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Long-Term Recurrent Intramedullary Arachnoid Cyst: Case Report and Literature Review

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Abstract

Objectives This article reports the management of a case of a 32-year-old male who presented with progressive weakness in the lower limbs and spastic paraparesis secondary to an intramedullary arachnoid cyst (IMAC). For literature review, the authors used the phrase "intramedullary arachnoid cyst" in PubMed search engine. 23 articles describing cases with IMAC were included in this review, with a total of 26 patients.

Materials and Methods We report a case with long term recurrant intramedullary arachnoid cyst and present a review on spinal intramedullary arachnoid cyst.

Result IMAC is showing bimodal incidence and trending to occur below 10 years and after 30 years. However, rarely, it should be considered in the differential diagnosis of intramedullary cystic lesions. Authors suggest doing laminoplasty or fusion for the pediatric patients to prevent kyphoscoliosis deformity in the long run, but doing early surgery to gain better outcome. Resection of the cyst wall should be done as much as possible; if it could not be achieved, then marsupialization or cysto-subarachnoid shunt should be considered. Aspiration alone or fenestration is not enough to eradicate the cyst. Long-term and prospective studies are recommended to achieve the best treatment options.

Conclusion Review supports early surgical treatment of symptomatic IMACs with resection of the cyst wall as much as possible.

Keywords
- intramedullary arachnoid cyst
- intramedullary lesion
- spinal surgery
- fenestration
- marsupialization

Introduction

Arachnoid cyst is a fluid-filled sac that can present in brain or spinal cord. Spinal arachnoid cysts can be extradural, intradural, or intramedullary.[1] Intramedullary arachnoid cysts (IMACs) are rare and more common in the thoracic spinal cord.

Since its first report by Aithala et al in 1999,[2] 26 cases have been reported sporadically around the world until the

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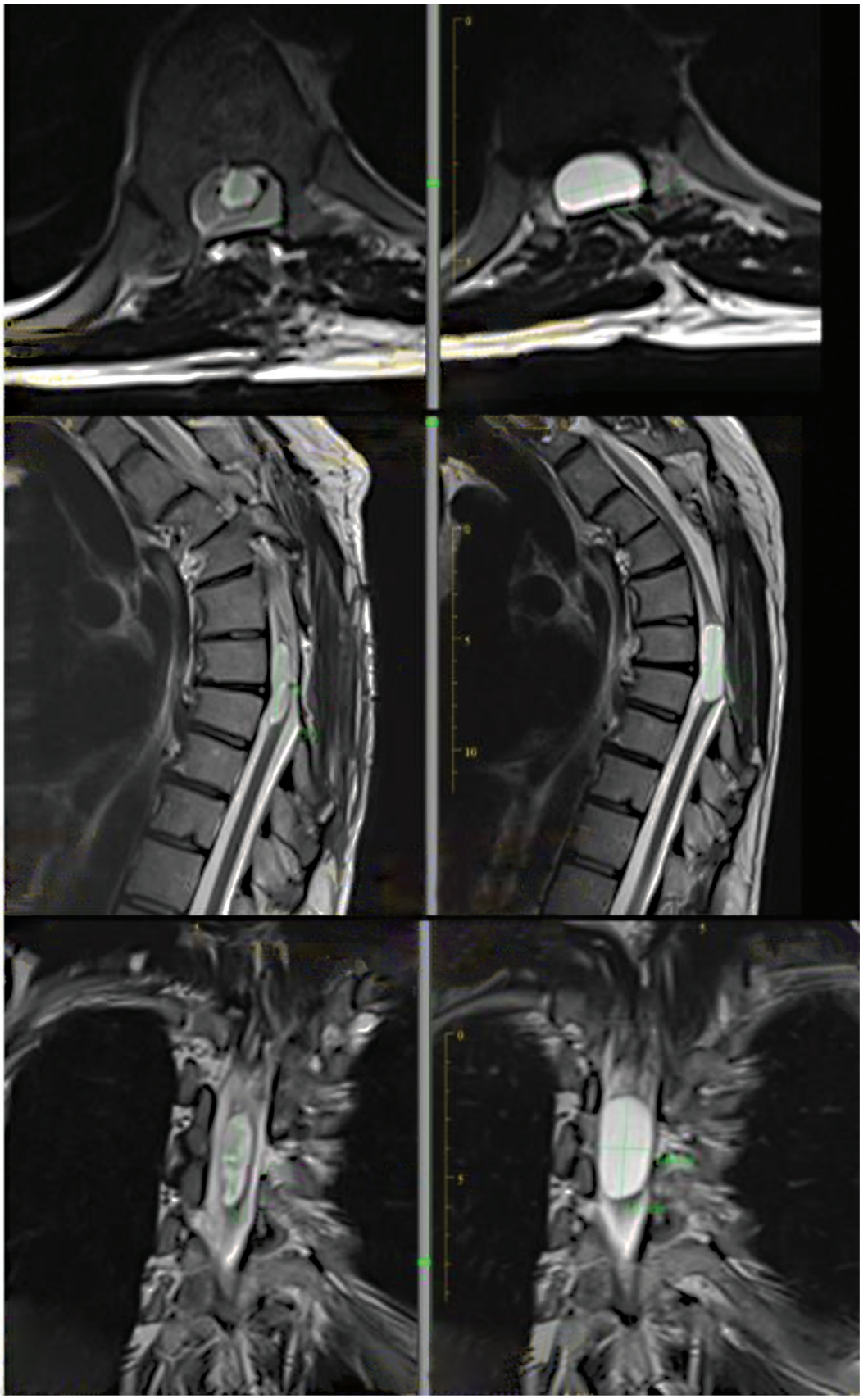

Fig. 1 Comparative axial, sagittal, and coronal T2-weighted magnetic resonance images before and after the latest surgery, associated with increased thoracic kyphosis on sagittal view.

present, with the common scenario of a deteriorating neurological status followed by the description of a cystic lesion in the spinal cord on magnetic resonance imaging (MRI). Surgery was done for all cases with only 2 cases reporting recurrence after 11 and 27 months.

This article also describes a case of a 32-year-old male who presented with progressive weakness in the lower limbs and spastic paraparesis secondary to an IMAC, which was detected on MRI and confirmed with histopathology study. Cyst wall fenestration and partial wall excision were performed. Postoperatively, the patient improved gradually.

Illustrative Case

A 32-year-old male presented with a history of paraparesis, when he was around 2 years old (1989), of unknown cause.

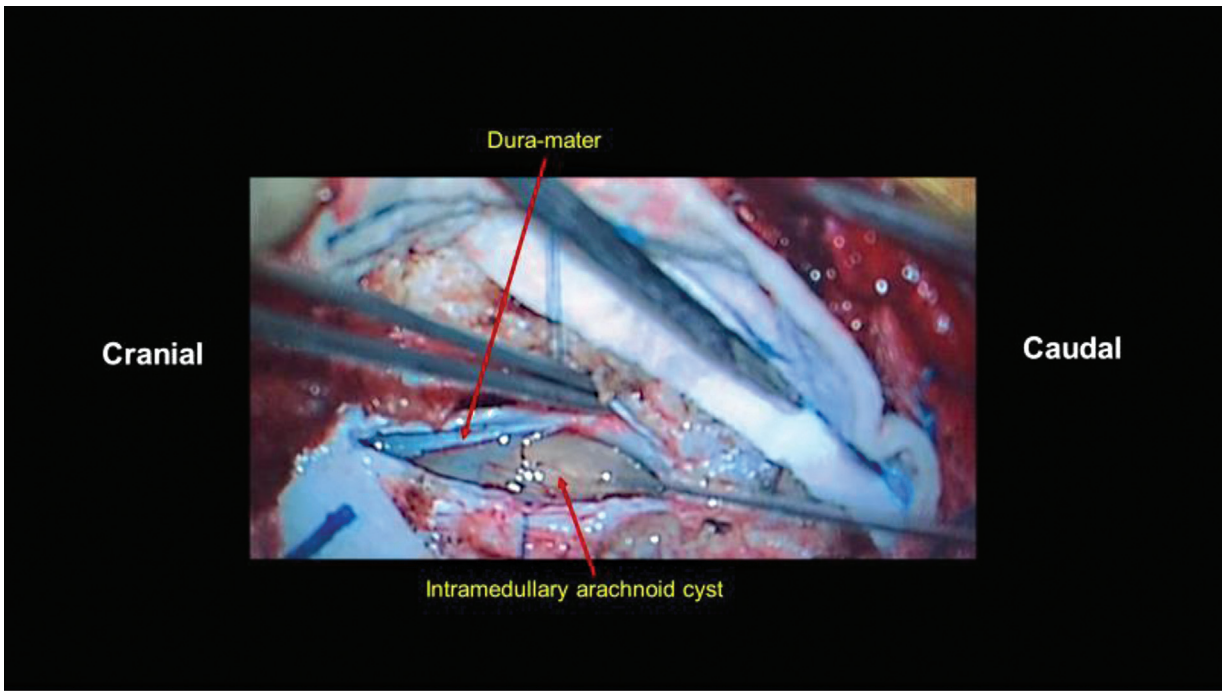

Fig. 2 Intraoperative image showing the posteriorly exposed T6-T7 intraspinal region, microscopic view of the microsurgical median myelotomy, and visualization of the intramedullary arachnoid cyst; a thin membrane was observed before the fenestration, for drainage of the cyst, biopsy, and partial removal of this cyst wall (April 2020).

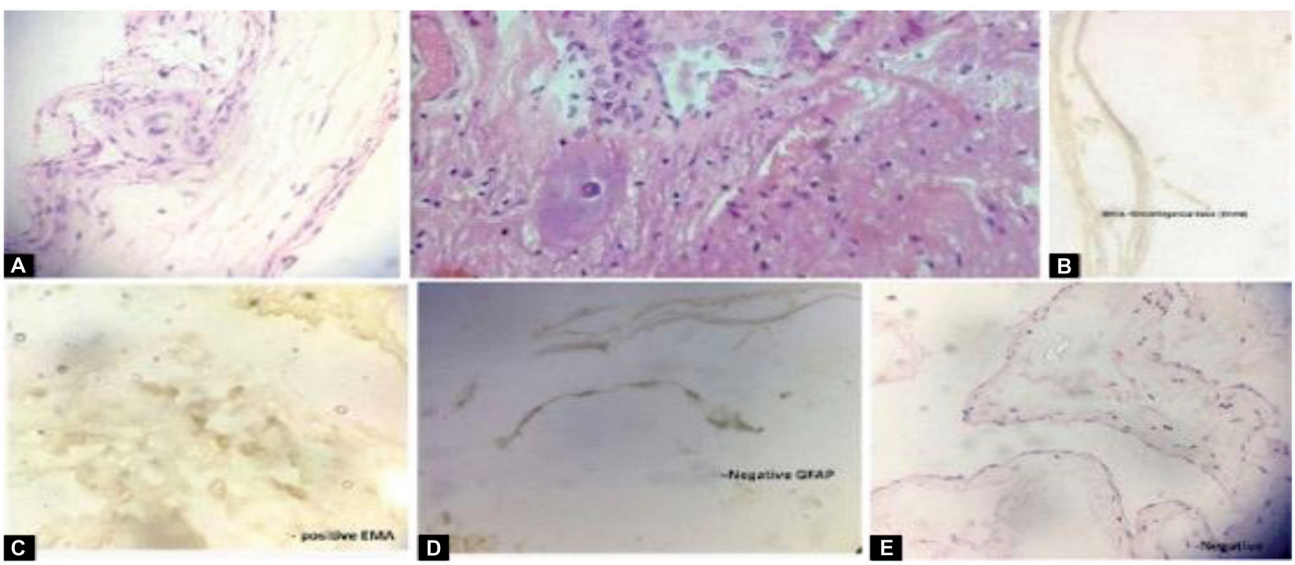

Fig. 3 Immunohistochemistry of intramedullary cyst (**A–E**) The linings of epithelial cells express epithelial membrane antigens. Negative control slide for S100, P53, carcinoembryonic antigen, and glial fibrillary acidic protein does not express target antigen (immunoperoxidases against epithelial membrane antigens are from Biogenix, ×10).

Then he had gradual spontaneous recovery of his initial neurological deficit. When he was around 10 years old, he presented with back pain and recurrence of weakness in his lower limbs, and he underwent a posterior thoracic decompression (1997), (unfortunately, the exact pathology and surgical details are not available), with partial recovery of his lower limbs' functions. The patient recovered relatively well, but his neurological deficit started to deteriorate again in 2012, getting worse with associated sphincter disturbance in 2016, compatible with neurogenic bowel (constipation) and bladder (partial urinary retention).

In February 2020, he came to our hospital, seeking specialized neurosurgical treatment, with no other relevant associated diseases. He had spinal examination revealing thoracic localized hyperkyphosis and mild scoliosis near a previous midline surgical scar. Motor exam was showing a spastic paraparesis (grade 3 right and grade 4 in his left lower limb), and he was walking with support. Lower limbs' deep tendon reflexes were hyperactive, with bilateral Babinski and clonus at his feet. Sensory examination showed hypoesthesia to pinprick and touch below T6 with spared sacral sensations.

A detailed clinical and themaging review of his case was performed. New MRI of the thoracic spinal region was showing a large well-defined thoracic intramedullary cyst (no enhancing intramedullary cystic lesion, hyper intense on T2-weighted image, and low-signal intense on T1 spin echo within the spinal cord and causing focal cord expansion) between T6 and T7, associated with increased thoracic hyperkyphosis, suggestive of IMAC (►**Fig. 1**).

It was hypothesized that he had a rare T6-T7 recurrent IMAC with myelopathy and gradual neurological deterioration. In April 2020, surgery was performed under general anesthesia in a prone position on the spinal table using intraoperative neuromonitoring and a lumbar drain shunt inserted to prevent postoperative cerebrospinal fluid (CSF) leakage. Adopting T6-T8 posterior midline approach, removal of a small portion of the remaining posterior elements at the level T6-T8 to expose the dura-mater was achieved. Microscopic midline durotomy was performed. At this moment, a localized bulging of the spinal cord related to the intramedullary cyst was observed. There was no shunt or suture (marsupialization), so we hypothesized that aspiration or fenestration/partial wall excision was done in the first surgery. A microsurgical median myelotomy was done, visualizing the IMAC (►**Fig. 2**). Aspiration of a clear fluid from the intramedullary cyst was observed. A thin membrane was found, and fenestration of it was done for drainage of the cyst. Biopsy and partial removal as possible of this cyst wall was accomplished. A watertight dural closure was done and other surgical planes were sutured. The fluid and tissue samples of the cyst wall were sent to the pathology department, for confirming the diagnosis (►**Fig. 3**).

The patient had a good postoperative recovery and bed rest for 2 days; the lumbar drain was removed and he was discharged on the third postoperative day. He had a new MRI after surgery (June 2020). In comparison with the previous MRI (February 2020) (►**Fig. 1**), the new image showed signs of bilateral laminectomy from T6 to T8 and an intramedullary fluid-filled cystic cavity inside the spinal canal surrounded by a low-signal rim that had regressed in size, suggesting a postsurgical status with reduced thickness of the arachnoid cyst.

At the latest follow-up, he was presenting with a partial neurological recovery (grade 4 for both right and left lower limbs), and improving gradually in his walking capacity and sphincter disturbance. The patient traveled back to his home country (June 2021), but he is still in contact (online), assuring that he is clinically stable.

Discussion

In April 2023, a review of the literature published in PubMed was conducted using the term "intramedullary arachnoid cyst" and 26 articles (27, including the presenting case) describing the presentation and management of IMACs were included. A descriptive outcome is demonstrated in ►**Tables 1** and **2**.

A total of 27 patients were included in this review (including our case report). All cases were investigated by MRI and diagnosis confirmed by histopathology of the surgically collected tissue samples. The main surgical technique was a posterior spinal approach for laminectomy of the desired site, then microsurgical cyst evacuation, and then fenestration, followed by partial to complete excision of the

Table 1 Qualitative table

Characteristics	Main descriptions
Papers included	23 papers
Level of medical evidence	All level IV of medical evidence
Patients	$n = 27$ patients
Age	Bimodal incidence: Below 10 years = 13/27 (48%) Above 30 years = 13/27 (48%)
Gender	Male = 40.7% (11/27) patients Female = 59.3% (16/27) patients
Pathology	Intramedullary arachnoid cyst
Location	Cervical = 33.3% (9/27) Thoracic (T1-L2) = 63% (17/27) Lumbar (L2-S5) = 3.7% (1/27)
Follow-up	Mean = 13.8 months (16–50 months)
Surgical technique	Laminectomy = 100% (24/27) Hemilaminectomy = 3.7% (1/27) Laminotomy = 3.7% (1/27) Posterior spinal fixation = 4.34% (1/27) Median myelotomy = 85.2% (23/27) DREZ myelotomy = 14.8% (4/27) Marsupialization = 11.1% (3/27) Fenestration = 100% (23/23) Cyst wall partial resection = 100% (27/27)
Recurrence	After surgery = 14.8% (4/27)

Abbreviation: DREZ, dorsal root entry zone.

Table 2 Literature review of spinal intramedullary arachnoid cyst

Patient	Author	Age/ Sex	Time from deterioration to surgery	Symptoms	Location	Treatment	Outcome	Follow-up	Recurrence
1	Aithala et al (1999)[2]	7y/M	5 days	Severe pain in the abdomen; bladder and rectal disturbance; weakness in the lower limbs	T1	Median myelotomy Cyst wall resection	Complete Rapid and complete	Not available	No
2	Gilabert-Gonzáles et al (2001)[3]	1y/F	Not available	Weakness in the lower limbs	L1-S1	Total excision of the cyst	Complete	Not available	No
3	Goyal et al (2002)[4]	63y/F	Over years	Severe weakness in the lower limbs (bedridden); bladder and rectal disturbance	T9-L2	Laminectomy Median myelotomy Partial excision of the cyst	Improved	3 months	No
4	Sharma et al (2004)[5]	10y/F	Not available	Progressive quadriparesis	C4-T1	Median myelotomy Partial excision of the cyst	Marked improvement	1 month	No
5	Sharma et al (2005)[6]	4y/F	20 days	Inability to walk/stand; weakness in the upper limbs	C4-C6	Median myelotomy Cyst was decompressed, and most of the cyst wall was excised	Complete	17 months	No
6	Ghannane et al (2007)[7]	4y/M	15 days	Weakness in the lower limbs	T3-T4	Laminectomy Median myelotomy Partial cyst wall resection	Complete	6 months	No
7	Ghannane et al (2007)[7]	8y/M	1 month	Weakness in the lower limbs	T3-T4	Laminectomy Median myelotomy Posterior fenestration of the cyst	Good	8 months	No
8	Guzel et al (2007)[8]	7y/F	1 month	Severe weakness in the lower and upper limbs	C2-C4	Laminectomy Median myelotomy Partially excised with fenestration	Good	2 years	No
9	Gezici and Ergün (2008)[9]	35y/F	2 months	Weakness in the lower limbs; progressed to complete loss of power and 2 months of urinary incontinence	T5-T6	Laminectomy DREZ myelotomy Majority of cyst wall excision Fenestration of the remainder cyst wall	Good	3 years	No

(*Continued*)

Table 2 (*Continued*)

Patient	Author	Age/ Sex	Time from deterioration to surgery	Symptoms	Location	Treatment	Outcome	Follow-up	Recurrence
10	Lmejjati et al (2008)[10]	12y/F	15 days	Weakness in the lower limbs	T3-T4	Median myelotomy Marsupialization	Full recovery	4 months	No
11	Medved et al (2009)[11]	1y/M	Sudden	Bladder and rectal disturbance; inability to walk/stand	T5-T6	Hemilaminectomy DREZ myelotomy Marsupialization	Complete	1 month	No
12	Diyora et al (2010)[12]	45y/f	10 days	Severe pain; weakness in lower limbs; progressed to paraplegia with urinary and fecal incontinence.	T4-T5	Laminectomy Midline myelotomy Cyst wall partially excised	Improved	1.5 months	No
13	Kataria et al (2012)[13]	9y/F	1 year	Back pain; weakness in the lower limbs; bladder and rectal disturbance	Thoracolumbar	Laminectomy Median myelotomy Partial excision of the cyst	Complete recovery	6 months	No
14	Kataria et al (2012)[13]	40y/F	6 months	Weakness in the lower limbs; bladder and rectal disturbance	L1	Partial excision of the cyst wall	Good	1–6 months	No
15	Bond et al (2012)[14]	2y/M	Not available	Back and lower limbs pain	T10	Fenestration of the cyst	Good/ remission	1 month	No
16	Rahimizadeh and Soufiani (2013)[15]	58y/F	3 months	Weakness in upper and lower limbs	C6-T2	DREZ myelotomy Wide fenestration with partial excision of the cyst wall Cervicothoracic instrumentation from C3 down to T2 was done	Improved	3 years	No
17	Novegno et al (2014)[16]	31y/F	3 months	Back pain; weakness in the lower limbs; bladder and rectal disturbance	T11-T12	Median myelotomy Fenestration Partial excision of the cyst	Complete	2 years	No
18	Thakar and Hegde (2016)[17]	64y/M	1 month	Weakness in lower and upper limbs	C6–T1	Cyst wall excision	Not available	Not available	Not available
19	Alugolu et al (2016)[18]	54y/F	2 months	Weakness in the lower limbs; paresthesia	T8-T12	Laminectomy Partial removal of cyst wall	Good	Year	No

Table 2 (*Continued*)

Patient	Author	Age/ Sex	Time from deterioration to surgery	Symptoms	Location	Treatment	Outcome	Follow-up	Recurrence
20	Panwar et al (2019)[19]	40y/M	3 months	Numbness in bilateral lower limbs	T11-T12	Median myelotomy Incomplete removal of cyst wall was done	Pain relieved completely	12 months	Suggestive of recurrence after 11 months
21	Panwar et al (2019)[19]	45y/F	3 months	Paresthesia and spasticity in lower limbs	T9-T10	No available approach Cyst wall excision	Pain relieved completely	11 months	No
22	Shaaban et al (2019)[20]	32y/M	3 months	Bladder and rectal disturbance; impotence; repeated falling	T6-T8	Median myelotomy Partial excision of the cyst	Improved	50 days	No
23	Ichinose et al (2020)[21]	4y/M		Weakness in the lower limbs; bladder and rectal disturbance	C2-C3	First fenestration of the cyst Median myelotomy Second excision of the cyst	Good	32 months	Yes, after 27 months
24	Aljameely and Baeesa (2020)[22]	47y/F	5 weeks	Weakness in upper and lower limbs; bladder and rectal disturbance	C3-C5	DREZ myelotomy Cysto-subarachnoid shunt	Complete recovery	5 years	No
25	Diyora et al (2022)[23]	45y/F	Not available	Difficulty in walking	Craniocervical Brain to C7	After two failed trials of needle aspiration, cysto-subarachnoid shunt	Complete recovery	4 years	Yes, after 4 months, then No
26	Thakur et al (2021)[24]	2y/M	6 months	Decreased sensation over the gluteal region along with persistent dribbling of urine	T12-L1	Laminotomy Median myelotomy marsupialization	Improved	3 months	No
27	Present case	2y then 32y/M	Many years	Back pain and progressive paraparesis	T7-T8	Cyst drainage and fenestration Partial excision of cyst wall	Improved	30 months	Hypothesis, yes

Abbreviations: DREZ, dorsal root entry zone; F, female; M, male; y, year.

cyst wall. Patients experienced immediate improved neurological outcomes in almost all cases with sometimes complete resolution of the symptoms, especially on acute presentation followed by early surgery.

In general, these studies demonstrate bimodal incidence (below 10 years =13/27 [48%]; above 30 years = 13/27 [48%]), with slight predominance in females (59.2%) in contrast to males (40.7%). The most common location of an IMAC occurrence was the thoracic spine (T1-L1: 17/27 [63%]), especially thoracolumbar region (T9-L1: 8/27), and then midthoracic (T3-T6: 6/27), followed by cervical (9/27 [33.3%]). Clinical presentation depends mostly on the cyst location and its volume, mainly caused by compression of the spinal cord and nerve roots. Most patients would present with weakness correlated with the compressed segment distribution and radiation along with upper or lower motor neuron signs. Almost all patients reviewed had a comparatively long course of symptoms of vague back pain. However, they all shared the rampant deterioration in their neurological exam during a short period of time, which is mainly attributed to the IMAC expansion. Surgery was of paramount significance in all reported cases after deterioration. The four main steps of surgery used were shared among the most reported surgically treated cases starting with: posterior spinal approach, laminectomy of the desired spinal segment, median myelotomy to gain access to the cyst, and cyst fenestration with cyst wall partial to complete resection.

The trigger point for deterioration and cyst behavior is still unclear,[15] but if the patient is asymptomatic, a wait and periodic scan approach could be tried.[25] The timing of surgery following deterioration in symptoms and examination is important; this article review is showing better outcome with early intervention after deterioration.

Although posterior spinal laminectomy approach is the standard approach for most of the reviewed articles, laminectomy in pediatrics itself is a risk factor for spinal deformity, especially kyphosis, like the presenting case, and sometimes it can progress to serious complications. Hence the authors suggest fusion, hemilaminectomy, or laminoplasty for pediatric patients.[26,27]

The surgery of the cyst was performed by posterior midline myelotomy approach in most of the literature, especially with posterior and central lesions. However, dorsal root entry zone approach was used in three articles, especially with ventral and unilateral lesions. Both approaches have their advantages and disadvantages, and careful localization of the cyst and clinical examination can help to choose one of them.[9,28–30] No significant postsurgical complication was reported except in one case with residual left upper limb weakness.[21]

Management options of the cyst range from aspiration, fenestration, removal of the cyst wall, marsupialization (suturing its wall to the arachnoid of the cord to prevent reclosure), or placement of the cysto-subarachnoid shunt. Most authors used wide fenestration of the cyst with excision, as much as possible, of its wall. Excising the cyst wall is usually difficult due to its adherence to the cord parenchyma, so other options should be considered.

Four articles report recurrence after management. A case in which the initial surgical management involved fenestration alone without cyst wall resection, a recurrence of the IMAC occurred after 27 months and the surgery was repeated with proper resection of the cyst wall achieved, indicating the potential benefit of an adequate cyst wall resection in preventing the recurrence of such cysts.[21] In another case, aspiration trial was done twice; the patient improved for some time, but then reaccumulated within 4 months. The case ended with surgery with cysto-subarachnoid shunt.[23] Third case reported reaccumulation after 11 months after incomplete wall resection; no data are available about resurgery.[19] In our case during the surgery there was evidence of laminectomy but no drain or stiches for marsupialization, so we hypothesized that fenestration with or without cyst wall excision was done during the first surgery. We do not know exactly when reaccumulation happened but the authors did adequate excision of cyst wall (second surgery) after 13 years.

Definite mechanism of the pathological development of an IMAC remains idiopathic and Valsalva maneuver did not show any CSF leakage or expansion of the cyst.[18] Zekaj et al reported a case of development of an IMAC on follow-up after near-total removal of an intradural extramedullary cyst, with a small part of cyst wall left behind.[31] Rahimizadeh and Soufiani report a case association of cervical spondylosis and IMAC.[15]

Radiologically, the IMAC presents as well-demarcated intramedullary cystic lesion, hypointense on T1-weighted and hyperintense on T2-weighted images, with absence of pointed craniocaudal poles of the lesion, and shows neither restriction on diffusion-weighted images nor enhancement after intravenous gadolinium administration.[16,32]

Lessons

IMAC is showing bimodal incidence and trending to occur below 10 years and after 30 years. However, rarely, it should be considered in the differential diagnosis of intramedullary cystic lesions. Authors suggest doing laminoplasty or fusion in pediatric patients to prevent kyphoscoliosis deformity in the long run, or to do early surgery to gain the best outcome. Resection of the cyst wall should be done as much as possible; if it could not be achieved, then marsupialization or cysto-subarachnoid shunt should be considered. Aspiration alone or fenestration is not enough to eradicate the cyst. Long-term and prospective studies are recommended to achieve the best treatment options.

Ethical Approval

This case report and literature review study was waived by the local Ethics Committee of the hospital and all the procedures being performed were part of the routine care.

Consent to Participate

Written informed consent was obtained from all individual participants included in the study.

Consent to Publish

The authors affirm that human research participants provided informed consent for publication of the images and data.

Availability of Data
All data are available in the database of the hospital.

Funding
None.

Conflict of Interest
None declared.

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