

# Embolization of Spinal Dural Arteriovenous Fistulae: Clinical Outcomes and Long-Term Follow-Up: A Multicenter Study

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## ABSTRACT

**Background Data:** The most frequent treatable vascular abnormality of the spinal canal is spinal dural arteriovenous fistulae (SDAVFs), which cause progressive para- or quadriplegia mostly affecting elderly males. SDAVFs are present in the thoracolumbar region. The main goal of treatment must be to obliterate the shunting zone via superselective embolization with the usage of a liquid embolic agent.

**Purpose:** This study aims to evaluate endovascular technique as a safe and efficient approach for the treatment SDAVFs, especially with long-term follow-up clinical outcomes.

**Study Design:** A retrospective clinical case study.

**Patients and Methods:** From May 2010 to May 2017, 15 patients who had symptoms attributed to SDAVFs underwent the operation in the Departments of Neurosurgery in Suhag, Tanta, and Al-Azhar Universities and Interventional Radiology, Ain Shams University. All the patients had varying degrees of progressive spastic paraparesis with and without sphincteric disturbances.

**Results:** Endovascular embolization was used in all cases. Fourteen were males, with ages ranging from 45 to 74 years old. After the treatment, good outcome was found in five patients (33.3%), a moderate outcome was delineated in six patients (40 %), and four patients revealed a poor outcome (26.7%).

**Conclusion:** Spinal AVF could be treated safely and effectively by the endovascular approach. Generally, there is no correlation between the disappearance of MRI abnormalities and significant clinical improvement. The preclinical state of the patient is directly proportional to the clinical outcome. Due to unexpected responses, embolization should be attempted even the patient is in a bad clinical condition. (2021ESJ239)

**Keywords:** spine, arteriovenous, fistula, endovascular, embolization

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## INTRODUCTION

Spinal vascular pathologies are considered rare. They represent less than 2% of all vascular neurological pathologies.<sup>1,11</sup> Among these lesions, spinal dural arteriovenous fistulae (SDAVFs) are by far the most common, accounting for around 80% of all spinal cases.<sup>5,15</sup>

SDAVFs mostly presented with back pain, progressive weakness, variable degrees of sensory deficits, and sphincteric problems.<sup>1,18,23</sup> The pathophysiology of SDAVFs is mainly due to elevated venous pressure resulting from shunting of the blood from the arteries to the veins, which is valveless, thus leading to reduction of the arterial supply with progressive necrotizing myelopathy, which may be irreversible if not treated.<sup>9,10</sup> The standard treatment for SDAVFs is usually surgically because it is a technically feasible and safe procedure with low rates of morbidity or recurrence. Other treatment options include endovascular embolization, which has achieved rising acceptance, either alone or combined with surgery.<sup>18,25</sup>

The aim of this study is to assess the efficacy and safety of the embolization technique in treating SDAVFs.

## PATIENTS AND METHODS

We retrospectively reviewed patients with symptoms due to SDAVFs in the period from May 2010 to May 2017 in Suhag, Tanta, Ain shams, and Al-Azhar University Hospitals. Fifteen patients were enrolled in this study with the mean age of  $54.2 \pm 10.3$  (range, 45–74) years.

Inclusion criteria were all patients with symptomatic SDAVFs confirmed by MRI and diagnostic spinal angiogram (DSA). Exclusion criteria were renal impairments, allergies to IV contrast agents, and blood coagulopathies. Patients that showed a shared artery supplying both the anterior spinal artery and the fistula were

excluded from the study; otherwise, embolization was primarily attempted for all spinal DAVFs.

All patients were subjected to clinical evaluation using the Aminoff–Logue motor disability scale (Table 1).<sup>2</sup>

**Table 1.** Aminoff–Logue motor disability scale.<sup>2</sup>

Disability	Degree
Hypesthesia without gait restriction	1
Reduced tolerance to exercise	2
Need a cane for walking	3
Need for two canes or crutches to walk	4
Inability to stand: patient in a wheelchair or in bed	5

According to the Aminoff–Logue scale, a good result was defined as motor power of Grade 4 or 5, indicating that the patient can ambulate and carry on with his daily activities (ALS1-ALS2); a moderate result was defined as motor power of Grade 3, indicating the patient needs support to walk or perform activities (ALS3-ALS4); a poor result was defined as motor power less than Grade 3 (ALS5) indicating the patient cannot perform any activities.

The epidemiological and perioperative data of our patients are summarized in Table 2. The Aminoff scores were checked by a neurosurgeon team that was not enrolled in the treatment plan.

Radiological assessments included noncontrast MRI and postcontrast-enhanced MRI that revealed flow void signals and cord increased intensity on T2W imaging and postgadolinium enhancement in all cases. Patients with findings suspecting typical characteristics of SDAVFs were scheduled for a conventional spinal angiogram. In only 1 case in our series that was not eligible for MRI due to placement of a cardiac pacemaker, a thoracolumbar CT angiogram was used that showed the appearance of the tortuous linear vascular channel in the spinal canal. After the MRI (or CTA) exam, all patients underwent a DSA; this

was done directly before the endovascular therapy in the same session.

The study was approved by the research ethics committee of our institutions and informed consent was obtained from each patient or their guardians. The study was conducted according to the WMA Declaration of Helsinki–Ethical Principles for Medical Research Involving Human Subjects.

#### **Endovascular Procedure:**

All patients underwent endovascular embolization under general anesthesia. A 5 or 6 French (Fr) femoral sheath is inserted, and a diagnostic catheter of 5 Fr is employed. Various catheter forms are frequently used during treatments; the Cobra shape and SIM II are the most used shapes because they may readily be placed into the arterial branches of the spinal cord. Starting with the internal iliac arteries and working up to the right and left vertebral arteries, the entire spinal vasculature is examined. Even though the fistula was found at lower levels, this procedure was followed to check any accompanying vascular pathologies that might not be seen. Moreover, we carefully evaluated the levels below and above the fistula bilaterally to diagnose and treat any additional feeders and check for the site of the anterior spinal artery (artery of Adamkiewicz).

If the proximal vein draining the fistula (foot of the vein) could be occluded with liquid embolic, embolization was regarded as satisfactory. In all cases, either Onyx or NBCA mixed with lipiodol was used as a liquid embolic administered through 1.2 Fr or 1.5 Fr microcatheters after superselective access of the involved radicular feeder arteries. A control angiogram finally confirms the disappearance of the SDAVFs.

Postprocedural follow-up included the following: clinical evaluation after 2 weeks, 1 month, 3 months, 6 months, and 1 year using the Aminouff–Louge scale comparing it with the preprocedure score, radiological investigations by MRI with contrast, and DSA after 1 month.

#### **Statistical Analysis:**

For statistical data analysis, IBM-SPSS, version 25 (IBM-Corporation, Chicago, USA; August 2017) was utilized. Mean, standard deviation (SD), number, and percentage were used to represent the data. To compare the means of the Aminouff–Louge motor disability scale, the paired *t*-test was utilized.

## RESULTS

A total of 15 patients with a mean age of  $54.2 \pm 10.3$  (range, 45–74) years, including 14 male patients (93.3%) and one female, were recruited for this study. All patients complained of progressive spastic paraparesis of different grades (100%) and eight patients (53.3%) had sphincter disturbances (Table 2).

Embolization with NBCA was done in four patients and Onyx in eleven patients. An arterial spasm occurred during embolization in patient 1, preventing the Onyx from getting to the vein. His follow-up angiogram revealed a reestablishment of the fistula (Table 3). Within two months, he was scheduled for another endovascular trial with Onyx re-embolization as he was surgically unfit, and we managed to treat the fistula in the second session. The fistulae in all patients were found between D4 and L1, and the feeder, in these cases, was a radicular artery. Fistulae arising from the lateral sacral artery, which is a branch of the internal iliac artery (IIA), were found in two cases: at the L5 root in one and at the S1 root in another one (Table 2).

The postembolization clinical result assessed according to the Aminouff–Louge scale is seen in Table 2. Five cases had a good result (33.3%), six cases had moderate results (40%), and four cases had poor results (26.7%). The comparison of the patient's pre-embolization and postembolization Aminouff–Louge scale scores showed a significant impact on their outcome. Patients who had a favorable outcome all had ALS level 3 before treatment, which meant they could walk with

**Table 2.** Summary of data of our study's patients (n=15).

No	Age	Sex	Presentation	Diagnosis	Material	Aminoff-Logue scale		Sphincter control
						Preoperative	Last follow-up	
1	47	M	Paraparesis, incontinence	Lt D4 SDAVFs	Onyx	5	5	Not improved
2	67	M	Paraparesis	Rt D4 SDAVFs	NBCA	3	2	-
3	47	M	Paraparesis, incontinence	Rt D12 SDAVFs	Onyx	4	2	Not improved
4	51	M	Paraparesis, incontinence	Lt D8 SDAVFs	Onyx	5	3	Improved
5	62	M	Paraparesis	Lt D10 SDAVFs	NBCA	5	5	-
6	51	M	Paraparesis	Rt D11 SDAVFs	Onyx	3	0	-
7	44	M	Paraparesis, incontinence	Lt L5 SDAVFs lateral sacral artery from Left IIA	Onyx	3	2	Not improved
8	23	F	Paraparesis	Rt L4 SDAVFs	Onyx	3	0	Improved
9	62	M	Paraparesis	Rt D5 SDAVFs	Onyx	4	2	-
10	45	M	Paraparesis, incontinence	Lt D6 SDAVFs	Onyx	5	5	Not improved
11	48	M	Paraparesis	Lt D8 SDAVFs	Onyx	3	0	-
12	51	M	Paraparesis, incontinence	Rt D11 SDAVFs	NBCA	5	4	Improved
13	47	M	Paraparesis, incontinence	Lt D10 SDAVFs	Onyx	5	4	Improved
14	64	M	Paraparesis, incontinence	Lt D7 SDAVFs	Onyx	5	5	Not improved
15	55	M	Paraparesis	Rt S1 SDAVFs (origin: Rt lateral sacral artery, Rt IIA)	NBCA	3	0	-

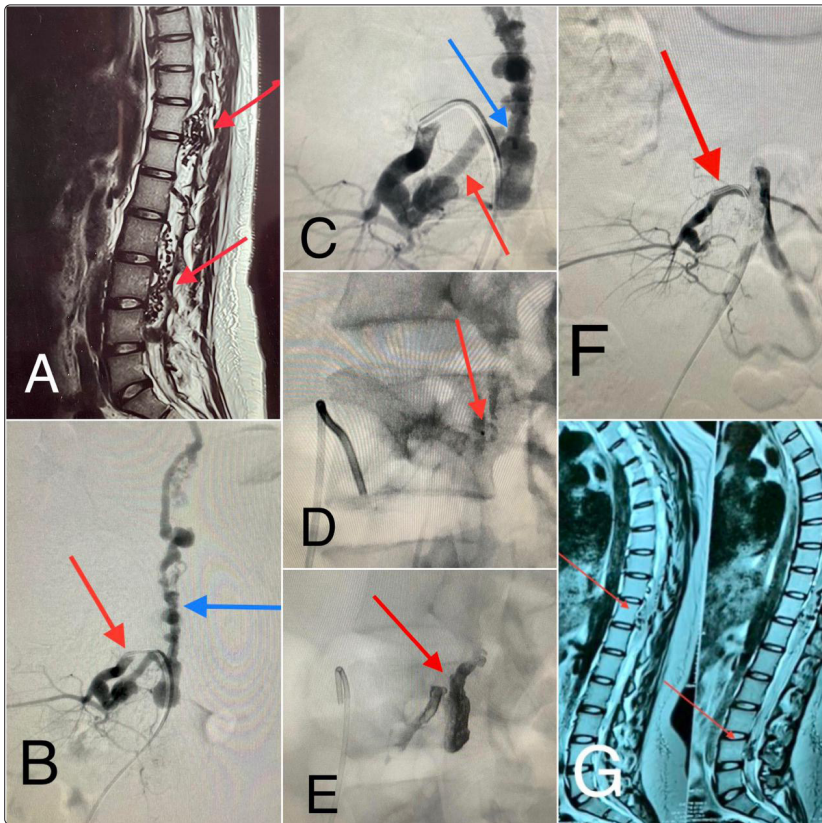
a support device and had good motor power before treatment. Regarding the eight patients with sphincter disturbances after treatment, three patients (37.5%) improved, and five patients had no improvement (62.5%).

It is crucial to mention that the case managed by Onyx (patient 9) had reestablishment of the fistula from a different level after three months of angiogram follow-up, with no recent clinical symptoms or even deterioration in his state at that time, no obvious morbidity, recurrence (except

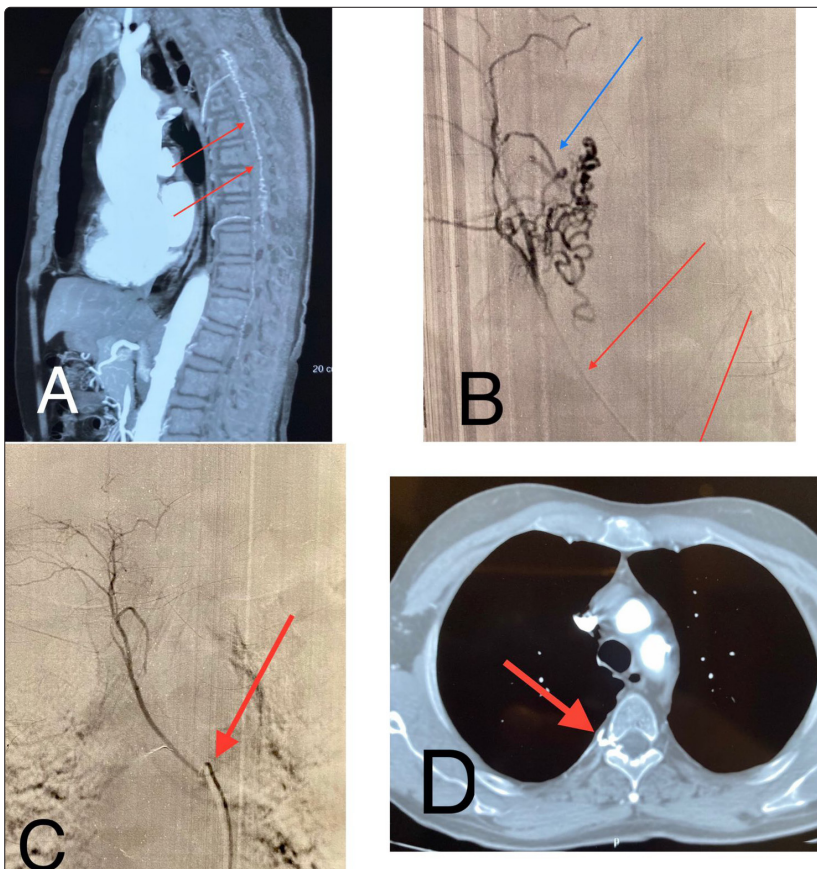
patient 9), or even mortality during follow-up.

On postoperative angiograms and MRI after 1 month, all cases except the one who developed arterial spasm had disappearance of the fistula on DSA, the disappearance of the edema of the cord, and signal void on MRI. However, there was no relation between the resolution of MRI signs and significant clinical improvement. Regarding long-term follow-up, we used the phone call interviews to simplify and none of our 15 patients were missed in our study follow-up.





**Figure 1.** A 23-year-old female patient presented with paraparesis and sphincteric disturbance. (A) T2 sagittal MRI 2 areas of entangled flow void signals inside the spinal canal suggesting an etiology of high-flow AV shunt; (B) red arrow: catheter in the right 4<sup>th</sup> lumbar artery, angiogram showing high-flow AV fistula between the radicular artery branch and the spinal vein (blue arrow); (C) oblique zoomed view of the radicular arterial branch (red arrow) and the blue arrow showing the foot of the vein in the AV shunt; (D) arrow shows the tip of the microcatheter at the fistulous point; (E) postembolization Onyx cast delineating the arterial feeder and the foot of the vein; (F) arrow points to the catheter in the lumbar artery during a control angiogram showing total cure of the AV fistula; (G) postembolization MRI showing marked reduction in the size of the vascular channels 16 months later.



**Figure 2.** A 67-year-old male patient presented with paraparesis. (A) CT angiogram of thorax shows a tortuous linear minute vascular channel in the spinal canal suggesting high-flow AV pathology; (B) red arrow shows the guiding catheter and the microcatheter accessing the right supreme intercostal trunk. Angiogram showing multiple radicular-dural arterial branches shunting into the spinal vein (blue arrow), with the latter assuming a varicoid appearance; (C) control angiogram via the guiding catheter (red arrow) showing a cure of the AV fistula, and (D) postembolization CT showing the glue cast in the right neural foramen 18 months later.

## DISCUSSION

SDAVFs are the direct connection between an artery (radicular-meningeal) and a vein (radicular-medullary).<sup>12,13</sup> Up to 80% of spinal vascular pathologies are caused by SDAVFs.<sup>17,14</sup> The majority of cases of SDAVF are idiopathic, yet acquired instances after trauma or surgery were described.<sup>16,19</sup> The most prevalent location of occurrence is the thoracolumbar area, with cervical SDAVFs being exceedingly rare. SDAVFs affect men predominantly over 40 years, with a ratio of male to female of 4:1.<sup>4</sup> The pathogenesis of myelopathy is a result of an increase in venous pressure caused by arterial shunting to the vein, leading to congestion in the spinal cord veins, lowering the cord arterial perfusion pressure, and finally causing cord ischemia and progressive myelopathy that is nonreversible in its end-stage.<sup>6</sup> That classic progression shows the following exceptions: 5% to 15% of cases with SDAVFs experience attacks of acute exacerbation of their complaints known as the “Foix-Alajouanine syndrome”; these attacks are thought to result from sudden thrombosis of the diseased veins draining the fistula; decline is unpredictable, and nonreversible loss of function may occur except if managed very early. In addition, a pattern of the acute progressive buildup of the condition with gaps of remission has been reported. If the condition is not managed early, it will cause progressive neurologic deterioration and marked impairment in 2–3 years, eventually leading to total paraplegia and incontinence.<sup>5,15</sup> The primary imaging that indicates the presence of the condition is MRI, which typically reveals a tortuous pattern of signal void in the subarachnoid region on T2-WI and a swollen edematous cord at a certain level manifested by abnormal elevated T2 intensity.<sup>1,12,19</sup> In weeks and months following surgery, removing the spinal AVF reduces spinal cord edema, causes the disappearance of the abnormal T2 increased intensity in the cord, and

abolishes enhancement of the pathologic vessels of the spinal venous plexus.<sup>19</sup>

The current study included 15 cases, with males accounting for more than 90% of the cases (14/14), which is in agreement with the studies of Rashad et al.<sup>19</sup> and Chibbaro et al.<sup>7</sup> In the studies of Gokhale et al.<sup>8</sup> and Andres et al.<sup>3</sup>, males accounted for 75% of the cases. The study of Ropper et al.<sup>21</sup> showed less manifestation of male predominance (11 males out of 15).

The age range of the current study is 45–74 years, in agreement with the age range of Rashad et al.<sup>19</sup> and Ropper et al.<sup>22</sup> studies, where the age range of the cases was 50–71 and 40–71 years, respectively. According to Andres et al.<sup>3</sup>, the age range was 44–77 years. The age range of Chibbaro et al.<sup>7</sup> and Gokhale et al.<sup>8</sup> studies was wider (20–76 and 24–76 years, respectively).

Progressive motor weakness represents the most common presentation of SDAVFs seen in up to 95% of patients, which is often associated with neurogenic claudication (worsening of weakness during physical exertion). Other manifestations of SDAVFs include pain, numbness, sphincteric disturbances, and sexual dysfunction.<sup>6</sup>

Regarding the presentation of the current study cases, all cases with progressive spastic paraparesis of different grades and 8 patients (53.3%) had sphincter disturbances. According to Ropper et al.<sup>21</sup>, the most common presentation was weakness (87%), followed by numbness (80%), paresthesia (60%), and bowel or bladder symptoms (47%). In the study of Gokhale et al.<sup>8</sup>, the most common presentation was back pain (93%) and lower extremity weakness (85%). Low back pain was the most common symptom in Andres et al.'s study.<sup>3</sup> According to Chibbaro et al.<sup>7</sup>, the most common presentation was lower extremity sensory disturbances (80%), followed by paresthesia (60%), medullary claudication (30%), paraplegia, and pain (20% each). In the study of Rashad and colleagues, all cases presented with progressive motor weakness (100%), followed by sphincteric disturbances (75%)<sup>19</sup>.



The follow-up of the current study showed that 33.3% of them showed good improvement after treatment, 40% showed moderate improvement and 26.7% showed poor improvement. In the study of Ropper et al.<sup>21</sup> and colleagues 45% of the cases improved after treatment, while 55% showed no improvement. In the study of Rashad et al.<sup>19</sup>, 16.7% of the cases showed good improvement, 58.3% showed moderate improvement, and 25% showed poor improvement. The failure rate of Andres et al.<sup>3</sup> cases was only 23.5%. On the other hand, Chibbaro et al.<sup>7</sup> claimed that they achieved cure in 100% of their cases.

Embolization with liquid is reported to have a very low rate of recurrence.<sup>9,3</sup> In this series, recanalization was encountered only in one case (6.7% was related to spasm), and we never delivered PVA in our case series. A reappearance of the DAVF at another level was never seen in our cases series except in one case (6.7%) based on the repeated MRI exams and many clinical follow-ups. The advantages of the embolization technique are the noninvasive nature and the option of an immediate angiogram test of the treatment. For that reason, we postulated it as the first choice in our study. Therefore, a primary trial of embolization with liquid can be justified. In this study, embolization did not push us through surgery. Other spinal DAVF series were reported by Ronald et al.<sup>20</sup> (49 cases), Song et al.<sup>24</sup> (27 cases), and Safaee et al.<sup>22</sup> (47 cases). In these studies, the primary success rate of embolization with liquid was reported between 30% and 90%. However, Ronald et al.<sup>20</sup> (90% primary success) stated that entry of the liquid embolic into the fistula without entry into the proximal vein is enough, which is seen in the high recurrence rate of 23%. In Song et al.'s<sup>24</sup> study, the Aminoff score was applied in 35 treated patients, supported by our series by a realization rate of 6.7% and a success rate of up to 73.3%.

Our study limitations were the small sample size due to the rarity of the pathology, being a retrospective, the short radiological follow-up

period, and the study design being single arm with no comparison to the surgical technique

## CONCLUSION

Spinal AVF could be treated safely and effectively by an endovascular approach. Generally, there is no correlation between the disappearance of MRI abnormalities and significant clinical improvement. The preclinical state of the patient is directly proportional to the clinical outcome. We recommend the intervention even if the patient is in bad clinical status because patients may show dramatic clinical improvements in some situations.

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## الملخص العربي

### عرض النتائج الإكلينيكية والمتابعة طويلة المدى لمراكز طبية متعددة بخصوص علاج الوصلات الشريانية الوريدية في غشاء الام الجافية بالحبل الشوكي عن طريق القسطرة

**البيانات الخلفية:** تعد الوصلات الشريانية الوريدية في الحبل الشوكي اهم سبب يمكن علاجه من أسباب الشلل المرتبط بالحبل الشوكي. وفي أغلب الأوقات تتواجد الوصلة في منطقة الفقرات الصدرية و القطنية و يعد الهدف من العلاج هو غلق منطقة الوصلة عن الطريق حق مادة صمغية من خلال قسطرة دقيقة توجه إلى المكان المحدد باستخدام الأشعة

**الغرض:** إثبات انه علاج وصلات الحبل الشوكي الشريانية الوريدية علاج آمن وفعال خاصة بعد المتابعة لفترة طويلة.

**تصميم الدراسة:** دراسة حالة سريرية بأثر رجعي.

**المرضى والطرق:** تم علاج 10 مريض ؛ 14 رجل وامرأة تتراوح أعمارهم من سن 40 عام إلى 74 عام ، تم علاجهم في جامعات : سوهاج و طنطا والإسكندرية وعين شمس ، ويعانون من أعراض مرتبطة بوجود وصلة شريانية وريدية بالحبل الشوكي مثل شلل الساقين وعدم التحكم في البول او البراز

**النتائج :** تم علاج الحالات جميعا بالقسطرة وكانت النتيجة أنه خمسة حالات تعافوا تماما وستة حالات تعافوا جزئياً و أربع حالات لم يتعافوا

**الخلاصة:** إذا تم علاج الوصلة الشريانية الوريدية في الحبل الشوكي سريعا ستكون النتائج جيدة وهي تعافى تام. حالة المريض قبل الحقن عنصر رئيسي في التنبؤ بالنتيجة. لابد من دراسة الحالة جيدا قبل العلاج. و يعد علاج القسطرة بديل مساوي للجراحة في العلاج.