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CASE REPORT

Covered Stent Treatment of an Extracranial Internal Carotid Artery Pseudoaneurysm in a Three Year Old Child with 12 Years of Follow Up: A Case Report

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Introduction: Extracranial internal carotid artery (ICA) pseudoaneurysms in children, although uncommon, are life threatening. Covered stents are a good alternative treatment, as they avoid the risk of open surgery and preserve the ICA. Until recently, long term outcomes were unknown.
Report: A three year old boy was hospitalised with an enlarged swelling on the left side of his neck and severe respiratory distress. He had been treated a month prior for a left deep cervical abscess, with bacteriological culture positive for *Staphylococcus aureus*. Computed tomography angiography (CTA) revealed a large pseudoaneurysm originating from the left ICA, approximately 2 cm above the bifurcation. A balloon expanded covered stent (Jostent Graftmaster; Abbott Vascular, Redwood City, CA, USA) was deployed via a left femoral approach, after selective angiography, to seal the carotid rupture without incident. Control angiography revealed immediate exclusion of the pseudoaneurysm and patent ICA. The bacteriological culture of the residual haematoma was negative. The child was discharged with full recovery and without neurological sequelae, under platelet anti-aggregation. He has been followed up and has remained asymptomatic for 12 years, with CTA confirmed ICA patency, without deformation or evidence of significant restenosis.
Discussion: This is the first report of the long term outcome of a covered stent in a child treated at three years of age, with a 12 year follow up. The good performance of the covered stent in this case reinforces its adoption as a first line option in the treatment of extracranial ICA pseudoaneurysms in children.

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INTRODUCTION

Pseudoaneurysms of the extracranial internal carotid artery (ICA) are rare in children. They are usually caused by trauma or infections in the deep cervical parapharyngeal compartments.[1] Rapid expansion with compression of the upper airways or extensive bleeding requires emergency management. The main treatment is carotid artery ligation, but this is associated with a neurological risk. Endovascular procedures, such as embolisation with coils or occlusive balloons, are low risk options, but they are associated with carotid artery sacrifice. Covered stents allow rupture coverage, while preserving the affected artery. There are a few reports of ICA pseudoaneurysms in children treated with covered stents. These showed good initial results, but long term outcomes were not reported.

CASE REPORT

In August 2008, a three year old boy was admitted to a private clinic in Concepción City, Chile, with an enlarged swelling on the left side of the neck and severe respiratory distress. He had been hospitalised a month earlier with a left deep cervical abscess that was drained via a cervicotomy. Bacteriological culture was positive for *Staphylococcus aureus*. After three weeks of targeted antibiotic treatment, the patient was discharged in a good condition. One week later, he presented to the emergency room. Emergency computed tomography angiography (CTA) was performed before endotracheal intubation and mechanical ventilatory assistance. The CTA showed a large pseudoaneurysm originating from the left ICA, approximately 2 cm above the bifurcation, that had extended to the entire retropharyngeal

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space with severe compression of the upper airway (Fig. 1). An endovascular approach was chosen over open surgery by a multidisciplinary team because the boy had a hostile neck. Using a left femoral approach, a 6 F guide sheath was placed within the left common carotid artery. A 5 F vertebral catheter was telescoped and positioned at the origin of the ICA. Selective angiography showed a jet of the ICA approximately 2 cm above the bifurcation. A 0.014 guidewire was positioned in the distal ICA. Under systemic heparinisation (80 UI/kg), a 4 × 19 mm Jostent Graftmaster (Abbott Vascular, Redwood City, CA, USA) balloon expanded stent was deployed to cover the carotid rupture without incident. Completion angiography showed immediate exclusion of the pseudoaneurysm and ICA patency (Fig. 2). The retropharyngeal residual haematoma was secondarily evacuated through a right cervicotomy. The child was extubated with full recovery without neurological sequelae. The bacteriological culture of the residual haematoma was negative. Antibiotic use was limited to the peri-operative period. One week later, the child was discharged home under platelet aggregation, using aspirin at a dose of 3 mg/kg/day. The patient has remained asymptomatic without complications until today. CTA performed at eight months and 12 years after surgery showed ICA patency, with correct placement of the covered stent without deformation or evidence of significant restenosis (Fig. 3).

DISCUSSION

Pseudoaneurysms of the extracranial ICA in children are rare. They are usually related to the proximity of the ICA to parapharyngeal abscesses.[1] They can also result from inadvertent trauma during tonsillectomy. In this case, a deep cervical abscess was drained through a left cervicotomy. Therefore, concomitant trauma may have contributed to the development of the carotid pseudoaneurysm. The frequent presentations include pulsation, increasing cervical swelling, Horner syndrome, or compression of the lower cranial nerves. In this child, the pseudoaneurysm expanded rapidly, and was associated with compression of the upper airway, which led to emergency management. This unusual presentation has been reported previously.[2]

Open surgery in a hostile neck is challenging for surgeons owing to the risk of iatrogenic injury. The previous mainstream treatment, which is proximal ligation of the ICA, has been abandoned because of the associated morbidity and mortality, as well as the complex arterial reconstructions.[3,4] Endovascular techniques, such as embolisation with coils or the use of occlusive balloons, reduce the risks of open surgery, but they result in the sacrifice of the artery.[5] Currently, the goal of treatment of carotid pseudoaneurysm is to achieve immediate exclusion from the circulation, while preserving ICA patency. Unlike uncovered stents, covered stents achieve this goal. Several materials have been used for covered stents, including synthetic polymers (polytetrafluoroethylene [PTFE] and Dacron) and biological materials (autologous vein, heterologous pericardium, endothelial cells, and fibrin).[6] Covered stents for small diameter vessels have been developed for coronary applications. This is the case for the Jostent Graftmaster coronary stent, which consists of a PTFE sandwich between two stainless steel stents. In a three year old child, the distal ICA reaches a diameter of 4 mm, and the Jostent Graftmaster stent adjusts to this measurement. As there was a punctiform rupture in the post-bulbar segment of the ICA, the precision of a short length balloon expanded stent seemed appropriate. Other types of stents, such as flow diverting stents (FDS) and coronary covered self expanding stents, were not available in Chile at that time. Otherwise, FDS does not ensure the immediate exclusion of the pseudoaneurysm, making the subsequent use of antiplatelet drugs difficult.[7]

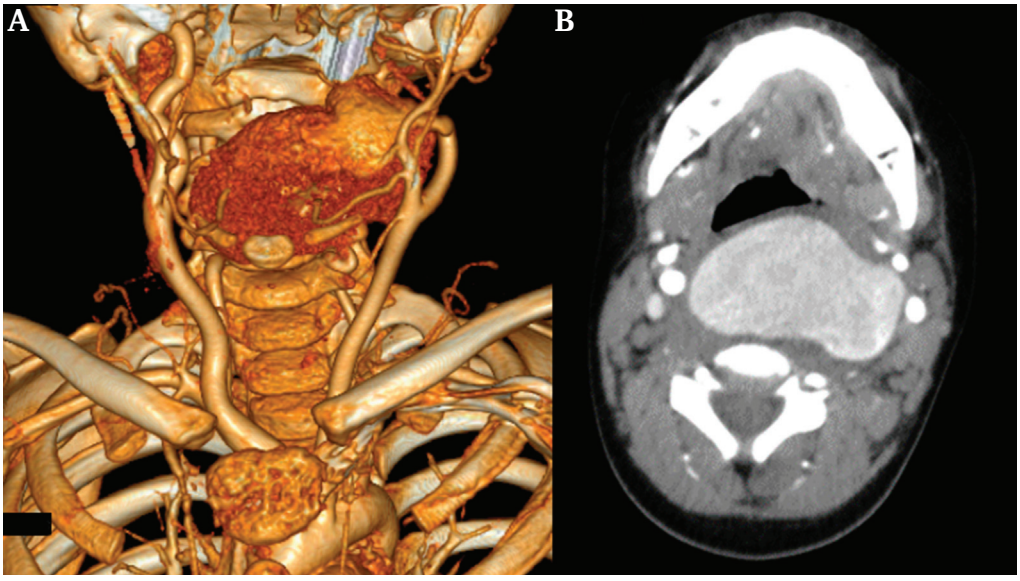

Figure 1. (A) Giant pseudoaneurysm arising from the extracranial left internal carotid artery. (B) Severe compression of the upper airway.

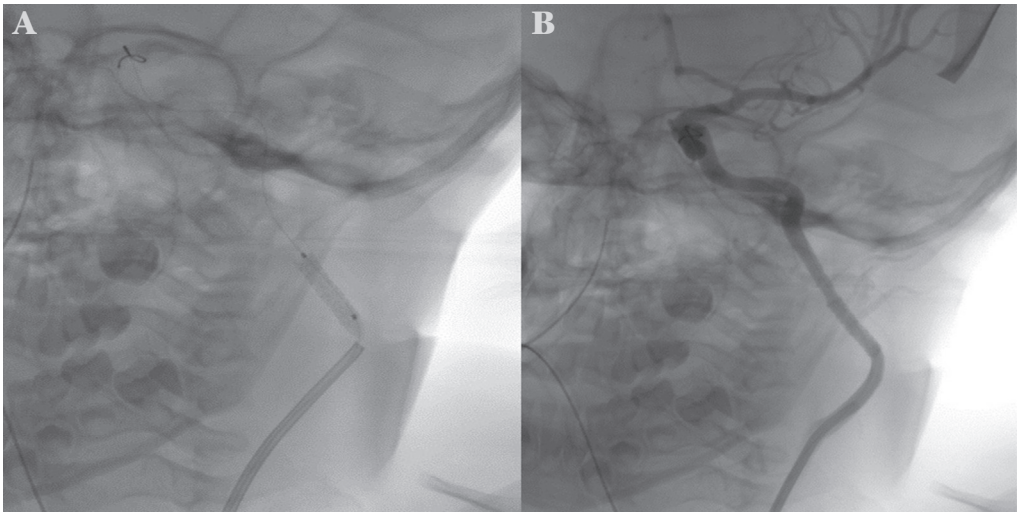

Figure 2. (A) Covered stent deployed at the site of internal carotid artery (ICA) rupture. (B) Angiographic control revealing immediate sealing and patency of the ICA.

There are a few reports on the treatment of pseudoaneurysms in children using covered stents.[8] These show good initial results, but long term results have not been reported.[9] The immediate complications of stenting are thrombosis, micro-embolisation, and carotid artery dissection. In stents covered with PTFE, thrombogenicity is added as a result of the hydrophobic properties of the surface that facilitate platelet adhesion. In the medium term, this thrombogenicity would be increased by the delay in the endothelisation of PTFE and the slowing down of the flow in a small diameter stent. Over time, localised restenosis is observed at the ends of the stent and not within the stent, as the covering material protects against intimal proliferation. The Jostent Graftmaster stent requires high pressure for post-expansion owing to its rigidity and lack of compliance. This may be difficult to use in growing children, who may require secondary open surgery during adulthood if the stent is not iteratively dilated. Based on the target height

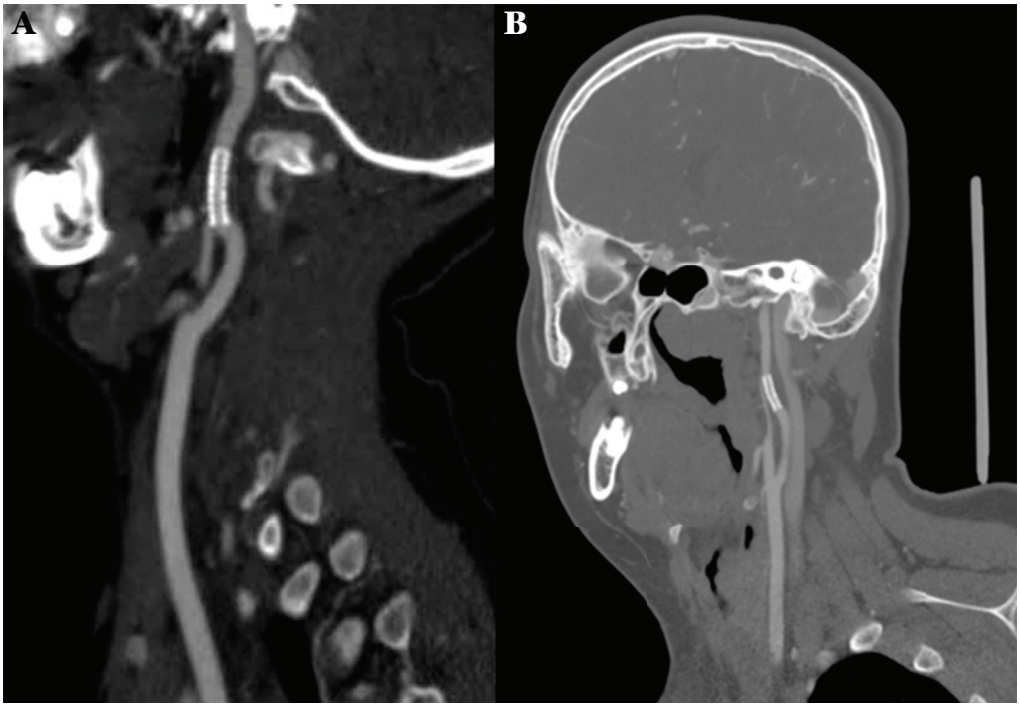

Figure 3. (A) Eight year and (B) 12 year follow up computed tomography angiograms confirm covered stent patency without significant restenosis.

according to the mid-parental height, this young man had almost reached his final height, and the diameter of the distal ICA could be stable, close to five mm; no invasive revision was warranted at that time. Follow up CTA in this patient showed a patent stent with a slight difference in diameter without significant restenosis.

Platelet aggregation with aspirin at the standard prophylactic dose has been shown to be safe. Aspirin is the most commonly used platelet anti-aggregant in children with cardiovascular disease, although the use of clopidogrel is increasing.[10] In this case, platelet anti-aggregation was started early with aspirin at a dose of 3 mg/kg/day for up to 100 mg/day, a dose that has been maintained until today. Owing to the satisfactory long term outcomes of this patient, it was not considered necessary to add another platelet anti-aggregant or oral anticoagulant. After the satisfactory initial outcomes and the 12 year follow up, the adolescent remains asymptomatic and without complications.

Summary

Pseudoaneurysms of the extracranial ICA are rare in children. They can present with abrupt compression of the upper airways and require emergency treatment. The use of covered stents is safe and effective for preventing morbidity and mortality associated with open surgery. The long term outcomes of the covered stent in this report are the first to be reported for a child treated at three years of age, with a 12 year follow up. The good performance of the covered stent in this case may facilitate its adoption as the first option in the treatment of extracranial ICA pseudoaneurysms in children.

CONFLICT OF INTEREST

None.

FUNDING

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