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REVIEW ARTICLE

Severe bacterial meningitis due to an enterothecal fistula in a 6-year-old child with Currarino syndrome: evaluation of surgical strategy with review of the literature

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Abstract
Meningitis is a rare but serious complication in patients with Currarino syndrome. We present a 6-year-old girl with a fulminant meningitis due to an enterothecal fistula involving the anterior sacral meningocele. Initial treatment consisted of broad-spectrum intravenous antibiotic therapy and laparoscopic construction of a deviating double-loop ileostomy. This was followed by an elective posterior neurosurgical approach with a sacral laminectomy, evacuation of the empyema, and securing the disconnection of the anterior meningocele from the thecal sac, 10 days after initial hospital admission. The girl made a good postoperative recovery. The treatment strategy in the setting of meningitis due to an inflamed anterior meningocele is discussed and the available literature on the topic is reviewed.

Keywords Bacterial meningitis · Currarino syndrome · Enterothecal fistula

Introduction

In 1981, Guido Currarino et al. were the first to describe a triad consisting of (1) partial sacral agenesis; (2) presacral mass (anterior meningocele, enteric cyst, teratoma); and (3) anorectal malformation/stenosis [10]. There are no generally accepted guidelines about the indication and timing of surgical correction of an anterior sacral meningocele. Neither are there known risk factors predicting which patients with Currarino syndrome are prone to develop meningitis due to an enterothecal fistula. It is also unknown whether there is a relationship between (increase of) the size of the anterior meningocele and the chance of developing an enterothecal fistula. Different surgical approaches (anterior/posterior/sagittal) to close and resect the meningocele have been reported in the literature [25]. Here, we present our experience with a young patient with Currarino syndrome suffering from meningitis due to an enterothecal fistula and give an overview of the available literature on the topic.

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Case report of an illustrative patient

A 6-year-old girl presented with headache, drowsiness, and opisthotonus. She had been diagnosed with familial Currarino triad with associated constipation and micturition problems. Repeated lumbosacral MRI scans over the years had revealed slight increase of the anterior meningocele (see Fig. 1). Four days before admission, the patient was treated with antibiotics in another hospital because of a suspected urinary tract infection. At presentation in the emergency room, she was drowsy (GCS 3-5-2) with severe opisthotonic posturing (see Fig. 2a), but without focal neurological signs. Her body temperature was 36.6 °C. Blood leucocyte count and C-reactive peptide were 23.2×10^9 /L and 214 mg/L, respectively. Analysis of the cerebrospinal fluid (CSF) revealed a pleocytosis with 6659×10^6 /L cells and glucose < 0.1 mmol/L. Antibiotic treatment was immediately started and consisted of intravenous ceftazidime and metronidazole for 6 weeks. Culture of the CSF rendered *Streptococcus anginosus* (milleri) and *Bacteroides fragilis*. Because of the multimicrobial culture and her medical history, an enterothecal fistula was suspected. Gadolinium-enhanced MRI of the lumbosacral region

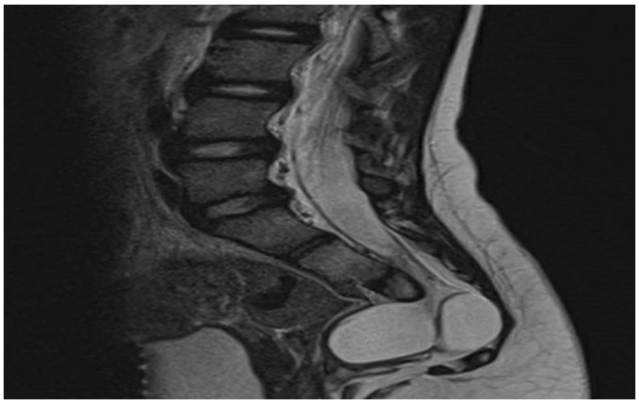

Fig. 1 Sagittal T2–weighted MRI shows the anterior sacral meningocele during follow-up, approximately 1 year before the patient developed meningitis

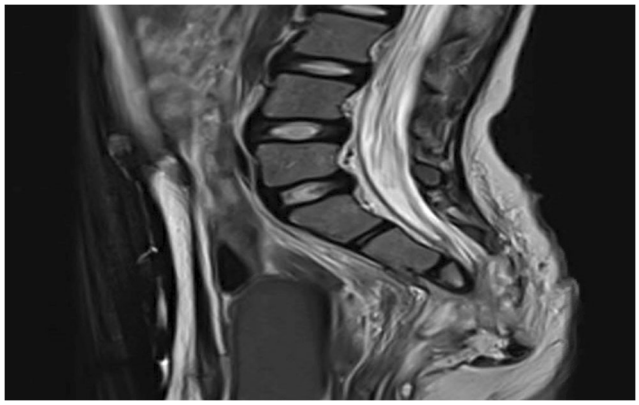

Fig. 3 Postoperative sagittal T2–weighted MRI shows obliteration of the anterior sacral meningocele after neurosurgical exploration and evacuation of the empyema

revealed inflammation of the anterior meningocele with abscess formation (see Fig. 2b).

Multidisciplinary rounds with pediatric neurologists, pediatric surgeons, pediatricians, microbiologists, and neurosurgeons resulted in the decision to first perform a laparoscopic deviating double-loop ileostomy in the acute stage, in order to stop the inflow of enteral commensals in the CSF space and inflamed retroperitoneal and epidural area. A few days after the formation of an ileostomy, a contrast study of the rectal stump confirmed leaking of contrast through a rectal fistula to the area of the anterior meningocele. Ten days after hospital admission, a posterior sacral laminectomy was performed with evacuation of a large amount of empyema and debris from the anterior meningocele and the region around the sacral roots. There was severe fibrosis in the operating field. The connection between the anterior meningocele and thecal sac had closed spontaneously due to inflammation. No patent fistula was found.

After uncomplicated surgery, the clinical course was dominated by the aftermath of the fulminant meningitis. Postoperative MRI confirmed obliteration of the anterior sacral meningocele/enterothecal fistula and a decline of inflammation (see Fig. 3). Initially, the ventricular system was dilated, which was managed by intermittent lumbar CSF-taps. No internal CSF shunt was needed as the hydrocephalus resolved after recovery from the meningitis. Successful restoration of the ileostomy was performed several months later. The girl made a good physical and neurological recovery.

Review of the literature

In an attempt to collect data supportive for any specific strategy or approach, we performed a literature search on the topic of Currarino syndrome and meningitis in the databases of PubMed, Web of Science, and Embase, using the following search terms: [currarino], [meningitis], [anterior meningocele], and [inflammation]. The search rendered 37 publications describing 38 patients (Table 1). All were single case studies, except for one article describing two young infants suffering from the condition. The series comprises 17 pediatric patients and 20 adult

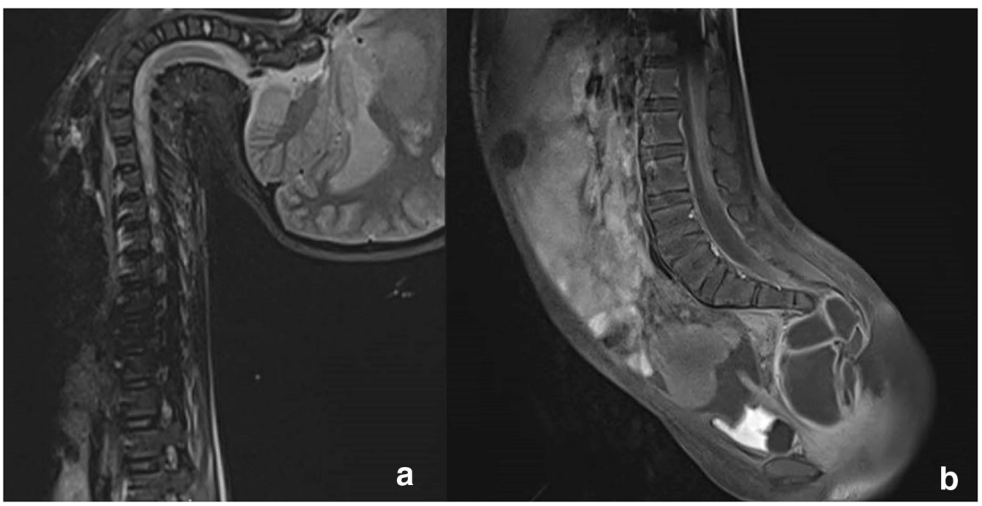

Fig. 2 **a** Sagittal T2–weighted MRI shows severe opisthotonic posturing at presentation in the emergency department. **b** Sagittal T1–weighted MRI with gadolineum shows enhancement of the anterior sacral meningocele with the formation of several inflamed empyema pockets

Table 1 Overview of the articles describing patients with Currarino syndrome/anterior sacral meningocele who developed meningitis

Author	Age of the patient(s) and sex (M/F)	Brief summary	Microbial pathogen(s)	Information on surgical approach/strategy and timing of surgery
Braczynski et al. 2017 [7]	71 years (F)	Meningitis because of an infected anterior meningocele in a patient suffering from colorectal carcinoma.	*Fusobacterium nucleatum*	Posterior approach. Laminectomy S1–3 with closure of the thecal sac and amputation of the meningocele. Surgery approximately 2 weeks after the presentation with meningitis.
Al Qahtani et al. 2016 [1]	14 months (F)	Passing away of an infant with Currarino syndrome because of a sacral fistula causing meningitis.	*Extended-spectrum beta-lactamases Escherichia coli*, *Enterococcus faecium*	No surgery performed.
Paul et al. 2015 [32]	30 years (F)	Pyogenic meningitis in a patient with an anterior meningocele	*Enterococci*	N/A
Morgenstern et al. 2015 [28]	9 days (1st episode of meningitis) and 7 months (2nd episode of meningitis) (M)	The article describes 4 patients with recurrent meningitis. One of these patients had a fistula and Currarino syndrome.	*1st episode of meningitis Streptococcus bovis* *2nd episode of meningitis Enterococcus faecium, Klebsiella pneumonia, Escherichia coli, Citrobacter freundii, E. faecium*	The CSF fistula was surgically repaired NOS. Timing of surgery not specified.
Ganeshalingham et al. 2014 [17]	8 weeks (M)	Passing away of an infant due to polymicrobial (enteral) meningitis. Strong suspicion of Currarino syndrome, but no post-mortem investigation performed.	*Escherichia coli, B. fragilis*	No surgery performed.
Patnaik et al. 2013 [31]	8 months (M)	Meningitis in a patient with an anterior sacral meningocele and epidural abscess	*Staphylococcus aureus*	Posterior approach. Laminectomy L5–S3. Drainage of the epidural abscess and intradural closure of the meningocele with a fascial patch. Timing of surgery not specified.
Monseaux et al. 2013 [27]	29 years (F)	Presentation with paraplegia in the context of a meningeal infection in a patient with an anterior meningocele	*N/A*	N/A
Kansal et al. 2012 [21]	45 years (F)	Recurrent aseptic meningitis in a patient with an epidermoid tumor in her anterior meningocele.	*Aseptic meningitis*	Posterior approach. Sacral laminectomy. The dura was opened in the midline. An anterolateral dura defect was closed by direct suturing. Timing of surgery not specified.
Calleja Aguayo et al. 2012 [8]	8 months (M)	Recurrent meningitis in a patient with Currarino syndrome and an anterior sacral meningocele and a rectal fistula.	*Streptococcus bovis, Enterococcus faecium, Escherichia coli, Klebsiella pneumoniae*	Mass excision, complicated by a recto-cutaneous fistula NOS. A colostoma and VP shunt insertion was necessary. Timing of surgery not specified.
Antuna-Ramos et al. 2011 [2]	10 years (M)	Meningitis due to a pararectal abscess with connection to the anterior sacral meningocele	*Bacterial meningitis NOS*	Posterior approach. Timing of surgery not specified.
Koksal et al. 2011 [23]	44 years (F)	Meningitis in a previously healthy woman with an anterior sacral meningocele	*Escherichia coli*	Posterior approach. Sacral route. Excision of the fistula tract. Repair of the sac orifice with sutures and fibrin glue. Surgery performed on the 4th day of hospital admission.
Raczynski et al. 2010 [35]	5 months (F) 2 months (F)	Meningitis caused by a fistula in two young patients with Currarino syndrome.	*Escherichia coli and Proteus mirabilis (5-month-old patient) and Pseudomonas aeruginosa (2-month-old patient)*	Surgical repair NOS. Timing of surgery not specified.
Bahtia et al. 2010 [5]	9 years (F)	Meningitis in a child with multiple occult spinal dysraphism stigmata, among which an anterior meningocele, a dermal sinus tract, caudal regression syndrome, and tethered spinal cord. After surgery, a second episode of meningitis occurred.	*Klebsiella pneumoniae, Streptococcus species*	Posterior approach. Laminotomy L5 and intradural exploration. Disconnection of the pyogenic sac and the thecal sac and sectioning of the fatty filum terminale. Surgery performed after 3 weeks of intravenous antibiotic treatment.

Table 1 (continued)

Author	Age of the patient(s) and sex (M/F)	Brief summary	Microbial pathogen(s)	Information on surgical approach/strategy and timing of surgery
Simon et al. 2010 [38]	30 years (F)	Meningo-encephalitis and meningo-myelitis in a patient with Currarino syndrome and an anterior sacral meningocele, a dermoid cyst, and tethered spinal cord.	*N/A*	A surgical intervention with aspiration and ligation of the anterior meningocele and tethered spinal cord release NOS. Timing of surgery not specified.
Bergeron et al. 2010 [4]	40 years (F)	Ascending meningitis and cauda equina syndrome caused by a rectal-thecal fistula in a patient with Currarino syndrome.	*Escherichia coli, group F streptococci, Bacteroides fragilis, Peptostreptococcus anaerobius, Candida glabrata*	Anterior approach. Removal of the meningocele with a trans-abdominal approach. Closure of the sacral deficit by suturing a strip of well-vascularized omentum and fibrin glue. Surgery on the day of hospital admission.
Kiefer et al. 2009 [22]	20 days (F)	Treatment-resistant meningitis (CSF pleocytosis) in a patient with Currarino syndrome and a rectal-thecal fistula proven by myelography.	*No pathogen isolated from CSF and blood cultures*	Posterior approach. Sacral laminectomy and repair of the anterior dural defect with fibrin glue and muscle graft. Timing of surgery not specified.
Sanchez et al. 2008 [36]	64 years (M)	Bacterial meningitis in a patient with an anterior sacral meningocele.	*Enterococcus faecalis, Escherichia coli*	Anterior approach. Laparotomy. Resection of a portion of the rectum and the meningocele. A terminal colostomy was performed. Timing of surgery not specified.
Miletic et al. 2008 [26]	39 years (M)	Meningitis and a large anterior sacral meningocele.	*Escherichia coli*	Posterior approach. Laminectomy with dural opening. Obliteration of the communication of the intrathecal compartment and the meningocele. Timing of surgery not specified.
Fitouri et al. 2007 [12]	3.5 years (F)	Repetitive meningitis in a patient with Currarino syndrome including a mature teratoma	*Escherichia coli, Streptococcus B, Haemophilus influenzae*	Surgical curation of the presacral cystic mass NOS. Timing of surgery not specified.
Fleury et al. 2007 [15]	29 days (F)	Multiple family members with Currarino syndrome. A patient with meningitis and Currarino syndrome with a mature teratoma is described.	*Escherichia coli, Bacteroides*	Posterior approach. Repair by way of a sagittal approach. Surgical treatment 21 days after antibiotic treatment/meningitis.
Hatano et al. 2006 [20]	46 years (F)	Marfan syndrome and incomplete Currarino triad, presenting with recurrent meningitis and an anterior sacral meningocele.	*N/A*	Surgical approach was limited to plasty of the meningocele NOS. Timing of surgery not specified.
Phillips et al. 2006 [33]	48 years (M)	Meningitis due to a rectal-thecal fistula in a patient with an anterior sacral meningocele	*Anaerobic gram-negative bacillus*	Anterior approach. Laparotomy was performed. The neck of the meningocele was oversewn. Coverage of the defect with omentum. Timing of surgery not specified.
Schijman et al. 2005 [37]	1 month (F)	The patient developed multibacterial meningitis at the age of 1 month. At the age of 3 months, she developed paraplegia due to an intramedullary abscess.	*Pseudomona, Proteus, Escherichia coli, Aerobacter*	Posterior approach. Sacral laminotomy. A cystic teratoma was removed. Watertight closure of the spinal canal with an aponeurosis patch graft. Surgical treatment of the anterior meningocele was performed 3 weeks after the presentation with paraplegia.
Emans et al. 2005 [11]	N/A	Expression patterns of Currarino syndrome are described. In this article, one patient with meningitis is mentioned.	*N/A*	Operative treatment NOS. Timing of surgery not specified.
Bal et al. 2004 [3]	35 years (F)	(Possibly iatrogenic) infected anterior meningocele after transrectal puncture.	*N/A*	Posterior approach. Sacral laminectomy. The neck of the anterior sacral meningocele was tied off. Timing of surgery not specified.
Haga et al. 2003 [19]	58 years (F)	Patient with recurrent meningitis and Currarino triad (with intradural epidermoid cyst).	*Coryneform bacteria*	Posterior approach. Sacral laminectomy. Opening of the dura. Neck ligation was performed. Timing of surgery not specified.

Table 1 (continued)

Author	Age of the patient(s) and sex (M/F)	Brief summary	Microbial pathogen(s)	Information on surgical approach/strategy and timing of surgery
Chou et al. 2002 [9]	3 months (M)	Infant with meningitis and Currarino syndrome.	*Bacterial meningitis NOS*	N/A
Guerin et al. 2000 [18]	23 years (F)	Polymicrobial meningitis (meningitis after a gynecologic puncture) leads to discovery of an anterior sacral meningocele.	*Enterococcus faecalis, Prevotella bivia, Streptococcus constellatus*	Communication between the endodural lumen and the meningocele on the S2 level was closed with adipose tissue and biological glue NOS. Surgery on the 22nd day of hospital admission.
Fitzpatrick et al. 1999 [13]	31 years (F)	The patient developed meningitis after a diagnostic laparotomy as part of the investigation of her infertility. A presacral mass was found, but not further explored at that time. Surgery was performed 18 years later because of mucopurulent rectal discharge.	*Escherichia coli, Bacteroides*	Posterior approach. A sacral approach from the midsacrum to the anal margin was used. The neck of the meningocele was ligated. The sacral defect was repaired using adjacent fascia. The surgery was planned in elective setting.
Tamayo et al. 1999 [40]	24 years (M)	Patient with multibacterial (enteral) meningitis and Currarino syndrome.	*anaerobic Enterococci, Bacteroides fragilis, Escherichia coli*	N/A
Funayama et al. 1995 [16]	4 months (1st episode of meningitis) and 1 year (2nd episode of meningitis) (M)	The patient died 1 month after his last hospital admission, due to severe meningeal infection and sepsis. Autopsy confirmed an anterior sacral meningocele and intraspinal abscess formation.	*Proteus mirabilis, Klebsiella pneumoniae*	No surgery performed.
O'Riordain et al. 1991 [29]	15 years (F)	Ten family members with Currarino syndrome. Meningitis described.	*Bacterial meningitis NOS*	N/A
Blond et al. 1991 [6]	7 years (F)	The patient developed polymicrobial meningitis after falling on her os coccygis. A small anterior sacral meningocele was discovered together with spina bifida occulta at L5 level and a scimitar sacrum.	*Streptococcus species, Bacteroides species*	Posterior approach. The fistula was ligated. Surgery performed during the third week of antibiotic therapy.
Page et al. 1990 [30]	27 years (M)	Meningitis due to rectal fistulation of the meningocele	*Stercoral flora NOS*	Curation after seven surgical procedures NOS. Timing of surgery not specified.
Fiumara et al. 1989 [14]	36 years (F)	Purulent meningitis in a patient with an anterior sacral meningocele	*Bacterium coli*	N/A
Synowitz et al. 1988 [39]	19 years (M)	The patient presented with an infected anterior meningocele	*Escherichia coli*	Posterior approach. Surgical closure of the meningocele was obtained using a dorsal transdural approach with sutures and fibrinous adhesive. Timing of surgery not specified.
Quigley et al. 1984 [34]	21 years (F)	Recurrent aseptic meningitis in a patient with an anterior sacral meningocele and a dermoid tumor	*Aseptic meningitis*	Posterior approach. Transsacral approach with a laminectomy L4 to S1. Extirpation of the dermoid tumor, detethering and obliteration of communication between the thecal sac and the anterior sacral meningocele. Surgery was planned in elective setting after recovery from the last episode of aseptic meningitis.

N/A not available, *NOS* not otherwise specified

patients (information on the age of one patient missing). The known female predominance of Currarino syndrome was confirmed with a F:M ratio of 24:13 (information on the gender of one patient missing). These case reports show that meningitis due to formation of an enterothecal fistula can be fatal [1, 16, 17]. In the series here described, a posterior surgical approach was chosen in 16 patients and an anterior surgical approach was chosen in 3 patients. For 16 patients, the surgical technique was not described in detail. In 3 cases, no surgery was performed (these 3 cases related to the three deceased patients in the series). Concerning the timing of surgery in the setting of meningitis and an infected meningocele, only 8 publications give adequate information. The timing of surgery ranged between day 1 and day 22 after hospital admission (and starting of antibiotic therapy), with a mean of 16 days and a median of 21 days before surgical correction of the fistula and the meningocele. For two patients, it was decided to plan the surgical repair in elective setting, after initial discharge home from the hospital when the meningitis was treated sufficiently (this included one patient with aseptic meningitis due to a dermoid tumor).

Discussion

In the case presented, it was decided to first perform an ileostomy, to be completely sure of a stop of leaking of intestinal microbial flora through the enteral fistula(s). This was followed by an elective posterior neurosurgical exploration. This seems to be an effective strategy. In the literature, we noticed the preference for a posterior approach in the situation of severe inflammation. This is in line with our experience in the case here described. In a posterior approach, important neurological structures are directly visualized and can be spared. A detethering procedure, for example cutting of the filum terminale, or removal of an intradural tumor (e.g., dermoid or teratoma), is conveniently possible during the same approach. A possible disadvantage is the suboptimal view on the rectum and the retroperitoneal/enteral anatomy. This is especially true in a situation of severe inflammation. In our experience, successful surgical closing of an enteral defect in the setting of active inflammation is not possible. Hence, a temporary ileostomy is an indispensable and elegant solution to overcome this problem. It is known from a significant body of literature, mainly from the GE-surgical field, that rectal fistula will close spontaneously if there is no passage of fecal material for some time because of an ileostomy [24].

The current available literature on the topic of meningitis, due to an inflamed anterior meningocele caused by an enterothecal fistula, is limited to case reports only. Therefore, evidence-based guidelines/protocols for Currarino patients developing meningitis due to an enterothecal fistula cannot be formulated. There is no high-quality literature on the natural history of Currarino patients with an anterior sacral meningocele to justify the prophylactic surgical correction of an anterior meningocele in all Currarino patients, solely aiming to prevent meningitis. Numbers needed to treat (NNT) to prevent one case of meningitis are unavailable. If the patient experiences other clinical symptoms that could be alleviated by surgical correction of the anterior sacral meningocele, this would of course justify a more aggressive surgical strategy towards closure and resection of the meningocele.

Conclusion

The present case and review of the literature illustrates that in patients with Currarino syndrome potentially lethal meningitis can occur due to the development of an enterothecal fistula. In our own limited experience and supported by the literature, the construction of an ileostomy in the acute stage seems a safe and rational start of the (surgical) treatment, together with administration of high-dose, broad-spectrum intravenous antibiotics. Subsequent surgical treatment of the enterothecal fistula and infected anterior sacral meningocele can be performed in an elective procedure, as soon as the patient has recovered from the most severe symptoms of the meningitis. A posterior approach is most often described in the literature, and seems to offer the best anatomical overview in the setting of (recent) inflammation. There is currently no supportive evidence for early prophylactic surgery in Currarino patients with an anterior sacral meningocele to prevent meningitis.

Compliance with ethical standards

Conflict of interest On behalf of all authors, the corresponding author states that there is no conflict of interest.

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