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Case report

Treatment of ruptured subclavian steal flow-related vertebrobasilar junction aneurysms: Case report on surgical and endovascular considerations from two cases

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ABSTRACT

Introduction: Subclavian steal phenomenon causes retrograde flow through the vertebral artery, ipsilateral to the affected subclavian artery, which rarely leads to flow-related vertebrobasilar junction (VBJ) aneurysms.
Case descriptions: We describe two cases of subarachnoid hemorrhage from such ruptured aneurysms in which the retrograde flow direction in the vertebral artery complicated surgical and endovascular treatment.
Discussion: Reversed flow in the vertebral artery, ipsilateral to the stenotic subclavian artery leads to a lack of proximal control in surgical clipping of these VBJ aneurysms and jeopardizes stability of coil and stent placement in endovascular aneurysm treatments in this setting.
Conclusion: From these 2 experiences over 7 years, treatment considerations emerged for future cases.

1. Introduction

A subclavian steal phenomenon occurs when blood flow to the upper extremity is provided by retrograde flow in the ipsilateral vertebral artery caused by a stenotic lesion of the subclavian artery proximal to the origin of the vertebral artery [1].

Sometimes this subclavian steal *phenomenon* leads to a subclavian steal *syndrome* if clinical signs or symptoms arise, typically presenting with arm claudication, dizziness or even syncope from vascular steal from the vertebrobasilar circulation. Exceptionally rarely this leads to aneurysm formation in the vertebrobasilar circulation like fusiform vertebral artery aneurysms [2,3], saccular aneurysm of intervertebral metameric collaterals [4] or vertebrobasilar junction (VBJ) aneurysms [5,6]. We only found two reports thus far of a ruptured wide-necked vertebrobasilar junction (VBJ) aneurysm treated by stent-assisted coiling [5,6].

Stability of the coils in such aneurysms is hard to achieve due to the strong retrograde flow, while stent-assisted coiling should be avoided in ruptured aneurysms because of the associated higher complication rate [7]. We present our surgical and endovascular experience in two consecutive patients over 7 years of time in a retrospective, single-center case series. Lessons learned prompted us to conceptual considerations for the treatment of future cases. This case report has been reported in line with the SCARE Criteria [8].

2. Case presentation

2.1. Patient 1

A 73-year-old man presented with worsening of non-traumatic

Abbreviations: VBJ, vertebrobasilar junction; CT, cerebral computed tomography; DSA, digital subtraction angiography; MRA, magnetic resonance angiography.

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headache, neck pain and somnolence after an episode of feeling generally unwell during a walk four days earlier. Medical history revealed hypertension and hyperlipidemia.

Cerebral computed tomography (CT) showed a Fisher III subarachnoid and intraventricular hemorrhage with a substantial amount of blood in the prepontine cistern caused by a ruptured left distal vertebral artery aneurysm adjacent to the vertebrobasilar junction (Fig. 1a and b).

A digital subtraction angiography (DSA) revealed a proximal left subclavian artery occlusion with retrograde flow through the left vertebral artery (Fig. 2). The aneurysm was located on the inferomedial side of the left V4 segment close to the vertebrobasilar junction and had a width of 4.9 mm, a height of 4.6 mm and a neck of 4.5 mm (Fig. 3) with a small bleb on the lateral side. Also, a small aneurysm on the posterior wall of the top of the right carotid artery was seen, measuring 1.5 mm in diameter, 1.7 mm in length and a neck of 1 mm. The location of the blood, the presence of the bleb and the size of the aneurysm made the vertebral artery aneurysm the most probable cause of the hemorrhage.

Endovascular treatment was performed via a right femoral approach, a 6 fr sheath was advanced into the proximal right V2 segment, an intermediate catheter (Navien 072, ev3, Irvine, CA, USA) was navigated into the distal V2 segment. Heparin was administered (5000 IU). The aneurysm was catheterized with a microcatheter (Echelon 10, 90 degree tip angle (ev3, Irvine, CA, USA)) over a Transend guidewire (Boston Scientific, Global Park Heredia, Costa Rica). During deployment of the first coil the coil dislocated repeatedly into the parent artery lumen due to the wide neck and the strong retrograde flow. Therefore, an Eclipse 9 balloon catheter (Balt extrusion, Montmorency, France) was placed over the neck of the aneurysm using a Traxcess guidewire (Microvention, Saint-germain-en-laie, France). However, during inflation, the balloon also dislocated easily due to the high retrograde flow (Fig. 4). After repeated attempts, a stable position of the balloon was established. A 4/12 coil (Cosmos Complex; Microvention Saint-germain-en-laie, France) was introduced and consecutively 3/6, 3/4 and 1.5/4 coils (Hypersoft 3D and Hypersoft Helical; MicroVention, Saint-germain-en-laie, France). A small neck remnant remained open after the coiling procedure (Fig. 5). The puncture site was closed with an angioseal (Terumo, Somerset, NJ, USA).

2.1.1. Follow-up and outcome

The patient recovered uneventfully. MRA after 9 months showed coil impaction. At that time, restoration of the anterograde flow direction was decided upon by recanalization and stenting of the proximal left subclavian artery, after which a stable neck-remnant was visible on follow-up MRA for more than 3 years now. The patient gave written informed consent.

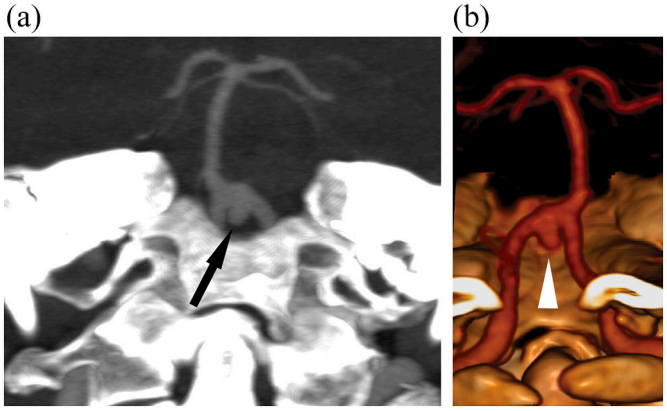

Fig. 1. a) Coronal maximum intensity projection of 7 mm of CT angiography exam, arrow points to the VBJ aneurysm, b) volume rendering of CT angiography exam, dorsal view, arrowhead points to the wide necked VBJ aneurysm.

2.2. Patient 2

A 73-year old woman presented with a thunderclap headache and Glasgow Coma Scale of 11/15. A Fisher grade III subarachnoid hemorrhage was diagnosed on CT with intraventricular blood and a hydrocephalus for which a ventricular drain was inserted. The subarachnoid hemorrhage was caused by a ruptured wide neck VBJ aneurysm pointing laterally with aneurysm width of 5.9 mm and a neck of 5 mm (Fig. 6). A proximal subclavian artery occlusion was present on the left side which caused strong retrograde flow in the left vertebral artery (Fig. 7).

Under general anesthesia an attempt was made to treat this aneurysm by coiling, however, the high retrograde flow and the wide neck caused repeated dislocation of the coils, which made endovascular treatment impossible. Stent placement was deemed less attractive in this ruptured case. Therefore, the next day, the patient was operated upon through a left, far-lateral, juxtacondylar approach for successful surgical clipping of the vertebral confluence aneurysm. Due to an intra-operative rupture of the aneurysm, the left glossopharyngeal and vagal nerve was functionally damaged.

The patient experienced a protracted postoperative course with a pneumonia, metabolic acidosis from total parenteral nutrition, dysphagia and hypophonia. Despite this, the patient's motor function and cognitive ability recovered well. Six months postoperatively, the patient was independent for activities of daily living, speaking well after an additional vocal cord plasty but with a permanent percutaneous endoscopic gastric tube. A 6-months and a 5-years follow-up CT angiography (Fig. 8) both showed a stable result without recurrence of the aneurysm. The patient deceased 11 years later from an unrelated cause. Before her death, the patient gave verbal consent to the authors and the family confirmed this in a written consent before publication.

3. Discussion

We present two older patients with wide neck, saccular, vertebral artery aneurysms in a remarkably similar location adjacent to the vertebrobasilar junction. Both patients had an asymptomatic subclavian steal phenomenon with flow reversal in the ipsilateral vertebral artery but suffered from a subarachnoid hemorrhage caused by a ruptured flow-related aneurysm at this VBJ location (Fig. 9).

Endovascular treatment by coiling alone was attempted in both patients, however the coils repeatedly herniated into the parent artery lumen because of the wide neck of the aneurysm and the strong retrograde flow. In one patient coiling was finally possible with the use of a balloon. An alternative technique to prevent coil dislocation has been described by Horowitz et al. by using stent-assisted coiling [5]. However, in case of a ruptured aneurysm, stent-assisted coiling is not attractive because of the need to start a dual antiplatelet regimen directly after stent placement [7]. Furthermore, the endovascular approach by balloon assisted coiling was not durable and lead to coil impaction on 9 months follow-up. After restoration of the anterograde flow-direction in the vertebral artery by elective transradial stenting of the ipsilateral proximal subclavian artery stenosis, follow-up showed a stable neck remnant.

Also surgical clipping of these aneurysms in this particular setting can be demanding: especially given the lack of proximal control, the retrograde flow in the vertebral artery on the ipsilateral side of the surgical approach has the potential to seriously complicate the procedure.

The strong retrograde flow in the ipsilateral vertebral artery creates problems for both the endovascular treatment as well as the surgical treatment. Therefore, in future cases we would consider emergency treatment of the subclavian artery occlusion either endovascularly (recanalization and stent placement) or surgically (creation of carotidosubclavian bypass), immediately before aneurysm repair.

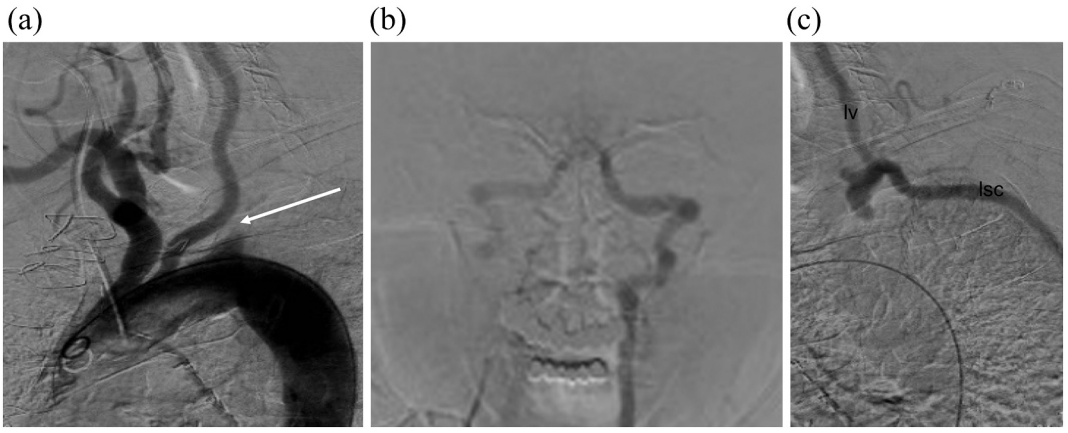

Fig. 2. Digital subtraction angiography (a, b, c) showing postostial left subclavian artery occlusion (arrow) with retrograde flow through the left vertebral artery (lv) to the left subclavian artery (lsc).

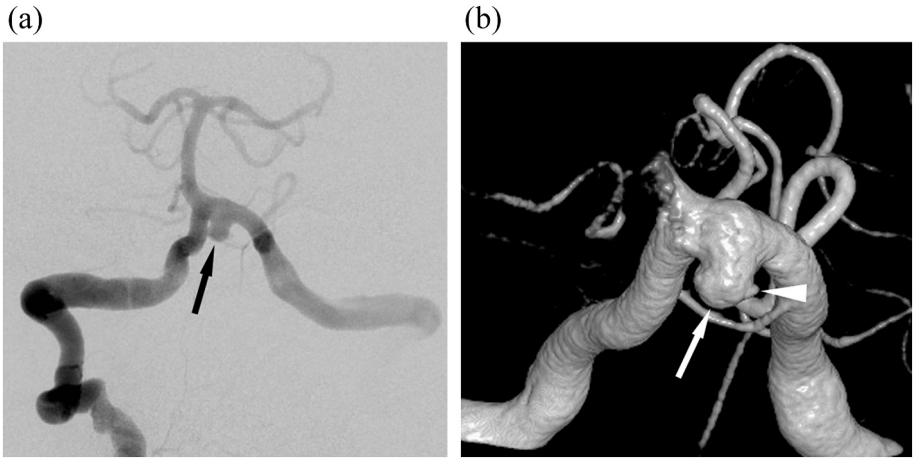

Fig. 3. Digital subtraction angiography in anteroposterior projection (a) and 3D angiography (b) showing the aneurysm (arrow) on the inferomedial side of the left V4 segment close to the vertebrobasilar junction with a width of 4.9 mm, a height of 4.6 mm and a neck of 4.5 mm with a small bleb on the lateral side (arrowhead).

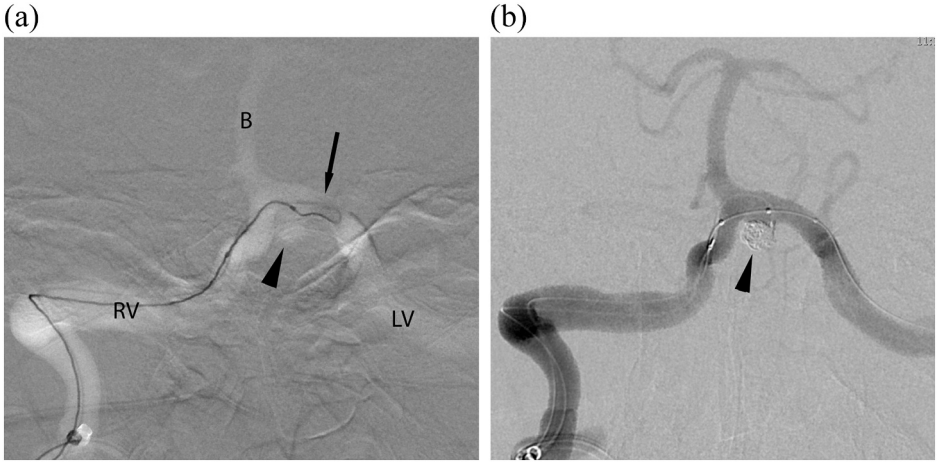

Fig. 4. a) Tip of guiding catheter distal in V2 segment of right vertebral artery (RV), coil (arrow) herniates from VBJ aneurysm (arrowhead) in lumen of left vertebral artery (LV). b) Successful balloon assisted coil embolization of the VBJ aneurysm (arrowhead).

4. Conclusion

We describe two cases of ruptured VBJ aneurysms in older patients with an asymptomatic subclavian steal phenomenon. Both surgical and endovascular treatment were complicated by the strong retrograde flow in the vertebral artery. Furthermore, endovascular treatment only resulted in a durable aneurysm exclusion after additional subclavian artery stenting.

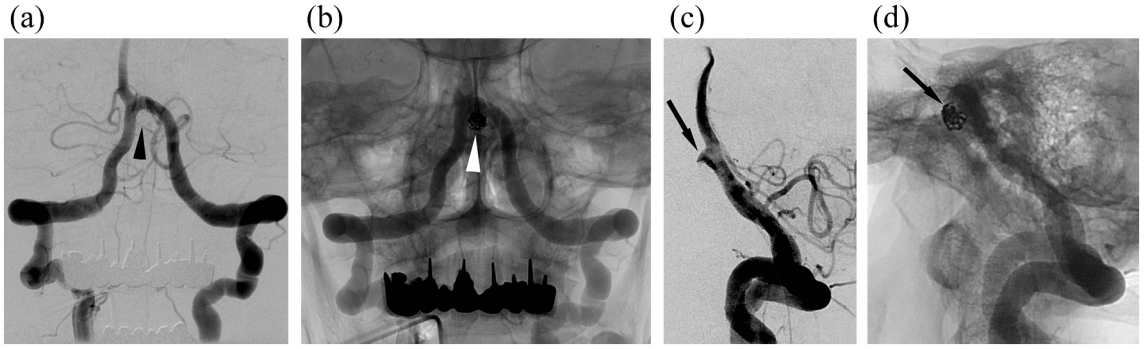

Fig. 5. Angiography after the placement of coils in the aneurysm, AP-projection with (a) and without (b) subtraction, arrowhead points to the coils. A small neck remnant (arrow) was present, visible on the lateral projection with (c) and without subtraction (d).

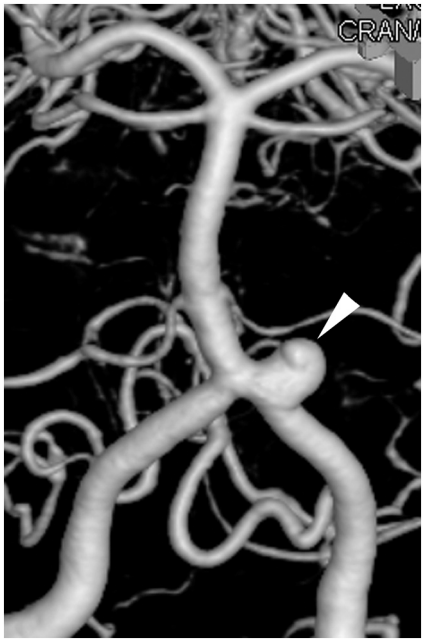

Fig. 6. 3D angiography after contrast injection in the right vertebral artery shows the laterally pointing wide necked VBJ aneurysm (arrowhead) in patient 2, aneurysm width is 5.9 mm, the neck measures 5 mm.

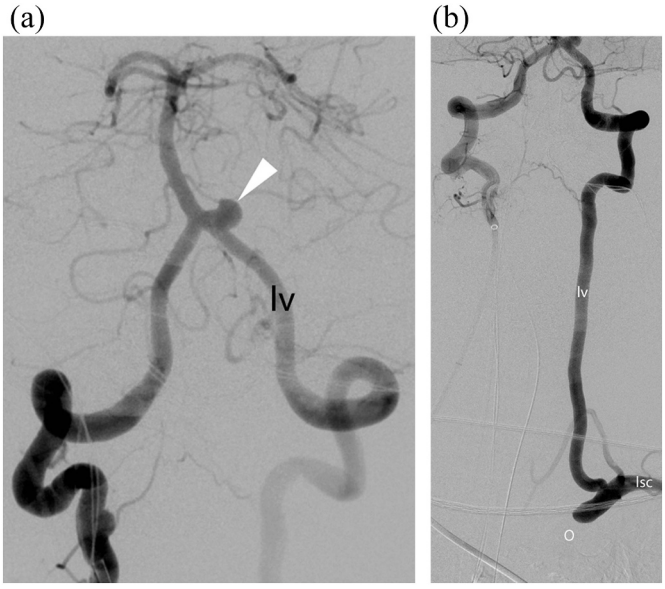

Fig. 7. Selective angiography of the right vertebral artery (a, b) shows retrograde flow in the left vertebral artery (lv) due to proximal left subclavian artery (lsc) occlusion (o). The white arrowhead points to the VBJ aneurysm.

Consent

As stated in the manuscript and complying with the SCARE 2020 guidelines, written informed consent has been obtained as detailed in the rebuttal letter and stated and specified in the manuscript.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Ethical approval

This study is exempt ethical approval of the University Hospitals Leuven as a retrospective case report.

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All authors.

Research registration number

1. Name of the registry: researchregistry
2. Unique identifying number or registration ID: 7475
3. Hyperlink: https://www.researchregistry.com/browse-the-registry.

CRediT authorship contribution statement

Sandra Adriana Cornelissen was involved in the endovascular treatment of the first patient, acquiring the images, the drafting of the article, preparation of the images and the final approval of the manuscript.

Sam Heye was involved in the endovascular treatment of the first patient, acquisition of the images, drafting of the article and the final approval of the manuscript.

Geert Maleux was involved in the endovascular treatment of the second patient, acquisition of the images, drafting of the article and the final approval of the manuscript.

Kim Daenens was involved in the surgical treatment of the first

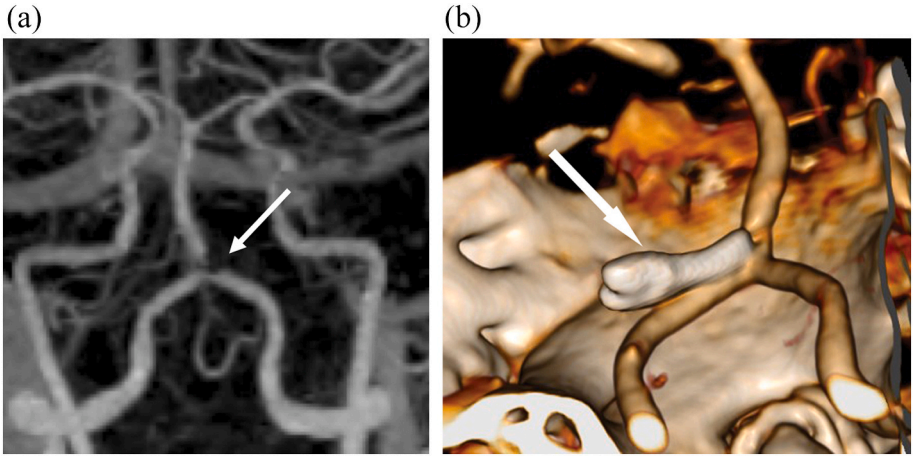

Fig. 8. Follow-up imaging by CTA of the clipped VBJ aneurysm. In a) coronal maximum intensity projection of a dual energy CTA with bone subtraction 6 months after treatment. The impression of a proximal basilar flow obstruction is caused by artifact of the Lazic clip (arrow) b) volume rendering (dorsal view) of CTA 5 years after clipping, arrow points to the clip. Both show a complete exclusion of aneurysm.

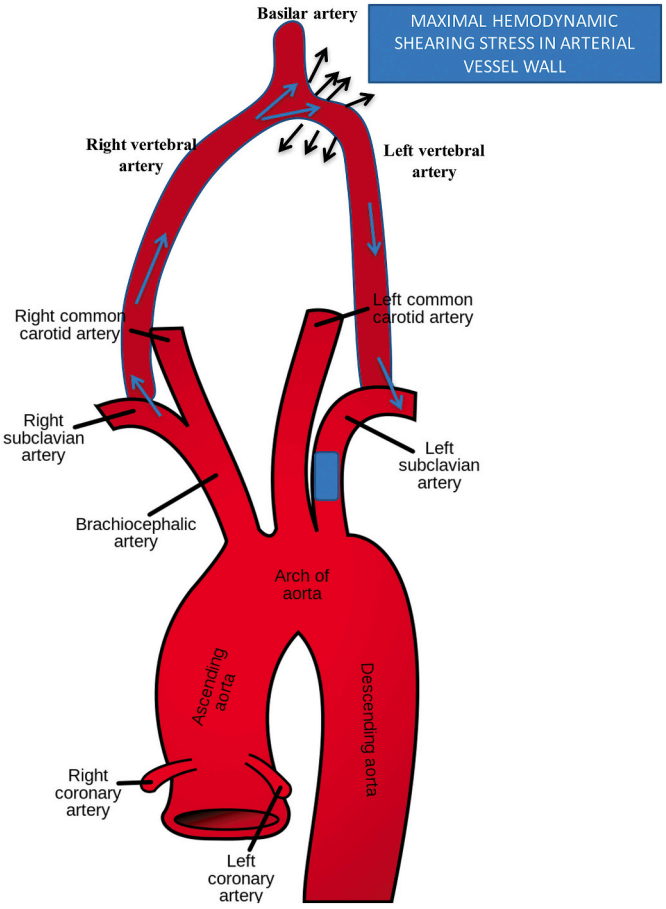

Fig. 9. Illustration of the pathophysiological mechanism leading to a flow-related left vertebral junction aneurysm in subclavian steal syndrome due to an obstruction (blue square) on the left subclavian artery, proximal to the origin of the left vertebral artery. Direction of intravascular flow is indicated by the blue arrows. Increased shearing stress indicated by black arrows.

patient, acquisition of the images, drafting of the article and the final approval of the manuscript.

Johannes van Loon was involved in the overall treatment of the 2 patients, drafting of the article and the final approval of the manuscript.

Steven De Vleeschouwer was involved in the conceptualization of this article, the neurosurgical treatment of the second patient, the overall treatment of the 2 patients, drafting of the article and the final approval of the manuscript.

Declaration of competing interest

Nothing to declare.

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