy revealed communication between the spinal canal and an intestinal reduplication. Laminectomies involving myelotomy, abscess drainage, dermal sinus or dermoid cyst removal were performed in a total of 32 (97%) of 33 patients. The spinal segments most often involved were the thoracolumbar segments. The midthoracic segments were the next most common involved, with the rest of the patients having spinal segments ranging from C1 to S2 (Fig. 4). Of the patients who underwent laminectomies, 2 patients underwent drain placement, and in 2 patients the wound was left open to drain. Three patients were taken back to the operating room to repeat or extend laminectomies for inadequate drainage. The median time from hospitalization until surgery was 5 days (range, 1 to 78 days).

**Antimicrobial treatment.** Thirteen patients received antibiotics for a median of 9 days before surgical drainage procedures or death. Three (23%) had sterile fluid obtained from the intramedullary abscess, and 10 (77%) had positive cultures despite prior antibiotics. Postoperatively 19 patients (50%) received antibiotics for a median of 20 days. Nine patients were not treated with antibiotics because they were managed in the preantibiotic era. One patient with IASC caused by *M. tuberculosis* received antituberculous therapy for 366 days after surgical drainage procedures were performed. Postoperative antibiotic administration was not reported in 10 cases.

**Outcome.** The only significant variables with an influence on outcome were administration of antibiotics and the time from onset of symptoms until surgery. The case fatality rate was 71% among 7 children who presented in the preantibiotic era (1944 and earlier) and 10% in the 31 children who presented after 1944 when antibiotics were available (*P* = 0.002). Children treated with antibiotics had a mean change in outcome of –1.05 (going from moderate to mild neurologic impairment, for example) which was twice the –0.5 change in morbidity and mortality outcome in children who were not treated with antibiotics. The length of pre- and postsurgical antibiotic administration was not significant.

Six of 7 patients without neurologic sequelae and 13 of 23 patients with neurologic sequelae had information in their case descriptions indicating the timing of their surgical drainage procedure with respect to onset of symptoms and hospitalization. Patients without neurologic sequelae had surgical drainage at a median time to surgery of 1.5 days (range, 1 to 8 days) after hospitalization, whereas the patients with neurologic sequelae had a median time to surgery of 4.0 days (range, 1 to 78 days) after hospitalization (*P* not significant). The median time to surgery from onset of symptoms was 7 days (range 1–365 days) in those without neurologic sequelae and 22.0 days (range, 2 to 1088 days) in those with neurologic sequelae (*P* not significant). Three of 6 patients (50%) without neurologic sequelae had a surgical drainage procedure performed within 5 days after onset of symptoms as compared with none of 13 patients (0%) with neurologic sequelae (*P* = 0.021).

**DISCUSSION**

Intramedullary abscesses of the spinal cord are uncommon in children accounting for only 38 cases identified in this review of the world medical literature since 1830. In children, unlike adults, the abscesses are associated with prior anatomic spinal canal defects. Fifty-three percent of all children and 63% of children younger than 5 years of age have prior anatomic spinal canal defects of which more than two-thirds have been identified before the development of neurologic deficits. These spinal cord defects arise from the failure of the neural tube to close during the fourth week of gestation and result in neural tube defects that involve tissues overlying the spinal cord: the meninges; vertebral arches; muscles; and skin. Spinal dermal sinuses, seen in association with IASC, result from defective separation of the epithelial ectoderm from the neural ectoderm and subsequent communication between the skin and deeper structures. They can terminate in soft tissue or in the epidural space, or they can form a connection with the dura and the spinal cord, leading to abscesses that arise at any point along the sinus tract. Epidural abscesses can develop outside the thick dura; the outside layer of the meninges adherent to the spinal cord. Subdural abscesses occur under the dura. Intramedullary abscesses occur within the spinal cord itself.

Twenty percent of cases in this series died, 60% had residual neurologic deficits and only 20% recovered without sequelae; rates higher than pediatric epidural6 and subdural7 abscesses and adult intramedullary spinal cord abscesses.8 Neurologic deficits arise from tethering of the spinal cord during growth, mass effect of a concomitant epidermoid or dermoid tumor or subsequent infection. It is widely accepted in the neurosurgical community that early imaging, exploration and correction of the spinal cord defect including complete excision of the dermal sinus is warranted before the development of neurologic symptoms.9 Because all cases of IASC with concomitant isolated dermal sinus presented at >6 months of age, this suggests that complete resection of the dermal sinus before 6 months of age may prevent the formation of IASC and it’s significant morbidity and mortality. Inadequate exploration and incomplete removal of the dermoid sinus tract can cause recurrent cyst formation and subsequent IASC, as seen in 4 (12%) of 33 patients with surgical records available and underwent surgical excision of their dermal sinus before presentation.
The diagnosis of IASC in infants and children may be difficult and delayed until significant neurologic deficits become apparent because of their indolent presentation, often without fever or elevated peripheral WBC count. Fever was absent in 44% of cases on presentation, and the peripheral WBC count was normal in 87% of children when it was obtained. Parents may not observe early signs and symptoms of IASC in diapered infants and toddlers unable to complain of back pain and where urinary and fecal incontinence is not unusual. The single most sensitive and objective indicator of IASC in children is the finding of paralysis, seen in 58% of the cases reviewed. It is, however, often a late clinical finding after irreversible neurologic impairment has already occurred.

An urgent MRI scan of the spine should be performed (if unavailable, a spine CT scan with and without contrast or spine ultrasound) can confirm the clinical suspicion of IASC. Once the diagnosis of IASC is made, early surgical exploration with incision, drainage and complete excision of the dermoid sinus within 5 days from presentation and postsurgical antibiotic use are critical in reducing the significant mortality and long term neurologic deficits associated with this condition. Exploration of the dermal sinus alone does not appear to be adequate and laminectomy with IASC drainage, and complete excision of the dermoid sinus tract is the surgical procedure of choice.

As with any abscess treatment with antibiotics alone is not sufficient, and pretreatment with antibiotics before surgery may not be beneficial. Although differences in surgical procedures and postoperative care may have contributed to the reduction in mortality rates in the postantibiotic era, it is likely that the concomitant antibiotic therapy contributed significantly to the reduction in mortality and morbidity seen in the patients who received antibiotics in the postantibiotic era compared with those who had only surgical drainage in the preantibiotic era. Duration of antibiotic administration postoperatively was not correlated with a significant difference in outcome, but most cases in recent years were treated successfully with intravenous antibiotics for at least 6 weeks. Given the wide range of microbes cultured from IASC, initially broad spectrum antibiotic combinations such as intravenous vancomycin, cefotaxime and metronidazole to cover *Staphylococcus* spp. *Streptococcus* spp., enteric Gram-negative bacilli and anaerobes should be used until identification of organisms and sensitivities are available.

Prevention of IASC resides largely in the early recognition of congenital defects of the spine, especially dermoid sinuses. MRI scan of the spine, surgical exploration and complete excision of the sinus before 6 months of age may reduce this infectious complication and its significant neurologic morbidity.
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