International Journal of Surgery Case Reports 85 (2021) 106173

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr

Case series

Symptomatic subclavian steal syndrome: Report of four Moroccan cases and literature review

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ARTICLE INFO

Keywords:
Subclavian steal syndrome
Vertebral artery
Symptomatic

ABSTRACT

Introduction: Subclavian steal syndrome (SSS) is the hemodynamic phenomenon of blood flow reversal in the vertebral artery due to significant stenosis or occlusion of the proximal ipsilateral subclavian artery.
Materials and methods: Four patients with subclavian steal syndrome were treated in our center. Percutaneous radial approach was used for angioplasty, primary stenting of subclavian artery was performed, surgical techniques in particular carotid-subclavian bypass and carotid-subclavian transposition were used.
Results: We report the cases of four patients, three of which are male, with an average age of 60 years. All of them were symptomatic. Diagnosis was made by duplex ultrasound, supplemented by CT-angiography and arteriography. Endovascular treatment was attempted in all four patients, which was successful in two patients, who underwent primary stenting, and failed for the two others, for whom surgical treatment was considered. One had a subclavio-carotid bypass graft with a polytetrafluorethylene (PTFE) prosthesis and the other had a subclavio-carotid transposition. The technical results were satisfactory in all patients with symptoms resolution. The postoperative evolution was without notable complications and the postoperative checkups were satisfactory.
Discussion: There are excellent screening tools and effective medical therapies which can be instituted if the SSS is diagnosed early. When the need for revascularization arises, percutaneous modalities are favored given their proven long-term efficacy, decreased morbidity and mortality, and cost-effectiveness. Nevertheless, large, prospective, randomized and controlled trials are needed to compare the long-term patency rates between the endovascular and surgical techniques.

1. Introduction

Subclavian artery steal syndrome (SSS) is a hemodynamic phenomenon caused by the reversal of blood flow in the vertebral artery (VA) associated with significant stenosis or occlusion of the proximal ipsilateral subclavian artery that can result in significant vertebrobasilar ischemia [1].

Clinically it manifests by symptoms of vertebrobasilar insufficiency, ischemic symptoms of the upper limbs and recurrent angina, especially in patients who have undergone coronary bypass surgery with the internal mammary artery.

In the absence of associated lesions of other cerebral arteries, this syndrome remains asymptomatic [2].

Cervical Doppler ultrasound and transcranial Doppler are the ultimate screening tools for diagnosis, supplemented by CT angiography or MRI angiography.

The management of the SSS has for a long time remained the prerogative of surgical treatment, the evolution and improvement of endovascular techniques has fundamentally changed the management of subclavian steal syndrome which has become the therapeutic approach often used as a first-line treatment.

We report through this work four observations of symptomatic subclavian steal syndrome treated in the vascular surgery department, University Hospital Ibn Sina, Rabat, Morocco. We will discuss the different methods of diagnosis and management of this pathology while recounting the results of various studies and recent publications concerning this syndrome.

The work has been reported in line with the SCARE criteria and cite the following paper [3]: Agha RA, Sohrabi C, Mathew G, Franchi T, Kerwan A, O'Neill N for the PROCESS Group. The PROCESS 2020 Guideline: Updating Consensus Preferred Reporting Of CasE Series in Surgery (PROCESS) Guidelines, International Journal of Surgery

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https://doi.org/10.1016/j.ijscr.2021.106173
Received 24 March 2021; Received in revised form 3 July 2021; Accepted 6 July 2021
Available online 7 July 2021

2020;84:231–235.

2. Case presentation 1

The procedure was performed by the chief of the vascular department with 30 years of experience.

67-year-old female patient with no pathological history, presented to the vascular surgery consultation for sudden occurrence of two episodes of vertigo and fainting with the loss of balance and visual fog, preceded by temporal headaches and hot flashes. Physical examination revealed an abolition of the pulses of the left upper limb with a difference of 20 mmHg between upper extremity systolic blood pressures and the presence of a murmur in the left supraclavicular region. No neurological deficit was reported. The 12-lead electrocardiogram was normal. Laboratory examinations were within normal limits.

Patient initially underwent a normal brain CT scan, Doppler ultrasound of the cervical arteries demonstrated a severe stenosis of the left pre-vertebral subclavian artery with additional permanent retrograde blood flow on the ipsilateral vertebral artery, and the presence of a loop of the right vertebral artery without stenosis. A CT angiogram of the cervical arteries was performed and has demonstrated a pre-vertebral sub occlusive stenosis of the left subclavian artery located 10 mm from its birth extended over 15 mm. Patient was admitted for endovascular treatment. Angioplasty with direct stenting of the lesion without predilation was performed (Fig. 1). Through an ultrasound-guided puncture of the left radial artery, a 7 mm × 27 mm balloon expandable stent (ev3 Visi-Pro*) was deployed with good final aortography control. The patient has been discharged one day after the intervention with dual antiplatelet medication: clopidogrel 75 mg/day for a period of three months and acetylsalicylic acid 75 mg/day for life. The evolution is marked by the disappearance of symptoms and the absence of restenosis in the 1-year and 2-year postoperative duplex ultrasound.

3. Case presentation 2

The procedure was performed by the chief of the vascular department with 30 years of experience.

A 48-year-old male patient was referred from cardiology department to our department following failed catheterization for coronary angiography in the lower and upper limbs. His medical history included active smoking for 30 years, ischemic heart disease with left ventricular dysfunction with 32% ejection fraction under medical treatment.

The patient reported recurrent vertigo, both lower extremities and left upper extremity muscle weakness for the past month. Physical examination revealed an abolition of the pulses of the lowers extremities and left upper limb, with a difference of 15 mmHg between upper extremity systolic blood pressures and no neurological deficit. The 12-lead electrocardiogram showed left atrial and ventricular hypertrophy with secondary repolarization disorders. Laboratory examinations were unremarkable. A duplex ultrasound of the cervical arteries revealed a sub occlusive stenosis of the origin of the left subclavian artery responsible for an intermittent subclavian steal syndrome, reversed blood flow in the ipsilateral vertebral artery and an amortization of the upper left limb artery flows. CT angiography of the lower limbs showed thrombosis of the renal abdominal aorta spreading to the primary iliac arteries.

Patient was admitted for an endovascular treatment. Due to his heart disease and aortic thrombosis, catheterization of the left subclavian artery was performed by ultrasound-guided puncture of the left radial artery and under local anesthesia. A direct stenting of the lesion without predilation was performed with a 7 mm × 39 mm balloon expandable stent (Cordis Genesis*) maintaining vessel patency of the ipsilateral vertebral and CCA with good final aortography control (Fig. 2). Patient was discharged the next day receiving dual antiplatelet medication: clopidogrel 75 mg/day for a period of three months and acetylsalicylic acid 75 mg/day for life. After 30 months of follow-up, no restenosis was observed and the patient reported resolution of his symptoms.

4. Case presentation 3

The procedure was performed by the chief of the vascular department with 30 years of experience.

It's a 65-year-old man, his medical history included diabetes on insulin and ischemic heart disease under medical treatment. The patient was admitted to the emergency department for intense throbbing and sleepless pain of the upper left limb for 15 days associated with coldness and appearance of trophic disorder in the fingers. The patient also reported the occurrence of two episodes of drop-attacks. Physical examination revealed an abolition of the pulses of the left upper limb with a difference of 30 mmHg between upper extremity systolic blood pressures, and a necrosis of the pulps of the 2nd and 3rd left fingers. No neurological deficit was observed. The electrocardiogram and the laboratory examinations were normal. Duplex color ultrasound objectified a prevertebral stenosis of the left subclavian artery with retrograde blood flow on the ipsilateral vertebral artery, findings indicating the development of an intermittent subclavian steal syndrome. It also showed thrombosis of the ulnar artery with a barely perceived flow at the radial artery. A CT angiography of the cervical arteries was also performed showing a sub occlusive stenosis of the left subclavian artery at its proximal portion.

Following the endovascular treatment failure (Fig. 3A) in this patient and given the short and deep nature of the pre vertebral subclavian artery, a sub-clavio-carotid bypass in PTFE prosthesis was performed by left subclavicular approach (Fig. 3B). Patient was discharged after five days receiving one antiplatelet medication, based on acetylsalicylic acid 75 mg/day for life. During 3 years of follow-up, no thrombosis of the bypass was observed and the patient reported resolution of his symptoms.

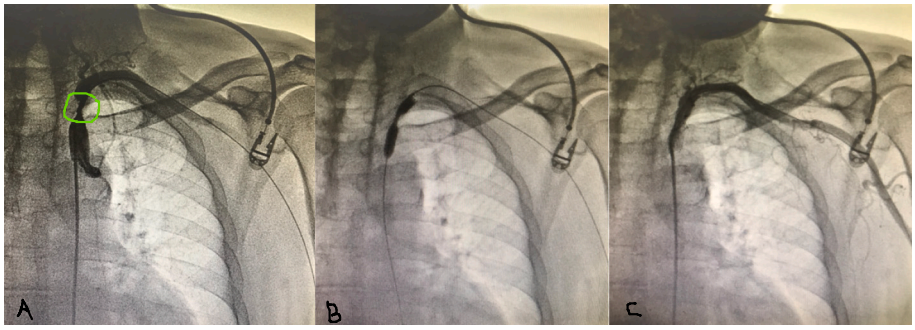

Fig. 1. Intraoperative angiogram showing: stenosis of the proximal left subclavian artery (A), success of recanalization with release of a balloon expandable stent (B) satisfactory angiographic control (C).

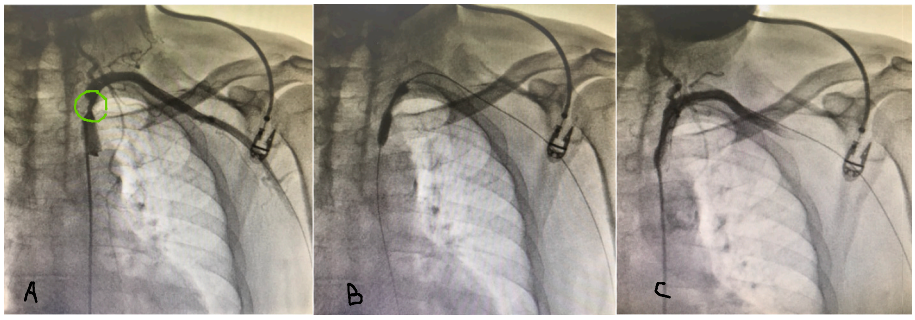

Fig. 2. Intraoperative angiogram showing: prevertebral stenosis of the left subclavian artery (A), release of the balloon expandable stent (B) successful subclavian artery stenting (C).

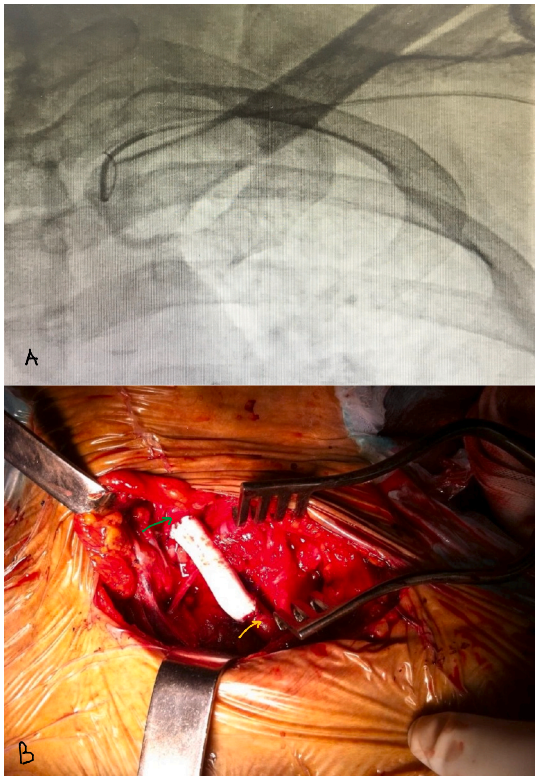

Fig. 3. Angiogram showing failure of recanalization of the left subclavian artery by radial transcutaneous puncture (A), intraoperative view showing the bypass in PTFE prosthesis between the left subclavian (yellow arrow) and the left common carotid artery (green arrow) (B). (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

5. Case presentation 4

The procedure was performed by the chief of the vascular department with 30 years of experience in vascular surgery.

A 55-year-old man, with medical history of smoking, was admitted to vascular consultation for a weakness and paresthesia of the left upper limb on exertion, he was a writer by profession. Physical examination revealed an abolition of the pulses of the left upper limb with a difference of 18 mmHg between upper extremity systolic blood pressures. Laboratory examinations and cardiac assessment were normal. Duplex color ultrasound objectified a prevertebral stenosis of the left subclavian artery with retrograde blood flow on the ipsilateral vertebral artery. Endovascular treatment by approaching the left radial artery was attempted but failed. The patient underwent surgical treatment with subclavio-carotid transposition (Fig. 4). Patient was discharged after four days receiving one antiplatelet medication, based on acetylsalicylic acid 75 mg/day for life. After one year of follow-up, the patient reported the disappearance of symptoms, he was very satisfied.

6. Discussion

Arterial disease of the upper limbs is less common than that of the lower limbs, it is most often asymptomatic and rarely severe due to the development of sufficient collateral circulation in most cases. When it becomes symptomatic, it is manifested by ischemic symptoms of the upper limbs and often by symptoms of vertebrobasilar insufficiency [2,4]. These are secondary to stenosis or occlusive lesions of the subclavian artery in its pre-vertebral portion leading to retrograde flow in the ipsilateral vertebral artery, defining the subclavian steal syndrome.

This retrograde flow was recognized and described first in 1960 by Contorini [5], in patient presented with absent radial pulse. In 1961, Reivich [6] established a correlation between clinical neurological manifestations and the subclavian steal syndrome. The term “subclavian steal syndrome” was formulated by Fisher in the same year [7].

The prevalence of SSS is 0.6% to 6.4% in the general population [8,9]. Fields et al. in a meta-analysis of 6534 polyvascular patients found 168 cases of SSS (2.5%) with 5.3% symptomatic patients [10].

The most recent large prevalence study was conducted by Labropoulos et al. in 2010 about 7881 polyvascular patients presenting for duplex ultrasound of the carotid arteries, it showed 5.4% cases of SSS

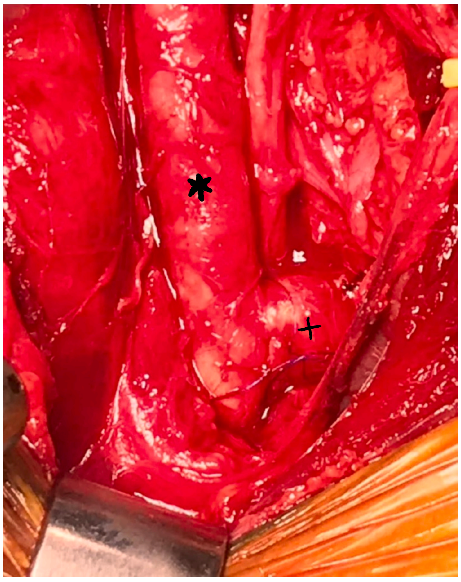

Fig. 4. Intraoperative view showing the transposition of the left subclavian artery (+) to the left common carotid artery (*).

[11].

Lesions predominate on the left side in 82.3% of cases [8,11,12]. It is explained by the acute angle at the origin of the left subclavian artery which makes the blood flow more turbulent accelerating the constitution of the lesions at the junction between the aortic arch and the left subclavian artery [13]; the small diameter of the left subclavian artery compared to the innominate artery may also explain this predominance.

Men are more affected than women with a sex ratio of around 2/1. In our study 3 patients were male.

Physio pathologically, the pressure is higher at the origin of the vertebral artery than at its termination and circulation is established from the origin to the basilar artery. The significant reduction in the diameter of the proximal subclavian artery leads to inversion of the pressure gradient. Hence, inversion of the blood flows at the ipsilateral vertebral artery.

SSS is most often asymptomatic, an incomplete circle of Willis or simultaneous presence of a contralateral carotid or vertebral stenosis can interfere with the cerebrovascular support system which results in the onset of symptoms [14–16].

In symptomatic patients, the SSS may have different clinical aspects: signs of upper limb ischemia manifested by pain or fatigue during exercise, paresthesia, intermittent coldness and sometimes trophic disorders [17,18]; and symptoms of vertebrobasilar insufficiency secondary to a flow reversal in the ipsilateral vertebral artery, these symptoms are often transient, such as paroxysmal dizziness, diplopia, ataxia, dysarthria, syncope and cerebellar syndrome. The drop-attacks are also quite characteristic of vertebrobasilar insufficiency but not pathognomonic [17,19].

Another symptom that should suggest SSS is recurrent angina after myocardial revascularization related to a coronary-subclavian steal phenomenon [20,21]. A steal syndrome may also occur in dialysis patients with an ipsilateral arteriovenous fistula [4]. Recently, a case of Room tilt illusion was also attributed to an SSS [22].

Atherosclerosis is the most common cause of subclavian stenosis and, thus, steal syndromes [11,23,24]. Other possible etiologies are: large artery vasculitis, especially Takayasu's disease, birth defects of the aortic arches such as a right-sided aortic arch with an isolated left subclavian artery [25], this malformation is often associated with other congenital heart anomalies, in particular the tetralogy of Fallot [26]. A left aortic arch can lead to a left or right SSS in case of hypoplasia or atresia of the proximal segment of the subclavian artery, or a right subclavian artery isolated from the brachiocephalic artery. To our knowledge, approximately 40 cases of SSS related to congenital aortic arch malformations have been reported in the literature [27].

Other causes are: radiation arteritis, Horton disease, thoracic outlet syndrome, fibromuscular dysplasia and stenosis after surgical repair of aortic coarctation. In our cases, Atherosclerosis was the cause of SSS in all the patients.

Difference in systolic blood pressure of the upper limbs>15 mmHg during the physical examination is an atypical finding, and should look for an SSS. An abolition of the peripheral pulses of the upper limb and the presence of a supraclavicular bruit should also suspect an SSS [28]. All of these signs should prompt additional radiological examinations.

Color duplex scan of the cervical region is considered by many authors as the ultimate screening tool [29]. It provides information on the morphology of the lesion and specifies its hemodynamic impact, thus, the study of the vertebral arteries and the carotid axes. It's an operator dependent examination. Color duplex scan can differentiate 3 grades of SSS: latent steal, intermittent steal and permanent steal.

CT angiography and MRI angiography of cervical arteries are tow examinations that allow the assessment of lesions and study of the brain parenchyma and adjacent structures. They complete the results of color duplex scan.

Conventional contrast arteriography has long been considered the standard tool for the assessment of cervical arteries. Nowadays, it is indicated in case of discrepancy between two noninvasive techniques, or a suspicion of an associated lesion of the carotid siphon, and for providing the possibility of an endovascular treatment after initial confirmation of the diagnosis.

Management of patients with SSS depends on several parameters, and includes either medical treatment alone or associated with a surgical or endovascular revascularization. Treatment of symptomatic SSS is always indicated, percutaneous transluminal angioplasty with or without stenting represents the treatment of choice for symptomatic SSS, surgery is especially indicated in case of failure of the endovascular approach or its unavailability. Surgery includes subclavio-carotid transposition, carotid-subclavian bypass and carotid-axillary bypass.

In a retrospective study of 110 patients (102 subclavian angioplasty with 90 stenosis and 20 occlusions) by De Vries et al., the technical and clinical success rate was 93% and the primary and secondary patency rates were 77% and 91.7% respectively at 2 years [30].

In Patel et al. in a series of 170 patients who underwent subclavian artery stenting, the technical success rate was 98%, the primary and secondary patency rates were 83% and 96%, respectively over a 5-year follow-up [31].

Cina et al. in a meta-analysis of 1027 patients compared subclavio-carotid transposition (SCCT) to carotid-subclavian bypass, they found a long-term patency rate of SCCT of 99% compared to that of bypass which was 84% [32].

Six studies, published between 1989 and 2012 to date, have compared the results of an endovascular technique (balloon angioplasty, angioplasty with expanding stent, angioplasty with self-expanding stent) to surgical revascularization of the proximal subclavian artery. They concluded that both techniques are effective and safe in the short and medium term. The long-term results were assessed just for open surgery [33–38].

Simple angioplasty patency rates were between 20% and 50% in case of occlusion. With stenting the patency rate increased and varied between 70% and 100% according to Queral and Criado [39].

Rodriguez-Lopez et al. reported primary and secondary patency rates of 93% and 90%, respectively, in 69 patients with occlusive and symptomatic lesions of the proximal subclavian artery treated with angioplasty [40].

A recent meta-analysis comparing angioplasty and stenting that included 1726 patients found significantly higher recanalization rates with stenting (92.8% vs. 86.8%, $P = .007$). However, long-term patency of the vessel, symptom alleviation, and stroke or death occurrence were not statistically different between the two groups [1].

Medical treatment based on antiplatelet aggregation and control of cardiovascular risk factors is always recommended.

In our cases endovascular treatment was the first-line approach in all patients, failure of recanalization in the 3rd and the 4th patient led to surgical treatment.

7. Conclusion

SSS is a rare entity and is often underestimated due to the asymptomatic nature of the lesions. Revascularization is always indicated in symptomatic patients.

Endovascular techniques should be tried first. Surgery is tried in case of failure or unavailability of endovascular techniques.

The choice between angioplasty alone or with stenting must be made on a case-by-case basis depending on the type and extent of the lesion, as well as the patient's comorbidities.

Ethical approval

The study is exempt from ethnical approval.

Funding

None.

Author contribution

EL BHALI Hajar: conception, methodology, data curation, data collection and writing the paper.

BOUNSSIR Ayoub: data collection and data analysis.

BAKKALI Tarik: data analysis and data curation.

JDAR Asmae: data collection,

EL KHLOUFI Samir: critical revision and final approval.

LEKEHAL Brahim: critical revision and final approval of the version to be submitted.

Consent

Written informed consent was obtained from the patients for publication of those cases reports and accompanying images. A copy of the written Consent is available for review by the Editor-in-chief of this journal on request.

Registration of research studies

N/A.

Guarantor

El Bhali Hajar.

Declaration of competing interest

The authors have not declared any conflict of interest.

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