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SPINAL DERMAL SINUS COMPLICATION IN CHILDREN: CASE SERIES AND LITERATURE REVIEW

COMPLICAÇÃO DO SEIO DÉRMICO ESPINHALEM CRIANÇAS: SÉRIES DE CASOS E REVISÃO DE LITERATURA

COMPLICACIÓNDEL SENO DÉRMICO ESPINAL EN NIÑOS: SERIE DE CASOS Y REVISIÓN DE LA LITERATURA

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ABSTRACT

Introduction: Dermal sinus (DS) is a rare dysraphism. It can be asymptomatic, become infected, and produce severe neurological symptoms. Our objective is to present a series of pediatric cases with spinal DS complicated byinfections (DSCI), describe the findings correlated with the anatomy in a stillbirth, as well as the associated pathologies and their treatment. Method: We analyzeddifferent variables in the clinical histories of 5 children with spinal DSCI. In addition, an anatomical dissection of a stillbirth with lumbar DS was performed. Results:Two males and 3 femaleswith DSCI and a mean age of 2 years and 9 months were included: 2 lumbar (one in the midline and the other in theparamedian region), 1 in the thoracic region, 1 in the upper cervical region,and 1 in the lumbosacral region. The forms of presentation were 3 meningeal profiles (one with pain andlocalized swelling) and 3 neurological deficits (one associated with the meningeal profile and another associated with pain and a tumor). In all cases, the tract of the DS was identified by magnetic resonance imaging (MRI).Associated lesions included 1 dorsal intramedullary dermoid cyst, 1 tethered lumbar spinal cord with syringomyelia, 1 partial cervical medullary disconnection, and 2 spinal dysraphisms. Four were operated on and one died of infectious complications before surgery. In the 12-week-old male fetus with lumbar DS, a permeable tract to the subarachnoid space was verified. Conclusions: DSCIsshould bestudied with MRI to identify their tracts, infectious complications of thecentral nervous system, associated malformations, inclusion tumors, and to enabledifferential diagnosis. Once diagnosed, they should be urgently treated both surgically and with prolonged antibiotictherapy. ***Level of Evidence IV; Therapeutic Study (Treatment Outcome Investigation)***

Keywords: Spinal dysraphism; Infections;Children.

RESUMO

Introdução: O seio dérmico (SD) é uma disrafia rara. Pode ser assintomático, sofrer infecção e produzir quadros graves.Nosso objetivo é apresentar uma série de casos pediátricos com SDs espinhais complicados por infecções (SDCI),descrever os achados correlacionados com a anatomia em um natimorto, as patologias associadas e seu tratamento. Método: Foram analisadasas variáveis da históriaclínica em 5 crianças com SCDI espinhal.Além disso, foi feita uma dissecção anatômica de um natimorto com SD. Resultados: Foram incluídos 2 meninose 3 meninas, com média de idade de 2 anos e 9 meses, com SDCI: 2 lombares(um na linha média e um paramediano), 1 na região torácica, 1 na região cervical superior e 1 lombossacral. As formas de apresentação foram 3 quadros meníngeos, 1 com dor e tumefação lombar local e 3 déficits neurológicos (um associado ao quadro meníngeo e outro associado à dor e tumor). Em todos os casos, o trajeto do SD foi identificado por ressonância magnética (RM).Aslesões associadas foram 1 cisto dermoide intramedular dorsal, 1 medula ancorada lombar com siringomielia, 1 desconexão medular cervical parcial e 2 disrafias espinhais. Quatro participantes foram operados e um foi a óbito decorrente de complicações infecciosas antes da cirurgia.No feto masculino de 12 semanas com SD lombar, foi verificado um trajeto permeável até o espaçosubaracnóideo. Conclusões: Os SDCIs devem ser estudados com RM para identificar o seu trajeto, complicações infecciosas do sistema nervoso, malformações associadas, tumores de inclusão e permitir o diagnóstico diferencial. Eles devem ser tratados cirurgicamente com urgência uma vez diagnosticados e com tratamento prolongado com antibióticos. ***Nível de evidência IV; EstudosTerapêuticos– Investigação dos Resultados do Tratamento***

Descritores: *Disrafismo espinal; Infecções; Crianças.*

RESUMEN

Introducción:El seno dérmico (SD) es una disrafia infrecuente. Puede ser asintomático, infectarse y producircuadros severos. Nuestro objetivo es presentar una serie de casos pediátricos con SD espinalescomplicados porinfecciones (SDCI), describir los hallazgoscorrelacionadoscon

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la anatomía en un mortinato, las patologías asociadas y su tratamiento. Método: Seanalizaron diferentes variables enlas historias clínicas de 5 niños con SDCI espinales. Además, se realizó unadisección anatómicade un mortinato con SD lumbar. Resultados: Se incluyeron2varones y 3 mujeres,de 2 años y 9 meses de edad promedio, con SDCI:2 lumbares (uno en línea media y otro paramediano), 1 en región torácica, 1 cervical superior y 1 lumbosacro. Las formas de presentación fuerontres cuadros meníngeos, 1 con dolor y tumefacción local lumbar y 3 déficits neurológicos (uno asociado a cuadro meníngeo y otro asociado a dolor y tumor). En todos los casos se identificó el trayecto del SD por resonancia magnética (IRM). Como lesiones asociadas hubo1 quiste dermoide intramedular dorsal,1médula lumbar anclada con siringomielia,1 desconexión medular cervical parcial y 2 disrafias espinales. Cuatro fueron operados y uno falleció por complicaciones infecciosas antes de la cirugía. En el feto masculino de 12 semanas con SD lumbar, se verificóun trayecto permeable hasta el espacio subaracnoideo. Conclusiones: Los SDCI se deben estudiarconIRM, para identificarsu trayecto, las complicaciones infecciosas del sistema nervioso, las malformaciones asociadas,los tumores de inclusión y permitirel diagnóstico diferencial. Se deben tratar quirúrgicamente con urgencia una vez diagnosticados y con antibiótico-terapia prolongada. ***Nivel de Evidencia IV; Estudio Terapéutico (Investigación de Resultado de Tratamiento).***

Descriptores: *Disrafia espinal; Infecciones; Niños.*

INTRODUCTION

Spinal dermal sinus (DS) is a rare congenital malformation that results in the incomplete separation of the cutaneous ectoderm from the neuroectoderm during the first weeks of pregnancy.[1,2] Classified among the closed malformations of the spinal cord,[3,4] it is defined as an epithelium-lined tract that runs from the skin of the dorsal region of the trunk, usually along the midline (although not always), and passes through the soft parts to reach the spine, the dural sac, or even the neural elements.[5] They can be asymptomatic or become infected and produce severe neurological conditions. Occasionally, they are associated with other conditions, such as a tethered spinal cord, intradural lipomas, and inclusion cysts.[1-3,6,7] Complications include recurrent bacterial meningitis, intramedullary and cerebral cysts.[1,2,6]

The objective of this paper is to present a series of five children with spinal DS complicated by infections (DSCI), describing the clinical and imaging findings – especially via magnetic resonance (MRI), the anatomy of the entity, the associated pathologies, and their treatment.

METHODS

We evaluated a series of five children with DSCI, treated during the period 2005-2021, by reviewing their available medical records and images. The following variables were analyzed: age, sex, location of the DS, forms of clinical presentation, infectious complications, associated conditions, surgical treatment (type and complications), and follow-up results. All were studied in the same center from both Tesla 1.5 and Tesla 0.5 MRIs with contrast. In addition, an anatomical study of a stillborn with DS from the collection of the 1st Chair of Normal Human Anatomy,Facultad de Medicina, Universidad Nacional del Nordeste (UNNE), located in Corrientes [Argentina]. We measured the cephalo-caudal length and used the table published by Corliss[8] to determine the gestational age of the specimen. A Carl Zeiss OPMI Vario 700 neurosurgical microscope was used in the anatomical dissection to obtain the photographs and information applicable to this study.

RESULTS

Five children, 2 males and 3 females, aged from 2 months to 14 years (mean age of 2 years and 9 months) were evaluated; two with DSCI in the lumbar region(one over the midline and the other in the right paramedian region), one in the dorsal region, one at the upper cervical level, and one in the lumbosacral/sacral region. Four of the 5 patients had some formof neurological deterioration, the forms of presentation of which were 3 with meningeal profiles, one of them with pain and localized lumbar swelling, and 3 with neurological deficits, one of them associated with a meningeal condition and anotherwith pain and a tumor (Table 1).In all the cases, the DS tract was identified with the MRI scans.

Table 1. Case history.

Case no.	1	2	3	4	5
Sex	**F**	**M**	**F**	**F**	**M**
Location	Lumbosacral (S1-S2), midline	Dorsal (T5), midline	Cervical (C1), midline	Lumbar (L4), right paramedian	Lumbar (L4), midline
Formof presentation	Meningeal profile + fistula	Motor deficit	Meningeal profile	Pain and right lumbosacral tumor	Meningeal profile + motor deficit
Age at presentation	8 months	9 years	2 months	3 years	10 months
Associated pathology	Sacral bone dysraphism	Intramedullary dermoid	Partial cervical medullary disconnection	Spina bifida	Tethered spinal cord in L4 + Syringomyelia
Neurological status	Without deficit	Paraparesis	Quadriparesis	Incontinence of the sphincters and dysbasia	Paraparesis
MRI findings	Intradural collection from L4 to S1	Dysraphism in T5 with festering intramedullary, subdural, and extradural collection	Dermal sinus in C1 extending to the posterior side of the spinal cord	Dysraphism in L4 with epidural phlegmon	Dysraphism in L4 with tethered spinal cord, infected syringomyelia and arachnoiditis
Infectious complication	Intradural abscess	Infected dermoid	Pyoventriculitis	Epidural phlegmon	Infected syringomyelia and arachnoiditis
Surgery	Yes	Yes	No	Yes	Yes
Description of the technique used	Resection of the fistula + laminoplasty + durotomy + drainage and lavage	Drainage + Decompression + Resection + Instrumented arthrodesis	--	Resection of the fistula + evacuation of the phlegmon	Drainage + Detethering of the spinal cord
Surgical complications	None	None	Preoperative death	CSF fistula. Closed with dural patch	None
Microorganism isolated	Enterococcus	Staphylococcus aureus	Not identified	Enterobactercloacae + Streptococcus viridans	--
Age at FU	10 years 8 months	16 years 6 months	--	3 years 5 months	Lost to follow-up
Current status	Good, no deformity, full recovery	Good, no deformity, full recovery	Preoperative death	Good, no deformity, full recovery	Lost to follow-up

FU: Follow-up.

Regarding the associated pathologies, a dorsal intramedullary dermoid cyst was recognized (Figure 1). The associated congenital malformations were an infected syringomyelia with a tethered lumbar spinal cord (Figure 2) and a partial cervical medullary disconnection (See Figure 3). There were also 2 concomitant dysraphisms (Table 1): in one case with an epidural and intradural abscesscreating a meningitis profile (Figures 4, 5, and 6; Case 1),and in the other a lumbar epidural phlegmon that had not penetrated the subarachnoid space(Figure7).Four were treated surgically and received antibiotic treatment during a 2- to 3-month period (mean of 11 weeks). Three of these showed good long-term evolution with complete recovery (mean follow-up of 6 years [ranging from 6 months to 10 years]), while the fourth case, although without complications in the immediate postoperative period, was lost to long-term follow-up. One of the 5 patients died prior to surgery due to damage caused by meningitiswith pyoventriculitis.

The 12-week-old male fetus with lumbar DS and with a malformation associated witha cleft lip and palate was photographed in the laboratory of the institution both before and after the anatomical dissection. After dissection under high-resolution microscopy and prior marking of the anatomical faults, a 1x1 mm dermal orifice in the midline above the upper levelof the iliac crestswas immediately identified. Then, we identified a permeable path to the subarachnoid space with an associated dysraphismat level L5,at a depth of 1.5 mm from the skin to the arachnoid trabecula (Figure 8).

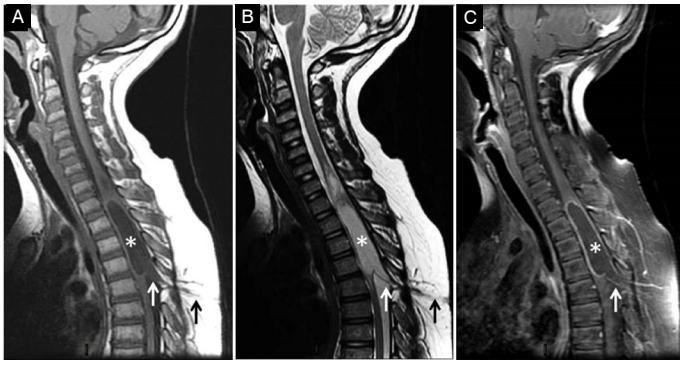

Figure 1. Sagittal Magnetic resonance images of the cervical and upper thoracic spine in a 14-year-old boy with DSCI in the dorsal region(Case 2). The DS tract in the subcutaneous (black arrows) and intradural (white arrows) cellular tissue with an intramedullary dermoid cyst (asterisk) can be identified. A. MRI in T1. B. MRI in T2. C. MRI in T1 with fat suppression and gadolinium.

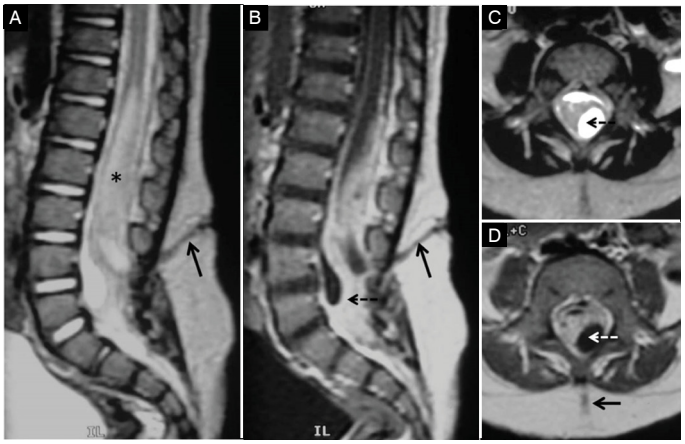

Figure 2. Magnetic resonance imagesof a 10-month-old boy with lumbar DS (Case 5). The DS tract is identified in the subcutaneous cellular tissue up to the dural sac (black arrows) associated with a tethered spinal cord and aninfected syringomyelia (asterisk and white arrow). A. Sagittal T2 MRI. B. Sagittal T1 MRI with gadolinium. C. Axial T2 MRI. D. Axial T1 MRI with gadolinium.

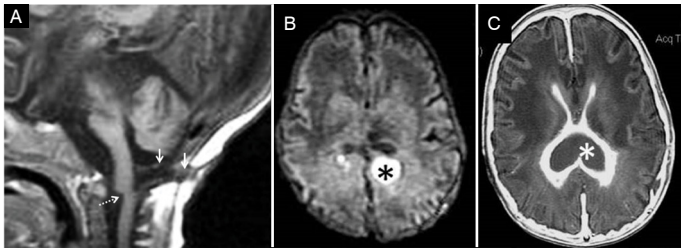

Figure 3. Magnetic resonance images of a 2-month-old girl with cervical DSCI (Case 3). We can identify the subcutaneous and intradural DS tract (white arrows) up to the posteriorside of the bulbo-medullary junction and the partial cervical medullary disconnection with an anterior notch (dotted arrow). In the brain images, the heterogeneous content of the lateral ventricleswith ependymal highlightingdue to pyoventriculitis (asterisks) is observed. A. Sagittal T1 MRI of the craniocervical region. B. Diffusion weighted axial MRI of the brain. C. Axial T1 MRI of the brain with gadolinium.

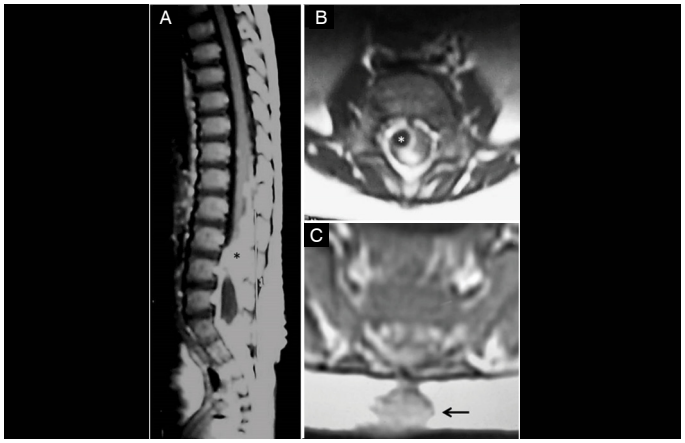

Figure 4. T1 magnetic resonance images with gadolinium of an eight-month--old girl with sacral DSCI at level S2 (Case 1). The DS tract is recognizablein the subcutaneous cellular tissue (arrows) with arachnoiditis and an intradural lumbar abscess (asterisk). A. Sagittal T1 MRI with gadolinium. B. Axial T1 MRI with gadolinium at lumbosacral-lumbar level 5. C. Axial T1 MRI with gadolinium at level S2.

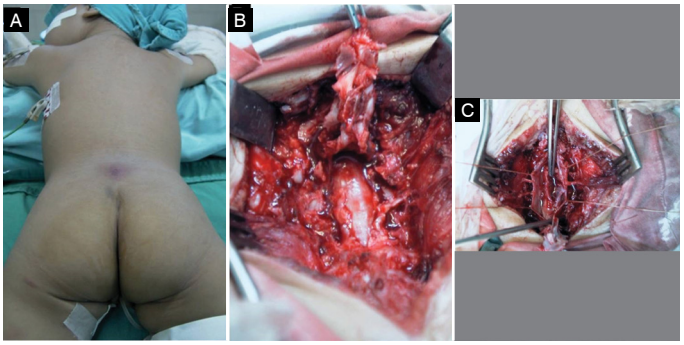

Figure 5. Clinical and intraoperativephotos of the patient (Case 1). A. The tract with clear signs of phlogosis. B. Intraoperative photo showing the area of the laminoplasty. Note the abnormal, bulging, and taut dural sac. C. Intraoperative photo of the durotomy and drainage of the intradural infection.

DISCUSSION

Dermal sinus (DS) is a rare dysraphism that results from the incomplete separation of the cutaneous ectoderm from the neuroectoderm between the third and eighth week of pregnancy.[1]The actual incidence of the condition is unknown,[1,7] being estimated at 1 out of every 2,500 live births,[3] although other authors estimate that it is found in up to 1.2% of newborns.[5] Clinically, it presentsas a dimple in the midline, as in our anatomical specimen (Figure 8A) or in a paramedian location in the lumbar, cervical, dorsal, or cranial region.

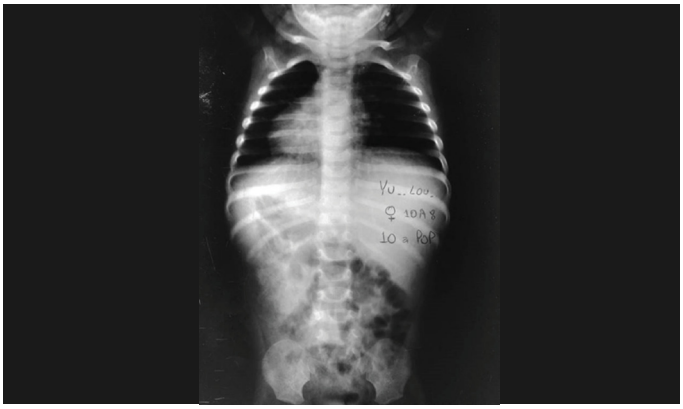

Figure 6. Frontal radiographof Case 1,10 years after surgery (post-laminoplasty, durotomy, and drainage).

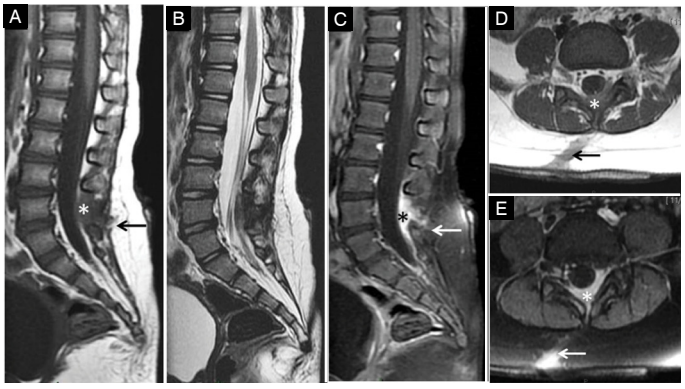

Figure 7. Magnetic resonance images of a 36-month-old girl with right paramedian lumbarDSCI at level L4 (Case 4). The DS tract is recognizable in the subcutaneous cellular tissue up to the epidural space (arrows) and an epidural phlegmon is highlighted with the contrast (asterisk). A. Sagittal T1 MRI. B. Sagittal T2 MRI. C. Sagittal T1 MRI with fat suppression and gadolinium. D. Axial T1 MRI. E. Axial T1 MRI with fat suppression and gadolinium.

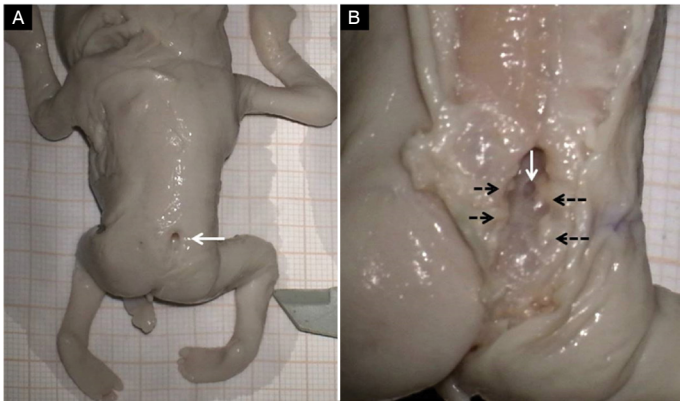

Figure 8. Photographs of a stillborn with lumbar DS. A. Photograph of a fetus at 12 weeks' gestation with permeable lumbar DS in the midline at level L5 (arrow). B. Communication with the subarachnoid space through a hole (white arrow) and the dysraphism (black dotted arrows) can be seen in the anatomical dissection.

They predominate in the lumbar region in 40-41% of cases and less frequently in the dorsal region (10%) and the cervical level (1%).[1,2] Rarely, they occur in a paramedian spinal location, as in one of the cases in our series (Table 1, Figure 7)and in other publications.[9]

DS may or may not be accompanied by cutaneous stigmata such as angiomas, hypertrichosis, skin tags, abnormal pigmentation, subcutaneous lipomas, or symptoms of local infection, like erythema and induration (Figure 5A).[1-3,5,10] In addition, it may be associated with inclusion tumors (dermoid, epidermoid, and teratoma), as in one of our cases (Table 1), or with other congenital malformations like diastematomyelia, tethered spinal cord, intradural lipoma, meningomyelocele, and lipomeningomyelocele (Figures 1 and 2).[1,2,5-7] Our anatomical specimen did not terminate in a dermoid or epidermoid tumor, and, although in the literature their association with DS varies from 30% to 50%,[3,5] the fact that it was found in only one of our cases, as well as the data from our stillborn, aligns better with the fact that only 60% of DSs terminate in one of these tumors.[5]

The DS tract is lined with scaly, multi-layered epithelium and it can extend from the deep fascia to the spinal cord. Around 60% of spinalDSs enter the subarachnoid space, as in some of our cases (Table 1) and in the anatomical specimen (Figure 8B), and 27% of these are attached to the cone, the cauda equina, or the filum terminale. The fact that allof our cases (Table 1) and the anatomical dissection of our stillborn specimen presented bone dysraphisms shows their strong association with DSs that end at the intradural level.[1] In any case, in 10-20% of cases it canend in a blind manner in the epidural spaceand in the subcutaneous cellular tissue in the others.[1,2,10,11] Due to the ascent of the spinal cord in relation to the greater longitudinal development of the spine during the fetal stage, the DS may pass through several levels within the epidural space before entering the subarachnoid space, following an ascending cephalic path, as observed in Case 1 (Table 1, Figure 4).

Diagnosis

Spinal DSs usually present in one of 5 ways:[10] (1) as a visible cutaneous tract; (2) as an central nervous system infection (meningitis, intra-spinal abscesses, etc.); (3) as aseptic meningitis (Mollaret's meningitis) resulting from the irritation produced by the desquamation of the epithelial cells of an associated dermoid or epidermoid cyst; (4) as neurological deterioration from medullary compression due to the growth of the associated cyst; and (5) as neurological deterioration from the tethering of the spinal cord. These last two forms of presentation are reported less frequently,[5] however, 2 of our 5 cases demonstrate the importance of keeping them in mind (Table 1, Figures 1 and 2).

Among the differential diagnoses ofspinal DSs are the pilonidal cyst andthe sacrococcygeal sinus, present in 2 to 4% of newborns. The latter is generally shallow, although it can reach as deep as the periosteum of the coccyx and does not require imaging study.[7]

Another differential diagnosis is limited dorsal myelochisis, a dysraphism that may be located at any spinal level. Although there are different clinical presentations, the most characteristic is a closed cutaneous depression in the midline,covered with pearly skin and with a fibroneural tract that connects the depression to the spinal cord just above the cone.[12-14] In most cases it is visible via MRI[13]and, since, unlike DS, the tract does not have a lumen, the probability of infection is low.[12,13] It is not accompanied by inclusion tumors, although it may be associated with intradural lipomas.[12-14]

Imaging studies of DS in children can begin with ultrasound, especially during the first year of life, and it is also permits the diagnosis of associated abnormalities, such as tethered spinal cord, inclusion cysts, lipomas, and syringomyelia.[15-17] But contrast--enhanced MRI is the method of choicefor the studying DSCI since it allows the identification of the path, the infectious complications, and the congenital anomalies associated with the condition.[18] Complete visualization of the DS tract is not always possible with MRI,[1,5] but in our series, as in that of Lane et al.,[19] it was identified in all cases (Table 1). Dermoid and epidermoid cysts that may be associated have a variable signal in MRI.[5]

Infectious complications

DS infection is the most serious complication for its high morbidity and resulting sequelae.[10] Infectious complications can occur in any part of the DS, from the opening in the skin to the nervous system. They include cellulitis, meningitis, intramedullary, cerebral, cerebellar, and subdural abscesses, pyoventriculitis, and infected inclusion cysts.[11,20,21] It has been reported that up to 48% of the patients reviewed may have infectious complications.[22] It is worth

noting thatDSCI of the infected pilonidal cyst, which is more distal, is usually located at the tip of the coccyx.[5]

MRI findings can be misleading in DSCI patients, since adhesions and inflammation of the cauda equina roots may mask an intradural lesion.[23] However, infected dermoids may exhibit an intense peripheralhighlight in the internal image of the abscess from the contrast agent (Figure 1).[24]

The bacteria found are diverse, among them we find *Escherichia coli*, *Staphylococcus aureus, Streptococcus viridans, Proteus*and*Klebsiella*;[5,7,11] but in the lumbosacral area fecal or cutaneous microorganisms are generally involved,[5] as was the case in 2 of our 3 lumbar cases (Table 1).

Treatment

The treatment of DS is surgical. Even if the MRI does not reveal any associated pathology, the tract must be removed,[1,5,10,25] even if it does not extend to the interior of the spinal canal.[3,7] Although classically it is said that when complicated it is better to wait for the infection to heal before performing surgery,[1,5] we disagree with this position and agree with those who support the need for urgent intervention.[11,18,20] This case series shows the importance of early surgery. Threeof the 4 operated patients had very good follow-up results and only 1 had an immediate postoperative complication that was fully resolved with a reintervention (Table 1) involving an extensive surgery that included the drainage and toilette of all the infected areas (skin, cellular tissue, the musculoaponeurotic layer, the epidural space, the subdural or intradural space, spinal cord, and inclusion cyst) is mandatory(Table 1, Figure 4 and 5).[11,18,20] Taking a sample during surgery for identification of the causal agent(s) is imperative,[11]and the resolution of the associated condition, as well as the repair of the dysraphism, desirable.[18]

We do not agree with the use of laminectomy in these cases in the pediatric population,[5,11,20] and we prefer laminoplasty in children as it is less likely to produce deformities in the long term (Figures5 and 6,Table 1).In cases where laminectomy is imperative for an adequate resection of the associated lesion and surgical debridement, localized instrumented arthrodesis is necessary to prevent future deformities (Case 2, Table 1).

Surgical resolution must be unfailingly accompanied by prolonged antibiotic therapy.[5,11] Our operated patients were all treated for a minimum of 8 weeks.

The surgical results aretypically good, even in cases of spinal cord or intradural infections (Figures 4, 5, and 6; Table 1).[5,11,18,20,26]

CONCLUSIONS

DSCI must be studied with MRI, since the method provides informationadditional to the simple observation of the entire DS tract, identifies the infectious processes of the central nervous system, the associated malformations, the inclusion tumors, and enables its differentiation from other entities. Once diagnosed, DSCI should be urgently treated surgically, and culture-guided antibiotic therapy should be prolonged.

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