Direct stick embolization of a rectal venous malformation via transanal minimally invasive surgery

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ABSTRACT

Rectal venous malformations (VMs) are rare clinical entities with variable patterns of presentation. Treatment requires unique, targeted strategies based on the symptoms, associated complications, and location, depth, and extent of the lesion. We present a rare case of a large, isolated rectal VM treated by direct stick embolization (DSE) using transanal minimally invasive surgery (TAMIS). A 49-year-old man had presented with a rectal mass incidentally detected on computed tomography urography. Magnetic resonance imaging and endoscopy revealed an isolated rectal VM. Elevated D-dimer levels concerning for localized intravascular coagulopathy warranted the use of prophylactic rivaroxaban. To avoid invasive surgery, DSE using TAMIS was performed successfully without complications. His postoperative recovery was uneventful, aside from a self-limiting and expected course of postembolization syndrome. To the best of our knowledge, this is the first reported case of TAMIS-assisted DSE of a colorectal VM. TAMIS shows promise for more widespread use in the minimally invasive, interventional management of colorectal vascular anomalies. (J Vasc Surg Cases Innov Tech 2023;9:101124.)

Keywords: Direct stick embolization; Rectal vascular malformations; Transanal minimally invasive surgery; Vascular anomalies

Although venous malformations (VMs) are the most common type of vascular malformation, colorectal VMs are exceedingly rare. They can occur as isolated, sporadic lesions or as part of a broader syndrome.[1] Isolated VMs usually have activating mutations in *TIE2,* a receptor tyrosine kinase involved in recruitment of mesenchymal cells during angiogenesis. Syndromic associations tend to include PIK3CA-related overgrowth syndromes—such as Klippel-Trenaunay syndrome and CLOVES (congenital lipomatous overgrowth, vascular malformations, epidermal nevis, spinal/skeletal anomalies/scoliosis) syndrome—and *TIE2*-associated blue rubber bleb nevus syndrome.[2,3] Regardless, colorectal VMs require tailored treatment based on the symptoms, associated complications, and location and extent of the lesion. Options vary widely and include endovenous or endoscopic embolotherapeutic techniques, endoscopic band ligation, suture ligation of feeder vessels with focal resection, segmental bowel resection, and combinations thereof.[4]

Transanal minimally invasive surgery (TAMIS) has emerged as a minimally invasive modality for the management of rectal pathology, primarily rectal cancer. This technique conventionally involves the usage of a single-incision laparoscopic surgery port introduced into the anal canal, followed by establishment of a pneumorectum. This technique facilitates a platform for transanal surgery with conventional laparoscopic instruments to achieve high-quality local excision of tumors.

To the best of our knowledge, we are the first to have used TAMIS to perform fluoroscopy-guided direct stick embolization (DSE) of an isolated, sporadic, exophytic, proximal rectal VM. The patient provided written informed consent for the report of his case details and imaging studies in accordance with institutional guidelines.

CASE REPORT

A 49-year-old man had with an incidentally discovered 6.5-cm, rectal mass found on noncontrast-enhanced computed tomography urogram performed to evaluate microscopic hematuria. He reported no abdominal pain, proctalgia, changes in bowel habits, or hematochezia. His microscopic hematuria was later deemed unrelated to the rectal mass. The initial laboratory studies were remarkable for elevated D-dimer levels (2.95 mg/L). Prophylactic rivaroxaban was started for a suspected mild case of localized intravascular coagulopathy.

Contrast-enhanced magnetic resonance imaging (MRI) was obtained to better characterize and determine the anatomic extent of the lesion.[5] MRI revealed a large, isolated 6-cm × 4.5-cm, multilobulated, T2-weighted hyperintense lesion with

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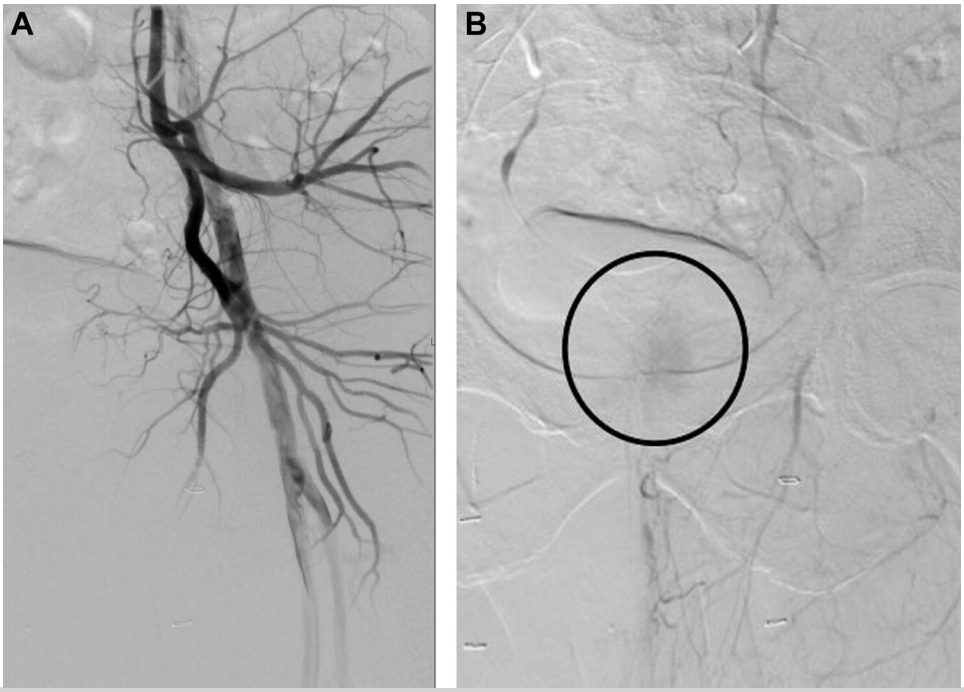

Fig 1. A, Selective angiogram of the internal iliac artery demonstrating no arterial component of a vascular malformation. **B,** However, delayed frames from the same angiogram demonstrated a region of contrast blushing projecting over the pelvis, congruent with a venous malformation (VM; *circled*).

numerous phleboliths along the posterior rectal wall at the rectosigmoid region with marked protuberance into the rectal vault. Colonoscopy revealed a cyanotic, nonpulsatile, multilobular mass causing significant rectal impingement and luminal narrowing. The findings from both MRI and colonoscopy were supportive of a diagnosis of a vascular malformation.

Selective visceral and internal iliac angiography revealed no evidence of arterial insufficiency or arteriovenous malformations. Delayed venous phase runs confirmed the presence of faint venous stagnation in the rectosigmoid junction consistent with an isolated VM (Fig 1).

A multidisciplinary discussion was held with patient and colorectal surgery colleagues. Treatment was suggested, given the extent of luminal impingement and ample phleboliths, and, most importantly, to prevent localized intravascular coagulopathy. Localized intravascular coagulopathy poses a significant risk for sequelae such as pain, hemorrhage, thromboembolism, and, most importantly, full progression to disseminated intravascular coagulopathy, which can result from surgical management or embolotherapy.[6] The patient and surgical team preferred the least invasive method of surgery and the avoidance of proctocolectomy, given the invasive nature of the latter, its potential complications, and the prolonged recovery. As such, a unique, tailored approach to DSE under direct endoscopic visualization and fluoroscopic guidance was devised using TAMIS.

Surgical technique. TAMIS facilitates minimally invasive intervention to rectal VMs by creating a gas-tight seal at the anus through which the rectum can be visualized while providing an aperture wide enough for laparoscopic instruments to be inserted for intervention. The GelPOINT mini advanced access platform (Applied Medical, Rancho Santa Margarita, CA) was used to conduct TAMIS. The platform consists of an Alexis O-wound protector/retractor, a GelSeal cap, and 5-mm AirSeal port (Fig 2). In the hybrid operating suite, the patient was placed in the modified lithotomy position. The Alexis O-wound protector/retractor was inserted into the anus, and the proximal ring was secured above the anorectal ring and tightened to achieve adequate securement across the anus into the rectum, creating an aperture for advancement of the interventional equipment. The GelSeal cap was then secured to the distal ring and pneumorectum established with the aid of a 5-mm AirSeal port inserted through the GelSeal cap. Additional 5-mm ports were placed through the GelSeal cap, allowing for insertion of a 5-mm, 30° laparoscope and laparoscopic instruments without disruption of the seal. On inspection of the rectal lumen, several large protuberant cavernous clusters consistent with VMs were identified just proximal to the second rectal valve (Fig 3).

The patient was placed in the lithotomy position and general anesthesia was induced, without administration of prophylactic antibiotics. A 21-guage spinal needle was inserted directly through the Gel Seal cap. In tandem with the laparoscope and laparoscopic graspers, the spinal needle was visualized and guided through the malformation. Blood return was appreciated on removal of the inner stylet, indicative of intraluminal

Fig 2. Image of the GelPOINT mini advanced access platform (Applied Medical, Rancho Santa Margarita, CA) used during transanal minimally invasive surgery (TAMIS), consisting of the GelSeal cap (*arrow*), attached to the Alexis O-wound protector-retractor (*bracket*). The GelSeal cap is permeable to ports (*arrowheads*) for implementation of laparoscopic instruments. In this case, the spinal needle used for direct stick embolization (DSE) directly punctured through the GelSeal cap.

access. Direct stick venography through the needle confirmed proper placement and delineated the contour of the VM. DSE was performed by injecting a foamed emulsion of 10 mL of 3% sodium tetradecyl sulfate, 2.5 mL of lipiodol, and 1 part air under direct fluoroscopic visualization until adequate deposition in the region of the VM was appreciated without evidence of nontarget embolization (Fig 4). The needle track was packed with Surgiflo gelatin matrix (Ethicon, Raritan, NJ), and gentle surgical packing was performed with laparoscopic forceps. No immediate postoperative complications occurred. His postoperative D-dimer level was also elevated; therefore, hematology was consulted, who recommended another 2-week course of rivaroxaban.[7] At 3 weeks, the patient endorsed a reassuring course of recovery, only complaining of one bloody bowel movement and a self-limiting 2-week course of intermittent fevers most consistent with postembolic syndrome.[8] The patient additionally underwent further hematologic follow-up, which demonstrated improvement in his D-dimer levels. Thus, no further anticoagulation therapy was provided to the patient. Follow-up MRI demonstrated a decrease in the size and intensity of the VM, congruent with a therapeutic response (Fig 5). The patient continued to follow-up in the clinic regularly.

DISCUSSION

TAMIS was initially described in 2010 for use in facilitating access to the proximal and mid-rectum for resection of benign and early-stage malignant rectal lesions. Its use has now expanded to a variety of indications, including treatment of fistulas, rectal hemorrhage, foreign body removal, and so forth.[9] TAMIS allows for insufflation of carbon dioxide into the lumen of the bowel through a laparoscopic insufflator and, hence, creates a stable endoluminal operating platform. Laparoscopic instruments can be deployed transanally to perform excisions and other operative maneuvers. Furthermore, the surgical field can be viewed in 360° with high-definition optics, rendering this an ideal platform to conduct DSE, a maneuver that has evolved as a widely accepted mainstay of therapy for VMs.[10] Although colonoscopic-guided treatment of VMs has been reported,[11] our colorectal colleagues have elected to use TAMIS, because colonoscopy is a less stable platform and the instrumentation can obstruct fluoroscopic visualization of the lesion during the procedure.

Literature on the treatment of distal alimentary tract VMs is sparse given the rarity of the lesions, lack of awareness, and persistent use of erroneous nomenclature. For the minimally invasive treatment of VMs, DSE is the preferred approach for achieving embolization.[6] Embolization of lesions has been performed using a variety of agents, including ethanol. Agostinho et al[12] reported a case of a patient with Klippel-Trenaunay syndrome who had presented with hematochezia resulting in anemia secondary to multiple giant rectal VMs. They reported resolution of hematochezia and a decrease in the size of the large rectal VMs after injection of 75% ethanol.[12] However, they acknowledged a set of major adverse events related to distal embolization, including pulmonary embolism and cardiovascular collapse.[12] We continue to advise against the use of ethanol for the treatment of more voluminous VMs owing to the high rates of serious complications, despite its highly effective therapeutic profile.[13]

Given the lack of standardization for the DSE technique, methods, approach, and embolotherapeutic agents, the definitive treatment of colorectal VMs has remained surgical resection. Bhattacharjee et al[14] reported a case of a rectal VM that had presented with anemia secondary to recurrent rectal bleeding that was treated via mesorectal resection with coloanal anastomosis. The investigators reported that surgical excision is the ideal modality for the treatment of rectal VMs given their propensity for recurrence if treated with conservative measures, in particular, lesions with transmural involvement.[14] Deciding between surgery and sclerotherapy can be challenging, with treatment decision often

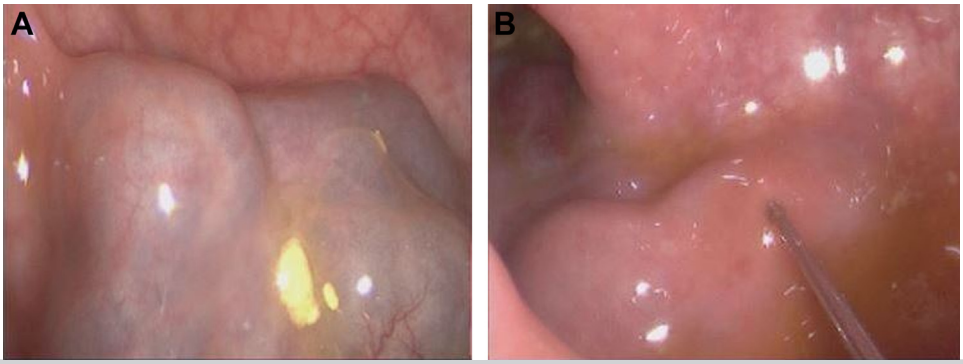

Fig 3. A, Laparoscopic visualization of the rectal venous malformation (VM) via transanal minimally invasive surgery (TAMIS). **B,** TAMIS allows for direct visualization of the trajectory and tip of the needle to ensure appropriate access without damage to surrounding structures.

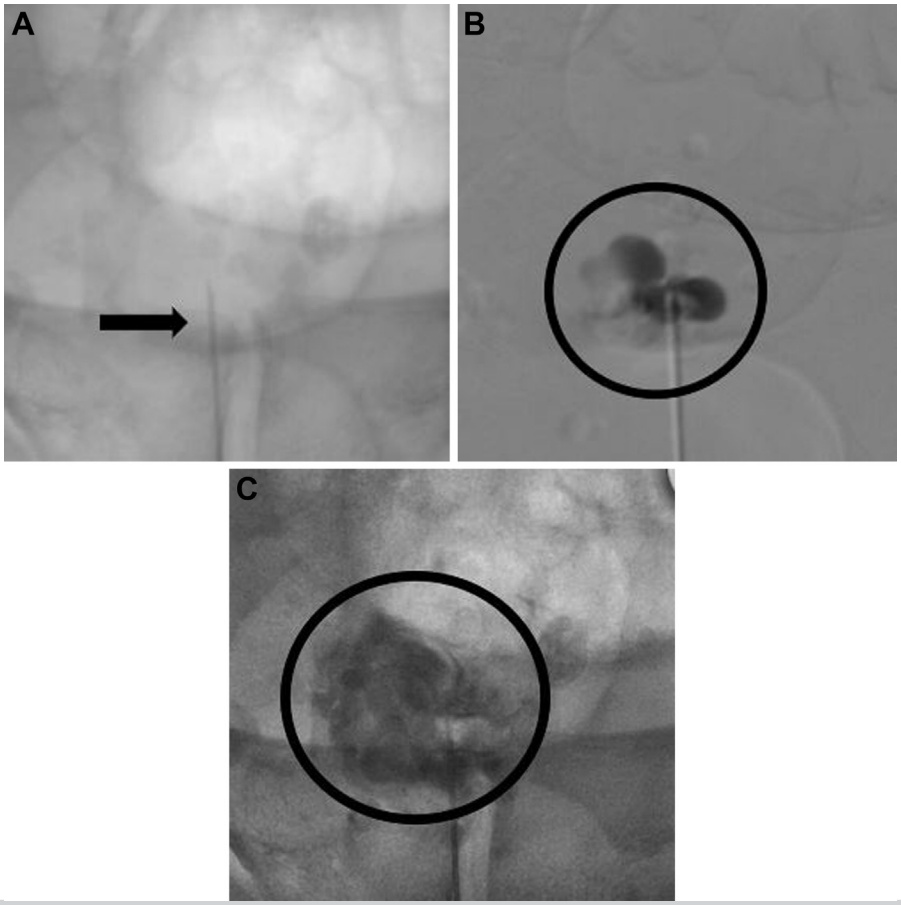

Fig 4. A, Fluoroscopy demonstrating the spinal needle (*arrow*) after puncture into the venous malformation (VM) before injection of sclerosant. **B,** Digital subtraction imaging through the spinal needle demonstrating contrast filling of the VM (*circled*), indicating adequate access. **C,** Fluoroscopy of VM after injection of sodium tetradecyl sulfate, lipiodol, air emulsion demonstrating radio-opacification of the VM (*circled*).

dependent on patient preference, disease extent, and symptoms.

Dasgupta and Fishman[15] stated that the management of visceral malformations (especially VMs of the gastrointestinal tract) depends on the extent of the anatomic distribution and symptoms. In the setting of congenital conditions such as blue rubber bleb nevus syndrome, which has the potential to present with

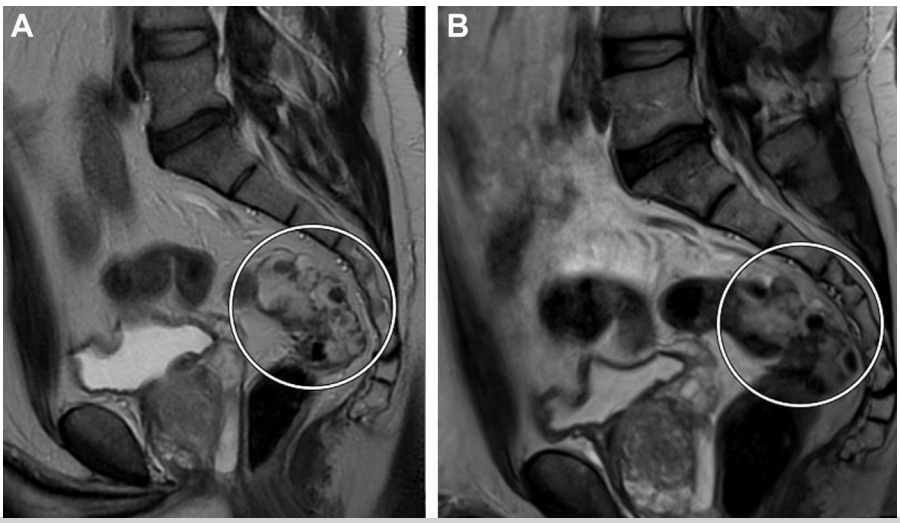

Fig 5. A, Sagittal section from T2-weighted magnetic resonance imaging (MRI) scan demonstrating a slight hyperintensity impinging on the rectum, consistent with a venous malformation (VM; *circled*). **B,** Follow-up MRI after embolization demonstrating a decrease in both the size and the intensity of the VM, congruent with a response to treatment.

transfusion-dependent anemia secondary to multiple foci of hemorrhaging VMs, multifocal surgical resection of all lesions with luminal contact might be the most judicious treatment modality, because it can be difficult to identify a single attributable lesion. However, for lesions limited to the distal anorectal VMs, Dasgupta and Fishman[15] suggested the efficacy of sclerotherapy via anoscopy. However, this might not provide permanent resolution, and surgical intervention could eventually be warranted.

Surgical resection and sclerotherapy need not be mutually exclusive interventions. Nassiri et al[1] reported a case of a patient with Klippel-Trenaunay syndrome who was experiencing hemorrhagic colorectal VM, resulting in transfusion-dependent anemia. The patient underwent multiple sessions of transcatheter embolization and DSE with sodium tetradecyl sulfate foam, which not only ameliorated the patient's symptoms and anemia, but also demonstrated reductive effects in the VM size, extent, and hemorrhage, which optimized the patient's candidacy for more definitive surgical resection by minimizing the risk of severe intraoperative hemorrhage.[1] The patient subsequently underwent resection of the involved colon, followed by coloanal anastomosis. This approach led to complete resolution of the patient's hematochezia.[1]

Additionally, for cases in which patients are not amenable to any intervention, pharmacologic therapies can be pursued to ameliorate the symptoms of colorectal VMs. Abematsu et al[16] reported a case of painless hematochezia resulting in anemia that was treated pharmacologically rather than with surgical intervention. The investigators reported stabilization of the serum hemoglobin levels with administration of propranolol and celecoxib via inhibition of aberrant vascular endothelial growth factor-directed angiogenesis. They also generally reported successful usage of octreotide as a first-line pharmacologic agent.[16] Tremendous value exists in using sirolimus as a systemically delivered pharmacologic method of addressing VMs by targeting the mTOR (mammalian target of rapamycin) pathway, although Nassiri et al[1] recognized that sirolimus is accompanied by side effects that could dissuade patients from its use.

Combination therapy is likely to be the evolving strategy of choice for patients with advanced, extensive, colorectal VMs with life-threatening complications. More focal lesions with a less severely threatening profile can be treated repeatedly when less invasive, highly targeted techniques such as TAMIS are used. TAMIS can also help expedite access for interventional treatment of these difficult-to-reach lesions. Although ideally used in a fixed-imaging operating or interventional arena, TAMIS can also be used with mobile imaging equipment, expanding access to more centers with the expertise to treat these complex lesions.

CONCLUSIONS

The management of rectal VMs can be successfully and safely managed in a coordinated, staged, and multidisciplinary approach. Although other treatment options have been used in the past for VMs, in the present case, TAMIS allowed for visualization, access, and maneuverability to manage rectal VMs via DSE. We encourage further use of TAMIS technology and continued multidisciplinary collaboration for the treatment of vascular malformations of the alimentary tract.

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