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Review Article

Delayed peritoneal shunt catheter migration into the pulmonary artery with indolent thrombosis: A case report and narrative review

Mayur S. Patel[1], Justin K. Zhang[1], Ali Saif Raza Khan[2], Georgios Alexopoulos[1], Maheen Q. Khan[3], Philippe J. Mercier[1], Joanna M. Kemp[1]

[1]Department of Neurosurgery, Saint Louis University School of Medicine, Saint Louis, Missouri, [2]Department of Neurosurgery, Texas Tech University Health Sciences Center, Lubbock, Texas, [3]Department of Neurosurgery, Saint Louis University, Saint Louis, Missouri, United States.

E-mail: *Mayur S. Patel - mayur.patel@health.slu.edu; Justin K. Zhang - justin.zhang@health.slu.edu; Ali Saif Raza Khan - alisaifrkhan@hotmail.com; Georgios Alexopoulos - georgios.alexopoulos@health.slu.edu; Maheen Q. Khan - maheen.khan@health.slu.edu; Philippe J. Mercier - philippe.mercier@health.slu.edu; Joanna M. Kemp - joanna.kemp@health.slu.edu

***Corresponding author:**
Mayur S. Patel,
Department of Neurosurgery,
Saint Louis University School of Medicine, Saint Louis, Missouri, United States.

mayur.patel@health.slu.edu

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ABSTRACT

Background: Ventriculoperitoneal (VP) shunts are the preferred surgical treatment for hydrocephalus, and rarely, these operations may be complicated by catheter migration to ectopic sites. We present the case of an asymptomatic VP shunt patient with delayed peritoneal catheter migration into the pulmonary artery shunt catheter migration into the pulmonary artery (SCMPA) complicated by knotting and indolent thrombosis, necessitating open-heart surgery for system retrieval.

Methods: We conducted a literature review in PubMed, Scopus, and Web of Science of prior similar reported cases and present the results of 24 cases of SCMPA.

Results: An asymptomatic 12-year-old male presented with SCMPA noted on routine annual follow-up imaging. Preoperative CT angiogram indicated extensive catheter looping into the pulmonary artery without evidence of thrombosis. Less invasive attempts to retrieve the retained catheter were unsuccessful, and open-heart surgery was required. Intraoperatively, a nonocclusive pulmonary arterial thrombus surrounding the knotted catheter was discovered that was lysed successfully before system retrieval.

Conclusion: VP shunt catheter migration into the pulmonary artery (SCMPA) with concurrent large vessel thrombosis can develop in pediatric patients incidentally without any clinical symptoms. Our report suggests that preoperative CT angiogram may be insufficient to detect arterial thrombosis in the presence of extensive intravascular catheter knotting. An open-chest approach may be the only viable surgical option for catheter retrieval in the presence of complex catheter coiling. The use of anticoagulation following open-heart surgery for retrieval of a migrated VP shunt catheter remains unclear, we here propose that continuation of long-term therapeutic anticoagulation may successfully prevent thrombus relapse.

Keywords: Catheter migration, Hydrocephalus, Shunt, Shunt complication, Shunt migration

INTRODUCTION

Ventriculoperitoneal (VP) shunting is the most common treatment for the management of congenital hydrocephalus, with more than 30,000 systems placed annually in the United States.[36] Although a lifesaving intervention, pediatric shunt failure rates can be as high as 23% in the

1st year of placement.[7,18,19] In addition, approximately 24% of pediatric patients will develop complications following shunt insertion and up to 22% will require a revision surgery.[7]

VP shunt complications include catheter obstruction, infection, system disconnection, hardware failure, and pseudocyst formation.[11,31] Less commonly, VP shunt catheters may migrate into ectopic sites, including the bowels, bladder, abdominal wall, thoracic cavity, and cardiovascular system.[18] Distal VP shunt catheter migration into the pulmonary artery (SCMPA) with concurrent large vessel thrombosis is an extremely rare occurrence. To the best of our knowledge, only 24 cases with SCMPA have been previously reported [Table 1].[3,5,8,9,13,22-25,27,32,39] We present the first case of an asymptomatic pediatric patient with SCMPA, further complicated by extensive knotting and indolent arterial thrombosis. This complication necessitated removal of the retained catheter through open-heart surgery, given our inability to retract the system after repeated manipulation using less invasive techniques. We provide a brief review of prior similar cases identifying SCMPA as complications of VP shunting and compare our findings with the literature.

A narrative literature review was performed in PubMed, Scopus, and Web of Science using the terms "ventriculoperitoneal shunt" and "migration" or "extrusion" or "misplacement." After screening the databases and removing duplicate entries, a total of 33 cases of intracardiac VP shunt migration which include the 24 cases of SCMPA were identified [Table 1].

CASE DESCRIPTION

A 12-year-old male with a history of open lumbosacral myelomeningocele treated with postnatal repair and Chiari-II-related shunted hydrocephalus presented to the neurosurgery clinic for his annual follow-up visit. The patient did not have any significant cardiac or pulmonary history. His initial shunt was placed at the age of 4 weeks, where a right VP system was utilized for permanent cerebrospinal fluid (CSF) diversion. The patient required a single revision at 11 years of age after presenting with incidental ventriculomegaly on routine follow-up imaging. He was found to have a disconnected distal VP catheter at the level of the neck. Only the distal portion of the VP system was revised, wherein the new peritoneal catheter was proximally connected to the existing shunt valve. The placement of the new catheter was uneventful; there was no copious tunneling during the procedure or damage to any adjacent structures of the neck or chest [Figure 1].

At 12 years of age, approximately 1 year following his single VP shunt revision, our patient was incidentally found with SCMPA on routine imaging during his regular follow-up clinic visit. On examination, the patient was asymptomatic with stable vital signs and denied any complaints at that time.

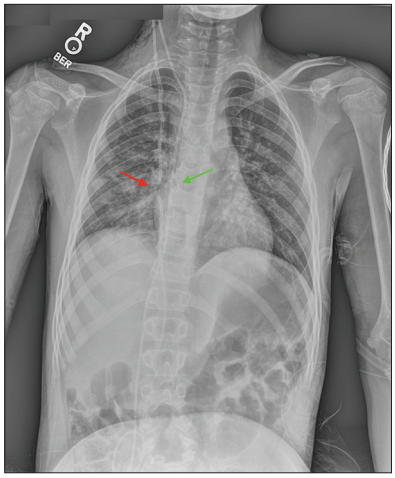

Figure 1: X-ray shunt series completed on postoperative day 1 after the initial revision at 11 years of age and 1 year before distal shunt catheter migration. Chest and abdominal AP X-ray shunt series demonstrated peritoneal VP shunt catheter disconnection. The abandoned system on the right (red arrow) represents the old, disconnected shunt catheter while the distal catheter on the left (green arrow) represents the newly inserted peritoneal shunt system.

Brain MRI revealed supratentorial ventriculomegaly, which was increased from baseline scans. Further workup with XR shunt series demonstrated distal shunt catheter migration into the thoracic cavity [Figure 2]. The patient was admitted for VP shunt exploration. Chest CT angiogram revealed that the peritoneal catheter had migrated through the right heart chambers into the pulmonary circulation, ultimately coiled multiple times within the pulmonary artery [Figure 2]. Transthoracic echocardiogram did not demonstrate any significant thrombosis, obstruction of flow, or right ventricular heart strain.

Extensive discussions with the patient's family regarding the extreme rarity of this condition were made, and we mutually agreed to proceed with surgical removal of the retained catheter to prevent any further cardiovascular complications. Initially, we attempted to remove the system through the existing retroauricular surgical incision, utilizing gentle traction. The migrated peritoneal catheter was identified at its proximal connection to the shunt valve, and after multiple traction attempts, we failed to obtain the system**.** We then proceeded with catheter manipulation under intraoperative fluoroscopy. Following multiple unsuccessful attempts, the patient developed premature ventricular contractions with any further catheter manipulation. Hence, a tortuous course into the pulmonary vasculature and/or extensive knotting was suspected. The interventional team was consulted, who stated that an endovascular approach for retrieval would not be feasible. It was soon made evident that the retained catheter could only be removed through an open-chest

Table 1: Literature review of intracardiac and pulmonary vasculature migration of malpositioned VP.

Authors	Patient age (years), gender (male/ female)	Time to detection from initial insertion	Symptoms on presentation	Pulmonary vascular migration?	Presence of knotting of catheter preoperatively?	Presence of intracardiac and/or pulmonary artery (s) thrombus(i) preoperatively?	Presence of intracardiac and/or pulmonary artery (s) thrombus(i) intraoperatively?	Presence of knotting of catheter in traoperatively?	Management
Morell *et al.*, 1994[26]	8, M	4 years	Headaches, vomiting, and hypertension	Y					Staged removal through an endovascular approach
Kang *et al.*, 1996[16]	12, M	2 months	Headaches, vomiting, fever, and chest pain	N	Y	NL	NL	Y	Thoracotomy, shunt replacement
Fraizer *et al.*, 2002[10]	14, M	1 month	Hypertension	N	Y	NL	NL	NL	Subxiphoid incision and shunt replacement
Imamura and Nomura, 2002[15]	76, M	18 days	Asymptomatic	Y	Y	NL	NL	Y	Cervical incision
Kubo *et al.*, 2002[21]	48, M	1 month	Neck pain	Y	N	NL	NL	NL	Cervical incision
Rodriguez-Sanchez *et al.*, 2003[33]	38, M	1 year	Fatigue, instability	Y	NL	NL	Y	Y	Initially: neck incision, catheter cut, and isolated anticoagulant therapy Few days later: interventional approach to remove knotted catheter and thrombus from pulmonary artery
Fewel and Garton, 2004[7]	16, M	33 days	Seizure	Y	Y	N	NL	NL	Retroauricular incision and shunt replacement
Kim *et al.*, 2005[20]	70, F	2 months	Gait disturbance, urinary incontinence	N	Y	NL	NL	NL	Mid-abdominal incision and shunt replacement
Chong *et al.*, 2008[4]	68, M	2 weeks	Abdominal pain	Y	Y	N	NL	NL	Cervical incision and interventional approach

(Contd...)

Table 1: *(Continued).*

Authors	Patient age (years), gender (male/ female)	Time to detection from initial insertion	Symptoms on presentation	Pulmonary vascular migration?	Presence of knotting of catheter preoperatively?	Presence of intracardiac and/or pulmonary artery (s) thrombus(i) preoperatively?	Presence of intracardiac and/or pulmonary artery (s) thrombus(i) intraoperatively?	Presence of knotting of catheter in traoperatively?	Management
Hermann *et al.*, 2009[14]	51, F	5 months	Fever, headache, pharyngitis	Y	Y	N	NL	NL	Failed removal through retroauricular incision, successful removal after using two Gooseneck snares, and removing the shunt in stages
Rizk *et al.*, 2009[32]	6, F	18 days	Respiratory distress	N	Y	NL	NL	NL	Retroauricular incision
Rizk *et al.*, 2009[32]	6, F	NL (Original shunt placed in infancy)	Chest pain	N	Y	NL	NL	NL	Retroauricular incision, lidocaine to prevent arrhythmia
Ryugo *et al.*, 2009[35]	50, M	2 years	Abdominal pain, hernia at incisional site	Y	NL	NL	N	NL	Catheter extraction under transluminal guidance, incision lateral to the sternocleidomastoid
Ruggiero *et al.*, 2010[34]	14, F	1 month	Abdominal pain, swelling around surgical site	Y	Y	N	NL	NL	Cervical Incision
Kano *et al.*, 2010[17]	6, M	6 days	Vomiting, somnolence	N	Y	NL	NL	NL	Subclavicular incision and shunt repositioned
Nguyen *et al.*, 2010[28]	38, M	8 months	Neck pain, swelling	Y	Y	NL	NL	NL	Neck incision and interventional approach
Nordbeck *et al.*, 2010[29]	6, M	NL	Fever, arrhythmia, abnormal cardiac sounds	Y	Y	NL	NL	NL	Open-heart surgery
Wei *et al.*, 2012[38]	72, M	4 years	Fever, cough, expectoration	Y	Y	NL	Y	NL	Cervical incision

(Contd...)

Table 1: *(Continued).*

Authors	Patient age (years), gender (male/ female)	Time to detection from initial insertion	Symptoms on presentation	Pulmonary vascular migration?	Presence of knotting of catheter preoperatively?	Presence of intracardiac and/or pulmonary artery (s) thrombus(i) preoperatively?	Presence of intracardiac and/or pulmonary artery (s) thrombus(i) intraoperatively?	Presence of knotting of catheter in traoperatively?	Management
Wei *et al.*, 2012[38]	37, M	5 days	Asymptomatic	Y	Y	NL	NL	NL	Retroauricular incision and shunt replacement
Zairi *et al.*, 2012[39]	63, M	7 days	Asymptomatic	Y	NL	NL	NL	NL	Retroauricular incision and shunt replacement
Manix *et al.*, 2014[25]	34, M	8 months	Headache	N					
Frahm-Jensen *et al.*, 2015[9]	77, F	2 months	Hypertension, gait unsteadiness	N	Y	NL	NL	Y	Venotomy, shunt replacement
Aboukais, 2015[2]	30, M	NL	Exacerbation of an asthma attack	Y	Y	NL	NL	Y	Cervicotomy, and shunt replacement
Lyon *et al*, 2016[24]	71, M	3 weeks	Gait disturbances, slow reactions, abdominal pain, worsening sleep apnea	Y	Y	NL	NL	NL	Retroauricular incision
Dossani *et al.*, 2017[5]	30, M	1 month	Headache, somnolence, lethargy	Y	NL	NL	NL	NL	Perivascular snare retrieval
Ralston *et al.*, 2017[30]	17, F	7 years	Headaches	Y	Y	NL	Y	Y	Open-heart surgery
Hajdarpasic *et al.*, 2019[13]	56, M	3 years	Fever, sepsis	Y	Y	NL	NL	NL	Supraclavicular and right cranial incision, a new shunt was not placed

(Contd...)

Table 1: *(Continued).*

Authors	Patient age (years), gender (male/ female)	Time to detection from initial insertion	Symptoms on presentation	Pulmonary vascular migration?	Presence of knotting of catheter preoperatively?	Presence of intracardiac and/or pulmonary artery (s) thrombus(i) preoperatively?	Presence of intracardiac and/or pulmonary artery (s) thrombus(i) intraoperatively?	Presence of knotting of catheter in traoperatively?	Management
Adib *et al.*, 2019[3]	38, M	7 months	Progressive thoracic pain and nocturnal cough	Y	Y	N	NL	NL	Neurosurgical removal of shunt, endovascular approach to remove knot, shunt replacement, prophylactic Enoxaparin
Moriarty *et al.*, 2019[27]	49, F	5 years	Chest pain, dyspnea on exertion	Y	Y	NL	Y	NL	Transfemoral endovascular removal and shunt replacement
Li *et al.*, 2019[23]	19, M	2 months	Dizziness and gait unsteadiness	Y	Y	NL	NL	NL	Retroauricular incision
Lancini and Shetty, 2019[22]	61, F	18 months	Acute chest pain and troponin rise	Y	N	NL	Y	N	Thrombus removal by atriotomy
Finneran *et al.*, 2020[8]	81, M	1.5 months	Bifrontal headache, nasal drainage, and congestion	Y	NL	NL	NL	Y	Retroauricular incision, transfemoral endovascular removal, and shunt replacement

All adult and pediatric patients are demonstrated in this table. We identified a total of 33 cases of cardiopulmonary VP shunt migrations, out of which 24 patients were presented with SCMPA. Only six pediatric SCMPA cases have been reported in the literature. Patients' demographics including age, gender, and symptoms are shown. The time from the last VP shunt insertion and presentation at shunt catheter migration is also depicted. Some authors emphasize the presence of cardiopulmonary knotting as well as pre-, intra-, and post-operative identification of the arterial thrombus. Finally, the revision technique including proximal catheter traction through an open approach, intraoperative fluoroscopy, endovascular retrieval, open-heart surgery, or a combination of techniques is demonstrated in addition to any administered prophylactic anticoagulation for each case. Abbreviations include M: Male, F: Female, Y: Yes, N: No, NL: Not listed, SCMPA: Shunt catheter migration into the pulmonary artery, VP: Ventriculoperitoneal

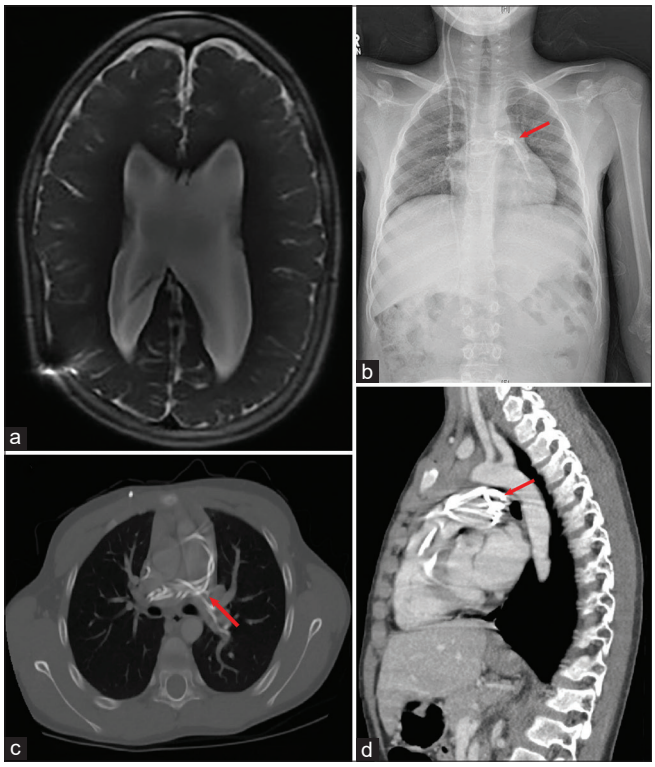

Figure 2: T2 MRI axial views with contrast revealed ventriculomegaly (a). X-ray shunt series demonstrated migration of the distal VP shunt catheter into the heart and pulmonary artery, with evidence of intracardiac knotting (red arrow) (b). Noncontrast chest CT in the axial and sagittal views showed extensive knotting of the distal shunt catheter in the pulmonary vasculature (arrow) without definite arterial thrombosis (c and d).

approach. After further discussions regarding the risks of abandoning the retained shunt catheter in the pulmonary vasculature, the patient was placed onto extracorporeal circulation and remained in the ICU for 48 h on therapeutic anticoagulation with IV heparin.

Our patient returned to the operating room on cardiopulmonary bypass approximately 48 h following the initial attempts for shunt retrieval. A midline sternal incision was made by our colleagues in cardiothoracic surgery. Temporary ligation of the pulmonary artery was performed using a purse string to obtain access to the migrated shunt catheter. A pulmonary arteriotomy was made, which revealed that the retained shunt catheter was coiled multiple times [Figure 3] in the main pulmonary trunk, resulting in nonocclusive arterial thrombosis. After the intra-arterial clot was lysed, the migrated catheter was retrieved successfully [Figure 3] and the pulmonary circulation was restored. Due to the necessity of long-term postoperative therapeutic anticoagulation secondary to intraoperative findings of pulmonary arterial thrombosis, the shunt system was externalized distally to the valve for temporary CSF

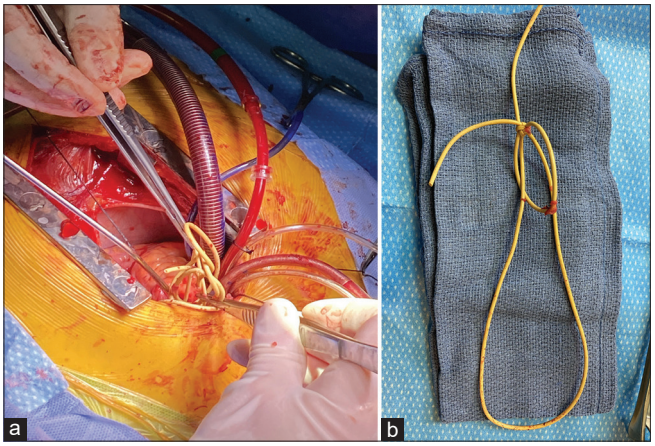

Figure 3: Intraoperative images demonstrating significant knotting on the migrated distal VP shunt catheter (a and b). There was extensive amount of intra-arterial pulmonary thrombosis that was lysed before successfully retrieving the catheter from the thoracic vasculature.

diversion until reinternalization, through a contralateral VP shunt system, was able to perform safely. Before discharge, the patient was transitioned to oral anticoagulation with Coumadin for a total course of 3 months. During his postoperative and annual follow-up clinic visits, the patient was completely asymptomatic with an intact VP shunt system and normal positioning of the distal catheter.

DISCUSSION

VP shunting is the first-line treatment of congenital hydrocephalus. Although an effective procedure, it is associated with failure rates up to 23% in the pediatric population.[7,18,19] Other therapies of hydrocephalus include ventriculoatrial and ventriculopleural systems. The long and tortuous route of these distal catheters makes the systems susceptible to malposition inside abnormal anatomic locations. Distal VP shunt catheter migration has been previously reported into the bowels, right heart chambers, pulmonary vasculature, urogenital tract, and other anatomic locations.[8] A BMI greater than 30 kg/m^2 in adults and the number of previous shunt revisions have been documented as independent risk factors for distal catheter migration.[1]

Historical background

VP shunt migration to ectopic sites is a relatively rare phenomenon in terms of shunt failure. The first cardiac migration of a distal shunt catheter was reported by Morell *et al*. in 1994 in a 12-year-old male. The patient presented with headaches, vomiting, and hypertension 4 years after the initial shunt placement. This was also the first reported case of shunt migration into the pulmonary vasculature.[26]

The first case of SCMPA in a pediatric patient who underwent an open-heart surgery was reported by Kang *et al*. in 1996. This patient was also symptomatic and presented 2 months after the initial shunt placement. Interestingly, the authors also observed intraoperative knotting of the distal catheter in the right ventricle. This patient had a pure cardiac migration without involvement of the pulmonary arteries.[16] In 2002, Imamura *et al*. described the first asymptomatic patient with a cardiac shunt migration in a 76-year-old, symptomatic, male patient who underwent a shunt replacement after a retrieval of the migrated catheter through a subxiphoid window.[15] Finally, Rodriguez-Sanchez *et al*. in 2003 were the first to report the presence of an intraoperative thrombus concurrently with a knotted catheter in the pulmonary vasculature.[33]

Clinical presentation

SCMPA is a rarer cause of cardiopulmonary shunt migration, where the catheter migrates to the pulmonary vasculature.[7,11,18,19,36] Patients with SCMPA usually develop signs and symptoms of increased ICP, including headaches, mental status changes, focal neurological deficits, and vomiting. In contrast, SCMPA patients with concurrent arterial thrombosis usually present with the right heart strain symptoms, such as, dyspnea, chest pain, exertion, and coughing in the emergency setting with signs of respiratory failure. SCMPA with arterial thrombosis can also lead to pulmonary embolism and respiratory arrest, hence, the need for urgent shunt revision.[7] In the two pediatric reports where extensive knotting was reported, all patients presented with clinical symptoms.[16,30]

Diagnosis, management options, and outcomes

There are no clear guidelines for the diagnosis of shunt migration and SCMPA.[40] Diagnosis can be made by an X-ray shunt series to assess the proximal and distal catheter failures or migration. Shunt failures and complications are diagnosed by clinical symptomology and CT scans of the head.[6,40]

Various surgical interventions exist to manage VP shunt migration. Minor surgery with posterior auricular or cervical incisions in adjunction with cardiovascular surgery has been reported.[3,7,15,21,32] Fluoroscopy can be used to detect knotting in the pulmonary vasculature. A combined surgical and interventional radiological approach has also been described.[26] Furthermore, open approaches include thoracotomy or using a subxiphoid window.[3,10,16] Interventional therapy may include procedures that involve loop snare devices, grasping forceps, helical baskets, Fogarty balloon catheters, and hook catheter/guidewire combinations.[3]

Data on long-term outcomes for VP shunt revisions and replacements are limited.[12] Patients who have had a shunt revision are more likely to experience a failure again.[12] Independent risk factors for another revision include cardiac diseases, chronic history of seizures, or a history of neuromuscular disease.[12]

Due to the high rate of VP shunt failures, especially in pediatric patients and regardless of revisions, it is recommended that patients are followed at least through the transition to adulthood as revisions and replacements are reported in the literature after 17 years of initial shunt placement.[37]

Literature review

Delayed SCMPA resulting in arterial thrombosis following VP shunt insertion is an extremely rare complication, with 24 published reports among the adult and pediatric population [Table 1]. Among the five SCMPA cases with extensive catheter knotting, all of them were found with pulmonary arterial thrombosis intraoperatively. Only six pediatric SCMPA cases were identified in the literature and three of them had an asymptomatic presentation. The timing of cardiopulmonary shunt migration at presentation varies widely from 2 days to 5 years [Table 1]. In 1994, Morell *et al*. were the first to report SCMPA as a VP shunt complication, while the patient was treated with staged removal through an endovascular approach.[26] Kang *et al*. described the case of an intracardiac shunt catheter migration and the shunt was retrieved by open thoracotomy, as knotting of the distal system posed significant difficulties for foreign body removal.[16]

Mechanism of shunt migration

Cardiopulmonary VP shunt catheter migration has been generally associated with two different proposed mechanisms in the literature.[3,5,10,20] The most common cause is iatrogenic perforation of the external jugular vein during tunneling of the distal catheter or subcostal placement of shunt tunneling instruments.[3,5] Adib *et al*. described how the suction effect of the venous system enhanced by the negative intrathoracic pressure with respirations could result in gradually pulling the catheter into the venous system after an iatrogenic perforation.[3] Subsequently, if the catheter migrates far enough, it can be lodged and subsequently knotted into the pulmonary arteries, leading to SCMPA, where patients classically present with the right heart strain symptoms.[3,20] To the best of our knowledge, no reports have previously documented an asymptomatic SCMPA patient with indolent arterial thrombosis [Table 1]. Another proposed mechanism suggests that the distal shunt catheter may progressively erode into the wall of the external jugular vein by forming a hairpin bend due to continuous traction from neck movements.[3,10] Other cardiopulmonary shunt migration theories proposed

by Wei *et al.* and Fewel *et al.* include size discrepancy between adjacent veins and hardware placement.[7,38] In our patient, we hypothesize that the migration could have been resulted by iatrogenic perforation of the external jugular vein given his recent VP shunt revision 1 year before presentation with SCMPA [Figure 4]. However, our patient did not report any subcutaneous feeling of distal catheter migration, neither any popping sounds that could have revealed an indolent fracture before his scheduled annual follow-up. Whether or not, patients with a fractured distal VP catheter migrating into the intrathoracic cavity can have a subjective feeling of migration remains to be determined. Subjective sensation of subcutaneous distal VP shunt catheter migration by the patients has not been previously reported in the literature in similar cases of SCMPA [Table 1].

Surgical management of SCMPA

Surgical strategies to correct SCMPA range from open revision surgery with catheter manipulation using gentle traction at the most proximal site to endovascular retrieval using loop snare device.[7,15,17,20,35] Alternatively, catheter manipulation under intraoperative fluoroscopy, open thoracotomy, or a combination of these techniques may be used.[10,16,26] In the presence of significant intravascular looping or catheter knotting, open revision with proximal retrieval may be inadequate.[30]

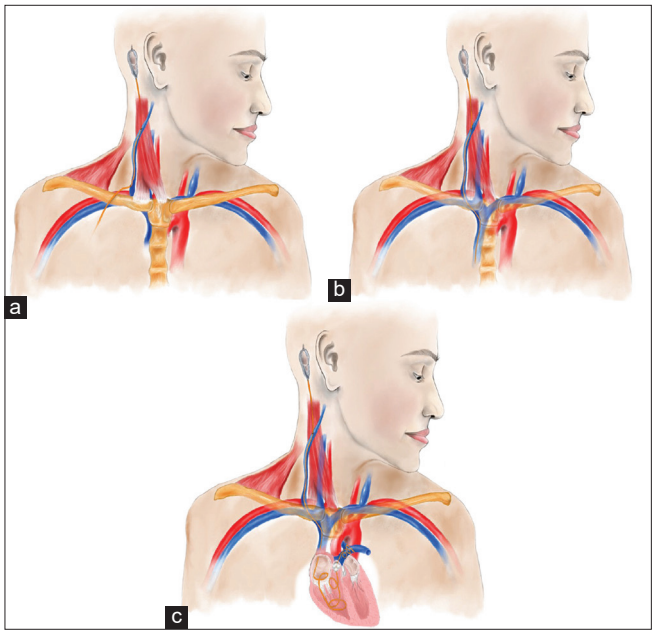

Figure 4: Medical illustrations describing proposed mechanisms of VP shunt migration. Erosion of the external jugular vein due to excessive traction form neck movements at varying levels of the vein (a). Iatrogenic perforation of the external jugular vein during tunneling of the distal catheter (b). In both (a and b), negative intrathoracic pressure along with venous flow further displaces the catheter into the vein, pulling it caudally. If the catheter migrates far enough, it can be lodged and knotted in the pulmonary arteries, as indicated in caption (c).

Among the six SCMPA pediatric reports, the catheter was retrieved through an open cardiac approach in 3 cases (50%), while two patients, as reported by Kang *et al.* and Ralston *et al.*,[16,30] demonstrated significant intravascular catheter knotting. Kang *et al.*[16] retrieved the retained catheter by a superior vena cava venotomy while the other two authors,[16,30] similarly to our case, retrieved the foreign body through a pulmonary arteriotomy. Less invasive techniques failed to retrieve the shunt catheter in our patient, and open-heart surgery was eventually performed as the last resort. In addition to obscuring thromboses, complex looping of catheters may result in developing knots during the removal process, in which case, interventional or less invasive attempts may be inadequate.[2,4,10,16,28,29] In our case, we attribute the failure of less invasive methods to the indolent pulmonary arterial thrombosis and extensive intravascular coiling that led to persistent cardiac arrhythmias on further catheter manipulation attempts. The least invasive technique should always be utilized to retrieve a retained shunt catheter before considering an open-chest approach, but in the presence of significant intravascular knotting with concurrent arterial thrombosis in a pediatric patient, catheter manipulation may lead to lethal cardiac arrhythmias.

Concurrent pulmonary arterial thrombosis

To the authors' knowledge, no study has noted the presence of pulmonary thromboses on preoperative imaging in the setting of SCMPA.[3,4,7,14,34] Interestingly, both preoperative echocardiogram and CT angiogram failed to demonstrate a thrombus surrounding the knotted catheter in our case. Our report is the first to describe how preoperative CT angiogram and echocardiography can be insufficient to detect arterial thrombosis in the presence of a coiled catheter. This is important to distinguish, as the pulmonary arterial thrombosis could have led to pulmonary embolization, shock, or cardiovascular complications, should open-heart surgery have been postponed for a later time. Although preoperative imaging missed vital information, it remains essential to assess the extent and course of catheter migration into the pulmonary vasculature. Therefore, despite negative findings on imaging, a thrombus could not be always ruled out in SCMPA patients [Figure 3].

The need for continuation of therapeutic anticoagulation after clot removal remains unclear. If detected intraoperatively, it can be managed medically through the same guidelines as central venous catheter-related thrombosis.[2,4,28] This includes a minimum of 3 months of anticoagulation or continued anticoagulation therapy based on patient risk factors.[28] At follow-up visits, our patient was clinically stable

without recurrent thrombosis on imaging, which suggests that the anticoagulation strategy was successful in our case. However, the utility of anticoagulation therapy in this patent population should be further explored in future studies.

CONCLUSION

SCMPA with concurrent large vessel thrombosis can develop incidentally in pediatric patients without any clinical symptoms. Preoperative CT angiogram may be insufficient to detect arterial thrombosis in the presence of extensive catheter knotting. Clinicians should be aware of the limitations of preoperative imaging and understand that the lack of symptoms may not necessarily be an optimal indicator of the extent of concurrent thrombosis. An open-chest approach may be the only viable surgical option for catheter retrieval in the presence of complex intravascular catheter coiling. Continuation of long-term therapeutic anticoagulation in these patients may successfully prevent arterial thrombus relapse.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

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