Vessel-preserving stent-assisted coil embolization of an extracranial internal carotid artery pseudoaneurysm that developed after tonsillectomy in a pediatric patient: initial case report

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Although there have been reports of carotid artery pseudoaneurysm formation after adenoidectomy and/or tonsillectomy secondary to iatrogenic injury, there are no case reports of successful endovascular reconstruction of the injured artery in the pediatric population. In most pediatric cases, the internal carotid artery (ICA) is sacrificed. The authors report on a 6-year-old girl who presented with odynophagia, left-sided Horner's syndrome, hematemesis, and severe anemia 6 months after a tonsillectomy. On examination she was found to have a pulsatile mass along the left posterior lateral oropharynx, and imaging demonstrated a dissection of the extracranial left ICA and an associated pseudoaneurysm. The lesion was managed endovascularly with stent-assisted coil embolization and ICA reconstruction. The child had a somewhat complicated postoperative course, requiring additional coil embolization for treatment of a minor recurrence of the pseudoaneurysm at 5 months after the initial treatment and then presenting with extrusion of a portion of the coil mass into the oropharyngeal cavity a year later. She underwent surgical removal of the extruded coils and repair of the defect and has since been free of symptoms or signs of recurrence.

The authors conclude that this strategy definitively protected the patient against an oral exsanguination or aspiration event secondary to aneurysm rupture and reduced her risk of stroke by preserving vessel patency and caliber. Moreover, they note that covered stent reconstruction surrenders endovascular access and cannot immediately provide these benefits.

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PSEUDOANEURYSMS of the carotid artery are rare medical entities occurring spontaneously in adults with an incidence of 2.5–3 per 100,000. Internal carotid artery (ICA) pseudoaneurysms are much rarer in pediatric populations and are often not spontaneous. Typical etiologies include infection, iatrogenic injury during surgery, and trauma. Multiple reports describe carotid artery pseudoaneurysm formation after adenoidectomy and/or tonsillectomy secondary to iatrogenic injury.^{9,13}

ICA pseudoaneurysms are formed from defects between the layers of the arterial wall and can extend to the

skull base and/or dissect into the oral cavity, presenting significant challenges for open surgical management. Typically, a carotid artery pseudoaneurysm presents as a pulsatile, tender mass in the neck and causes dysphagia and hoarseness. Additionally, some patients may experience life-threatening symptoms, including airway obstruction, hemorrhage, and stroke. Standard workup involves vascular imaging with ultrasound, CT angiography (CTA), MR angiography (MRA), or catheter angiography. Although patients may present in a stable condition with seemingly benign symptoms, this pathology requires urgent manage-

ABBREVIATIONS CTA = CT angiography; ICA = internal carotid artery; MRA = MR angiography; PA = posteroanterior.

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ment, as patients can decompensate rapidly due to stroke, exsanguination, or airway compromise.

Management strategies for these lesions should aim to sequester the pseudoaneurysm from the carotid circulation, given the low rate of spontaneous resolution.⁸ Prior to the advent of endovascular techniques, the standard treatment involved surgical sacrifice of the vessel with or without attempts at surgical reconstruction.⁵ However, morbidity and mortality rates for open surgical treatment of carotid artery pseudoaneurysms have been prohibitively high. Currently, endovascular treatment options include stent reconstruction with a covered stent, coil embolization of the pseudoaneurysm, or coil sacrifice of the ICA.^{6,9,11–13} The most common endovascular treatment technique used in pediatric patients is coil sacrifice of the involved segment of the ICA both distal and proximal to the pseudoaneurysm.^{1,2,4,7,10,11} Although vessel sacrifice does effectively protect the patient from pseudoaneurysm rupture, it can only be safely performed in patients with adequate collateral circulation.

In a systematic review of the literature, we discovered reports of several cases of stent-assisted coil embolization of extracranial ICA pseudoaneurysm, but we were only able to find successful treatment outcomes in the adult population.³ As of this writing, we know of no published report of a successful stent-coil reconstruction of a pediatric extracranial ICA pseudoaneurysm.

One paper described attempted direct coil embolization of a pseudoaneurysm of the cervical ICA in a 2-year-old boy who presented with recurrent epistaxis. Unfortunately, the coils subsequently migrated through the aneurysm wall into the oral cavity, and the patient ultimately required coil sacrifice of the ICA.¹⁴

In another reported case, a 5-year-old boy presented with pseudoaneurysm of his extracranial ICA after adenoidectomy and tympanostomy tube placement. He was initially treated with injection of thrombin into the pseudoaneurysm. However, he subsequently required treatment with a covered stent because of continued progression of the lesion.⁵

Covered stents permanently surrender endovascular access to the lumen of the pseudoaneurysm. If the pseudoaneurysm progresses in size or ruptures after treatment, the only salvage therapy is placement of an additional stent or vessel sacrifice. Moreover, these lesions often extend to the skull base and may require a series of overlapping stents up to the level of the petrous ICA to ensure complete endothelial apposition.

In this report, we present the first successful stent-assisted coil embolization and vascular reconstruction of an extracranial ICA pseudoaneurysm in a pediatric patient without the use of a covered stent.

Case Report

Clinical Presentation

A previously healthy 6-year-old girl presented with odynophagia, left-sided Horner's syndrome, hematemesis, and severe anemia (hemoglobin of 5) requiring transfusions 6 months after undergoing tonsillectomy in Mexico for recurrent tonsillitis. On examination, she had a pulsa-

tile mass along the left posterior lateral oropharynx adjacent to the left tonsillar pillar. She also had left-sided ptosis and miosis. She underwent MRI and MRA of the neck, which demonstrated a dissection of the extracranial left ICA and an associated pseudoaneurysm (Fig. 1). The patient was subsequently referred for endovascular management.

Endovascular Procedure

The patient was administered weight-based dosages of aspirin and clopidogrel. The right femoral artery was accessed and a femoral sheath was introduced using the modified Seldinger technique. Diagnostic angiography demonstrated a severe stenosis of the left ICA with an associated pseudoaneurysm. Also noted was collateral supply to the anterior circulation via the left anterior communicating artery and posterior communicating artery (Fig. 2).

The decision was made to attempt stent-assisted coil embolization of the pseudoaneurysm and reconstruction of the left ICA. A 5-Fr Guider Softip guiding catheter (Boston Scientific) was introduced into the sheath and advanced into the proximal left common carotid artery. Next, the pseudoaneurysm was catheterized with a PX Slim microcatheter (Penumbra) and a Synchro-2 micro-wire (Stryker).

After establishing our microcatheter position within the pseudoaneurysm, we deployed 11 individual Penumbra Coil 400 detachable 0.21 coils. Follow-up runs confirmed adequate packing density within the aneurysm. Next, we crossed the dissection flap with our microcatheter system and deployed two 4.5 × 37 mm nitinol Enterprise stents (Codman & Shurtleff and DePuy Mitek) along the length of the defect. Follow-up diagnostic angiography demonstrated restoration of luminal caliber, complete angio-

FIG. 1. Left: Fat-saturated contrast-enhanced axial T1-weighted MR image demonstrating a homogeneously enhancing mass along the left posterolateral pharynx. Right: 3D maximum intensity projection (MIP) demonstrating a flame-shaped narrowing of the left ICA with a large 5-cm pseudoaneurysm.

FIG. 2. Diagnostic cerebral angiography. **A and B:** Angiographic images demonstrating a large contribution to the left anterior circulation by the anterior communicating artery and posterior communicating artery, respectively. **C:** Oblique posteroanterior (PA) angiogram demonstrating the dissection with a characteristic flame-shaped narrowing and a large 5-cm pseudoaneurysm. **D:** 3D reconstruction from a 3D rotational angiogram demonstrating the pseudoaneurysm and dissection. Figure is available in color online only.

graphic occlusion of the pseudoaneurysm, and improved cerebral perfusion to the anterior intracranial circulation via the left ICA (Fig. 3).

Postoperative Course

At her 3-month follow-up visit, the patient's odynophagia, hematemesis, Horner's syndrome, and anemia had all resolved. Additionally, the posterior pharyngeal mass was nonpulsatile and reduced in size. The patient was normotensive and neurologically intact. This was in contradistinction to the patient's initial systemic hypertension on presentation to augment her collateral circulation in the face of poor left ICA flow.

Follow-up catheter angiography at 5 months after initial treatment demonstrated a 6-mm residual or recurrence within the left ICA pseudoaneurysm (Fig. 4). The patient underwent repeat endovascular treatment with additional coil embolization, which obliterated the recurrence (Fig. 5).

One year after treatment of the pseudoaneurysm recurrence, the patient developed throat pain and a sensation of a foreign body in her pharynx. She was otherwise at her clinical baseline. Upon examination, a portion of the coil

FIG. 3. Posttreatment angiography. **Left:** Oblique PA digital subtraction angiogram of the cervical left ICA demonstrating complete occlusion of the pseudoaneurysm and restoration of the luminal caliber of the left ICA. **Right:** PA digital subtraction angiogram of the anterior circulation performed by injection of the left ICA, demonstrating improved perfusion of the cerebral hemisphere via the left ICA as evidenced by complete opacification of both the middle cerebral artery and A₁ segment of the anterior cerebral artery with no evidence of washout by collaterals.

mass was noted to have extruded into the oropharyngeal cavity. CTA demonstrated no filling of the pseudoaneurysm and appropriate contrast filling along the course of the cervical ICA through the stent construct. Visual in-

FIG. 4. PA digital subtraction angiogram performed by injection of the left ICA at 5 months following initial repair, demonstrating recurrence of the left ICA pseudoaneurysm.

FIG. 5. PA digital subtraction angiogram performed by injection of the left ICA after repeat coil embolization at 5 months, demonstrating complete obliteration of the pseudoaneurysm.

spection and radiography demonstrated extrusion of a portion of the coils into the pharyngeal space (Fig. 6). The patient was referred to the Division of Otolaryngology and underwent resection of the extruded coils. The remainder of the coils were left in place, and the defect was repaired with a mucosal flap (Fig. 7).

At her most recent follow-up examination, 18 months after the endovascular reconstruction, the patient had experienced no further extrusion of coils or symptom recurrence (Fig. 8).

Conclusions

This case demonstrates that endovascular treatment can exclude an iatrogenic ICA pseudoaneurysm from circulation with coil embolization while preserving vessel patency using stent reconstruction. Iatrogenic pseudoaneurysms of the carotid artery carry significant risk for hemorrhage and stroke. Although tonsillectomy complications are rare, with reported mortality rates ranging from 1 in 1000 to 1 in 170,000 procedures, 30% of these deaths are secondary to vascular injury/hemorrhage of the carotid artery.⁹ Carotid artery pseudoaneurysm formation after tonsillectomy usually occurs secondary to iatrogenic

FIG. 6. A: Lateral radiograph of the soft tissues of the neck demonstrating extrusion of the coils into the pharynx. B: Endoscopic view of the oropharynx demonstrating the extruded coil mass protruding through the mucosa. C: Photograph of the coils removed from the mucosal defect. Figure is available in color online only.

injury to the vessel. Any disruption of the pharyngeal constrictor muscles during surgery risks injury to the carotid artery, especially if there is any tortuosity or looping of the vessel against the muscular wall. Inadvertent partial disruption of the vessel can lead to weakening and result in pseudoaneurysm formation, which may present in a delayed fashion.^{9,13} Although rare, these lesions require prompt diagnosis and treatment. Deconstructive strategies with open surgery or coil sacrifice may be associated with increased acute and long-term risk for stroke and may not be feasible in patients without adequate collateral circulation. Stent-coil reconstruction, in contrast to covered stent placement, preserves the potential for repeated endovascular access to the pseudoaneurysm. This is particularly critical if the aneurysm recurs, as was demonstrated in this case. Additionally, primary attempts at ICA preservation do not preclude deconstructive strategies as a salvage option.

FIG. 7. Left: Lateral radiograph of the soft tissues of the neck performed after the coils were removed, demonstrating the residual coil mass. Right: Photograph of the posterior pharynx after repair of the mucosal defect, revealing no residual extruded coils. Figure is available in color online only.

FIG. 8. Axial CT angiogram obtained 2 months after the final mucosal repair demonstrating a patent left ICA.

To our knowledge, this case report represents the first successful stent-coil reconstruction of an extracranial carotid artery pseudoaneurysm in a pediatric patient and demonstrates proof of principle. It further demonstrates the safe surgical management of delayed coil extrusion after endothelialization of the construct.

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Disclosures

Dr. Khalessi reports having consultant relationships with Medtronic, Stryker, and MicroVention.

Author Contributions

Acquisition of data: Goel, Pannell, Nation. Analysis and interpretation of data: Khalessi, Steinberg, Cheung. Drafting the article: Khalessi, Steinberg, Cheung, Pannell. Critically revising the article: Khalessi, Steinberg, Cheung, Pannell. Reviewed submitted version of manuscript: Khalessi, Steinberg, Pannell, Nation. Approved the final version of the manuscript on behalf of all authors: Khalessi.

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