





Case Report

Revascularization of the internal carotid artery through the hypertrophied vasa vasorum in traumatic carotid-cavernous fistula previously treated by ligation of cervical carotid arteries: A case report

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ABSTRACT

Background: Revascularization of the occluded internal carotid artery (ICA) through the vasa vasorum is exceedingly rare. Several previous studies hypothesized that the expansion of the vasa vasorum is associated with neovascularization related to the progression of atherosclerosis or plaque. The occurrence of reconstitution of the ICA through the vasa vasorum in traumatic carotid-cavernous fistula (TCCF) has rarely been reported.

Case Description: We described an extremely rare case of a 64-year-old woman who developed reconstitution of the occluded ICA through hypertrophied vasa vasorum supplying recurrent TCCF previously treated by ligation of the internal and external carotid arteries. Usual endovascular treatment may be challenging for this patient due to inaccessible route from the affected ICA through multiple small vessels. The patient was successfully treated with trapping procedure and obliteration the fistula using combined surgical and endovascular treatment.

Conclusion: We speculated that the hypertrophied vasa vasorum in TCCF may cause by a sequela of previous arterial injury, spontaneous recanalization of the occluded artery by the formation of vasa vasorum, and/or hypertrophy of the vasa vasorum due to the high flow of the fistula.

Keywords: Carotid artery ligation, Direct carotid-cavernous fistula, Trapping procedure, Traumatic carotid-cavernous fistula, Vasa vasorum

INTRODUCTION

Following complete occlusion of the internal carotid artery (ICA), recanalization through the vasa vasorum is extremely rare.^[14] Most cases of revascularization of an occluded ICA through the vasa vasorum have been caused by atherosclerotic occlusion.^[3,5,7] The proliferation of the vasa vasorum into atherosclerotic plaque may represent neovascularization and indicate that angiogenic factors may be involved.^[1] Reconstitution by a hypertrophied vasa vasorum rarely occurs in traumatic carotid-cavernous fistula (TCCF) previously treated by carotid artery ligation.^[10,16] Here, we reported a case of chronic recurrent TCCF previously treated by ligation of the internal and external carotid arteries. The occluded ICA was revascularized through hypertrophied vasa vasorum.

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CASE DESCRIPTION

A 64-year-old woman was admitted to the local hospital due to seizure with transient loss of consciousness. She complained of headache and mild cognitive impairment for the past 1 year. Cranial computed tomography (CT) scan and magnetic resonance imaging revealed markedly dilatation of the left cavernous sinus (CS), sphenoparietal sinus, and cortical veins along left cerebral hemisphere. There was some venous wall calcification [Figures 1a-g]. Her medical history showed an accident by a fall from a height of about 4 m followed by loss of consciousness and suffered subsequently from the right hemiparesis 25 years ago. She was treated by physiotherapy and admitted in the local hospital for 1 month. Two years after an accident, she developed left proptosis and underwent ligation of ipsilateral neck vessels from another tertiary hospital. Her proptosis completely resolved and she had no symptoms until the past 1 year. The patient was transferred to our institute for further investigation and proper treatment. Physical examination revealed the previous surgical scar at the left side of her neck [Figure 1h]. Visual acuity was 20/50 in the right eye and 20/100 in the left eye. She had no proptosis. On neurological examination, the patient had upper motor neuron facial weakness, nominal dysphasia, dyslexia, dyscalculia, and right-sided weakness (muscle strength 4/5) and numbness.

Cerebral angiography was performed before treatment. The left common carotid artery injection showed no existence of the external carotid artery, severe stenosis of the proximal

ICA, and long segment of multiple small vascular channels, probably representing the revascularization of the left occluded ICA through vasa vasorum, at C3 to C4 vertebral level. There was a direct high-flow fistula between the cavernous segment of the left ICA and the CS with retrograde venous drainage into markedly dilated left sphenoparietal sinus, superficial middle cerebral vein, superior sagittal sinus through the vein of Trolard, and the left transverse sinus through the vein of Labbé [Figure 2]. The fistula was also supplied by the contralateral ICA through the anterior communicating artery (ACoA) and vertebrobasilar system through posterior communicating artery (PCoA). In addition, 3D reconstruction and maximum intensity projection reformatted images of angiographic CT of the left CCA clearly illustrated multiple small serpiginous vessels, hypertrophied vasa vasorum, projecting over expected course of the ICA with antegrade filling of the distal ICA [Figure 3].

The patient underwent balloon occlusion test before treatment and could tolerate the vessel occlusion. Under general anesthesia and heparinization, transarterial detachable balloon embolization was attempted but the detachable balloon catheter could not be advanced into the fistula through ACoA from the right ICA, and PCoA from the VA. Then, the microcatheter was navigated into the fistula through ACoA from the right ICA. The largest GDC coil was attempted to place into the fistula, but the retrograde shunt into the fistula was so great that the coils mass was unable and kept migrating into the large venous pouch. Subsequently, the

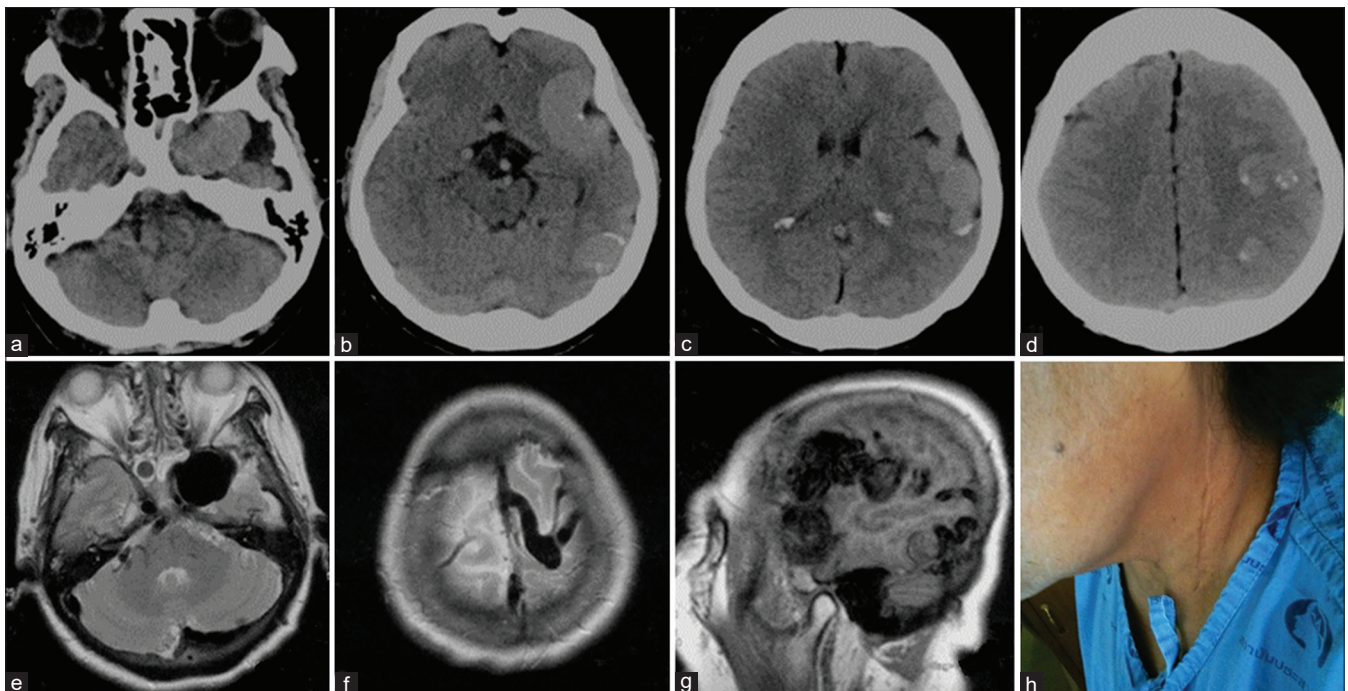


Figure 1: (a-d) Sequential axial images of non-contrast computed tomography scan, and (e and f) axial and (g) sagittal T2-weighted magnetic resonance images of the brain reveal markedly enlarged left sphenoparietal sinus and cortical veins along left cerebral hemisphere. There is some venous wall calcification. (h) Photograph of the patient's left-sided neck in oblique view shows the previous surgical scar.

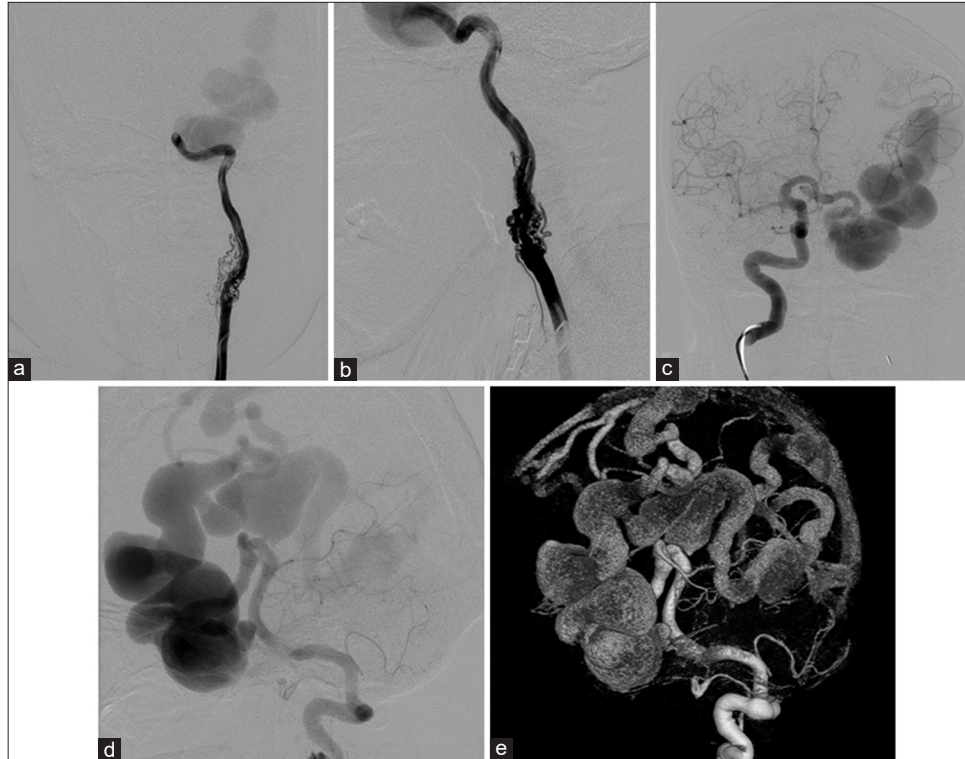


Figure 2: Cerebral angiography before treatment. (a) Anteroposterior (AP) and (b) lateral views of the left common carotid artery, (c) AP view of the right internal carotid artery (ICA), and (d) lateral views of the left vertebral artery with (e) 3D reconstruction angiographic computerized tomography reveal the left direct high-flow carotid-cavernous fistula supplied by the contralateral ICA through the anterior communicating artery, verteobasilar system through posterior communicating artery, and ipsilateral ICA through multiple small vascular channels, representing the revascularization of the left ICA through vasa vasorum, at the level of C3-C4. The venous drainage exclusively drains into markedly dilated tortuous cortical veins along left cerebral hemisphere. There is no existence of the left external carotid artery.

microcatheter was advanced further into the petrous segment of the left ICA proximal to the fistula which was occluded with Axium Detachable coils (Medtronic, Minneapolis, Minnesota, USA). Then, transarterial coil embolization was attempted at left supraclinoid ICA, but it was failed. The decision was made to proceed with surgery by clipping of the left supraclinoid ICA distal to the fistula.

On the following day, the patient underwent the left frontotemporal craniotomy. Anterior clinoidectomy was subsequently performed because it was difficult to place the clip on the ICA due to the large arterialized venous pouch. The clinoidal segment of the left ICA was clipped by the fenestrated clip encircling the left optic nerve. In addition, the left ophthalmic artery was clipped by another clip. Postoperative course of the patient was uneventful.

One month after surgery, cerebral angiography was obtained and revealed the remaining of severe stenosis of the proximal ICA with associated vasa vasorum and reduction in shunt flow and size of the dilated draining vein along the left cerebral hemisphere. In addition, the fistula received additional supply from the contralateral ICA and left VA. Prowler Plus microcatheter (Codman Neurovascular,

Raynham, MA, USA) was used over the Agility Steerable guidewire (0.016 soft, Codman Neurovascular, Raynham, MA, USA) was successfully advanced into the fistula through the small recanalized lumen of the left ICA. Embolization was performed with multiple fibered interlocking detachable coils (Interlock-35, Boston Scientific, Natick, MA). Post embolization angiography showed nearly obliteration of the fistula. Post embolization course of the patient was uneventful. Follow-up cerebral angiography, obtained 1 year after the second embolization, confirmed no recurrence of the fistula [Figure 4]. The patient had recovered completely from neurological deficits and had few episodes of seizure requiring the continuation of antiepileptic drug.

DISCUSSION

The vasa vasorum is a network of microvessels located in the adventitia of vessel wall of mid-to large-sized arteries. It supplies oxygen and necessary nutrients to the adventitia and the outer media of the arterial wall. On the other hand, the intima and inner media are nourished by direct diffusion of blood nutrients from the arterial lumen.^[5,7,12]

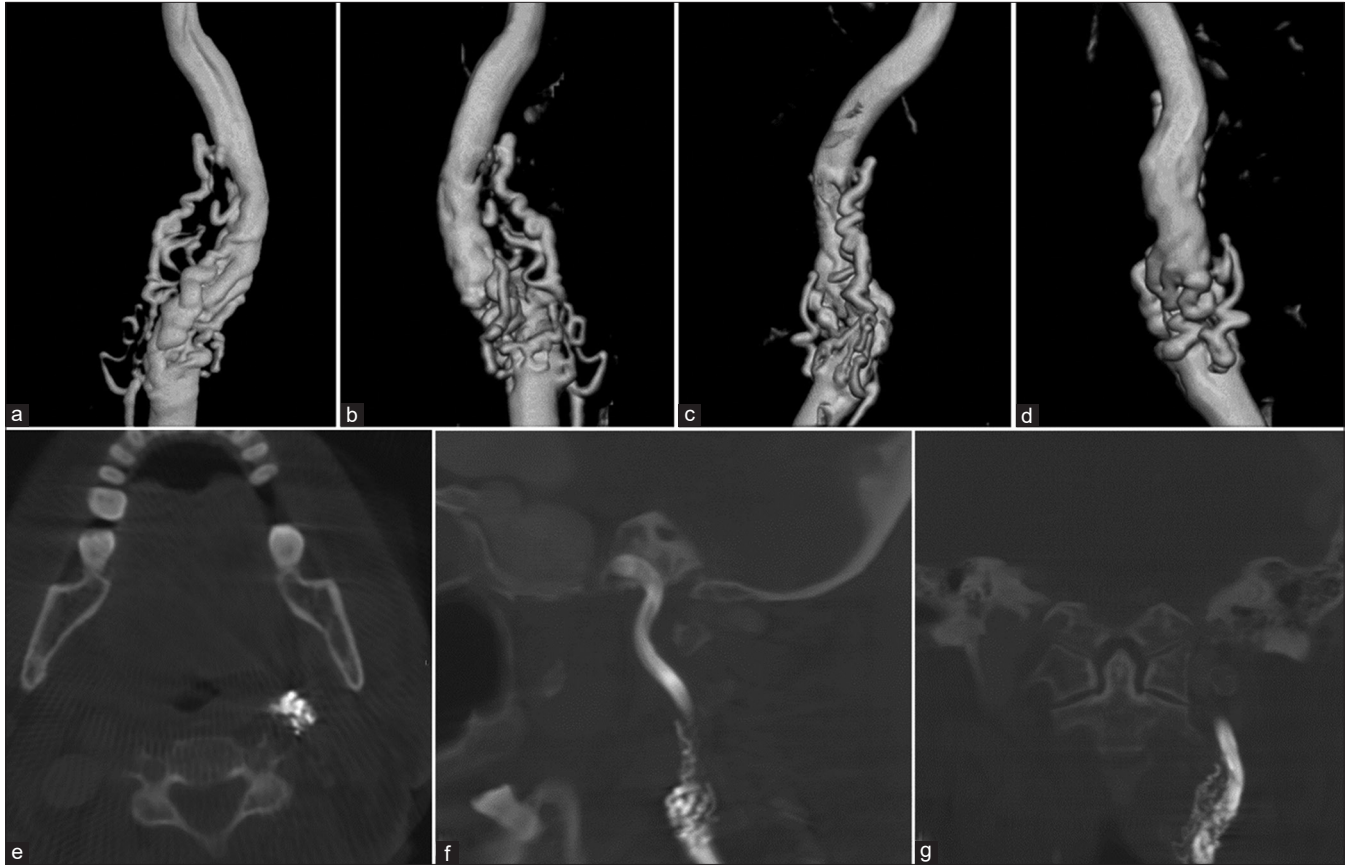


Figure 3: 3D reconstruction images in (a) anterior, (b) posterior, (c) right lateral, and (d) left lateral views, and maximum intensity projection reformatted images in (e) axial, (f) sagittal, and (g) coronal views of angiographic computerized tomography of the left common carotid artery clearly demonstrate multiple small serpiginous vessels, hypertrophied vasa vasorum, projecting over expected course of the internal carotid artery (ICA) with antegrade filling of the distal ICA.

The network of vasa vasorum surrounding atherosclerotic plaque may originate from the superior thyroid, ascending pharyngeal, common carotid, internal carotid, and external carotid arteries and fills up into the more distal ICA beyond the occlusion site.^[3] The fine vasculature through the vasa vasorum is located within and/or outside the vessel wall of the expected carotid artery.^[1] Cerebral angiography of the vasa vasorum may exhibit either single or multiple serpiginous vessels with marked tortuosity along the course of the occluded ICA.^[5,12]

Interestingly, when the ICA contains marked atherosclerotic plaque, the vasa vasorum may arise directly from lumen of the ICA distal to atherosclerotic plaque and pass inferiorly into network of the vasa vasorum.^[3] Alternatively, severe atherosclerosis may induce neovessels connecting bridging vasa vasorum to the arterial lumen.^[13] Furthermore, hypoxic condition, nutritional demand, and the recruitment of inflammatory cells may stimulate neoangiogenesis of the vasa vasorum, leading to the expansion of the existing vasa vasorum during the progression of atherosclerosis.^[17]

In non-arteriosclerotic cause, such as carotid artery dissecting aneurysm, and tumor, an occluded ICA might be recanalized through the vasa vasorum.^[1,4] Reconstitution of a previously ligated ICA in TCCF through the vasa vasorum is extremely rare. To the best of our knowledge, only three such cases have been previously illustrated from the literature review.^[10,16] O'Reilly *et al.*^[16] report a TCCF recurred 16 years after combination of clipping supraclinoid ICA, muscle embolization of the fistula, and ligations of carotid arteries. They speculated the anastomosis between the ascending pharyngeal and the cervical ICA through hypertrophied branches of the vasa vasorum of the ICA. The recurrent TCCF was closed with a balloon catheter introduced through a patent remnant of the cervical ICA. Subsequently, Halbach *et al.*^[10] demonstrated another two cases of TCCFs including one case previously treated with trapping procedure and another case with traumatic occlusion of the ICA. Both cases had vasa vasorum reconstituting ICA and were treated by direct puncture of the carotid artery above a proximal occlusion and occlusion of fistulas with coils or balloon. In addition, antegrade

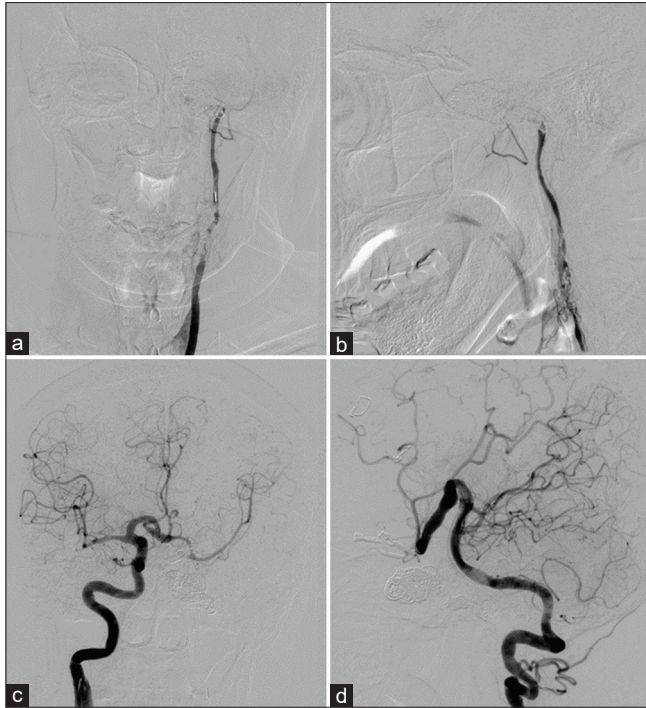


Figure 4: Cerebral angiography obtained 1 year after treatment. (a) Anteroposterior (AP) and (b) lateral views of the left common carotid artery, (c) AP view of the right internal carotid artery, and (d) lateral views of the left vertebral artery injections confirm complete obliteration of the fistula.

recanalization through the vasa vasorum may occur following parent artery occlusion by endovascular coiling or balloon.^[14,15]

Our case was the fourth case of TCCF supplied by the affected ICA through hypertrophied vasa vasorum. The patient underwent previous ligation of internal and external carotid arteries. We speculated that the fistula may remain persist or recur and the draining veins may reroute into cortical veins. Without other neurological symptoms and follow-up cerebral angiography, TCCF may sustain for more than 20 years until her seizure developed. The enormously draining veins and venous wall calcification may represent the presence of a long-standing fistula.

Following the occlusion of the ICA, revascularization of an occluded ICA may be explained by two possible mechanisms including luminal recanalization or reconstitution by a hypertrophied vasa vasorum, a source of collateral circulation.^[1,13] Marginal cerebral blood flow in the ipsilateral hemisphere may trigger the development of the vasa vasorum as potential collateral channels.^[5,15] The development of revascularization of an occluded ICA by the vasa vasorum seems to require several months to years.^[12,14] Based on by the experimental angiogenesis of arterial vasa vasorum model in rabbit by Bayer *et al.*,^[2] they found that endothelium-lined

microvessels were observed at 1 week but vessels as large as 300 μm with an organizing media were common by 3 weeks. In addition, they hypothesized that upregulation expression of the potent angiogenic factor, vascular endothelial growth factor, and its transcriptional regulator, hypoxia-inducible factor-1 α , may drive angiogenesis of vasa vasorum in their model.

In our case, we speculated that the rete mirabile-like arterial lesions of the left subpetrosal cervical ICA may cause by a combination of few factors including a sequela of previous arterial injury, spontaneous recanalization of the occluded artery by the formation of vasa vasorum, and hypertrophy of the vasa vasorum due to the high flow of the fistula.

The preferred access to the CS is certainly the affected ICA by transarterial route. Patients harboring TCCF with the previous occlusion of the affected ICA could not be successfully treated by standard endovascular techniques alone. These such cases may be cured by combining a surgical approach and endovascular technique, as shown in our case. Other options for the management of recurrent TCCF after surgical occlusion of the affected ICA have been previously reported.^[8,9,11,18] These techniques included surgical exposure or direct puncture of the CS and occlusion of the fistula with detachable balloon or coils, transarterial embolization with detachable balloon through the vertebrobasilar system through PCoA, and neck exploration for transarterial balloon embolization through the affected ICA above the occlusion point. Another option of endovascular trapping of TCCF described by Coley *et al.*,^[6] they successfully used a combination of proximal balloon occlusion and distal coiling of the parent vessel. The distal coil embolization was achieved by following retrograde catheterization of the distal parent vessel through the contralateral ICA or ipsilateral VA. They also suggested that if it had proved impossible to gain access to the distal carotid through the collateral circulation, surgical approach would have been required. In our case, we attempted to occlude the left supraclinoid ICA with coiling, but it was failed. Therefore, intracranial clipping of the left supraclinoid ICA distal to the fistula was performed.

CONCLUSION

It is well known that the proliferation or expansion of the vasa vasorum may occur during the progression of atherosclerosis. We reported the fourth case of recurrent TCCF developing the supply from the vasa vasorum after ligation of the affected ICA. We speculated that the hypertrophied vasa vasorum in TCCF may cause by a sequela of the previous arterial injury, spontaneous recanalization of the occluded artery by the formation of vasa vasorum, and/or hypertrophy of the vasa vasorum due to the high flow of the fistula. From our

review, antegrade recanalization through the vasa vasorum may occur following parent artery occlusion by endovascular coiling or balloon.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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