Internal Carotid Artery Pseudoaneurysm and Ischemic Stroke Secondary to Retropharyngeal and Parapharyngeal Abscess: A Case Report and Review of the Literature

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Abstract

Internal carotid artery pseudoaneurysm is an uncommon complication of retropharyngeal and parapharyngeal abscess in children. Treatment of the pseudoaneurysm has evolved in recent years from surgical ligation to endovascular techniques. Neurologic sequelae most commonly consist of Horner's syndrome with cerebral ischemia being uncommon. The clinical course of a 2-year-old boy with retropharyngeal abscess complicated by internal carotid artery pseudoaneurysm, is described and the literature is reviewed. A conventional angiogram confirmed the presence of a large pseudoaneurysm with no anterograde flow distal to the pseudoaneurysm and substantial collateral flow across the circle of Willis, with filling of the left anterior and middle cerebral arteries via the anterior and posterior communicating arteries. Endovascular occlusion resulted in nonfilling of the left internal carotid artery, pseudoaneurysm, and left internal jugular vein at the base of the skull. Following the procedure, the patient developed transient mild right hemiparesis associated with frontal lobe ischemia.

Keywords

acute ischemic stroke, endovascular coiling, internal carotid artery, pseudoaneurysm, retropharyngeal abscess

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Internal carotid artery pseudoaneurysm is an uncommon complication of retropharyngeal and parapharyngeal abscess in children. Unlike a true aneurysm, which is a dilation of the artery involving all layers of the vessel wall, the walls of a pseudoaneurysm consist of adventitia, periarterial fibrous tissue, and hematoma. The encapsulated hematoma communicates with the lumen of the vessel as a result of damage to one or more layers of the arterial wall. Ongoing enlargement of the pseudoaneurysm results in a pulsatile hematoma, which may result in clot propagation and embolization or hemorrhage. Treatment of the abscess may include antibiotics and drainage of the abscess; however, specific treatment of the pseudoaneurysm is also required in order to prevent hemorrhage and other sequelae. Treatment of the pseudoaneurysm has evolved in recent years from surgical ligation to endovascular techniques. Neurologic sequelae most commonly consist of Horner's syndrome, with cerebral ischemia being uncommon.

The clinical course of a 2-year-old boy internal carotid artery pseudoaneurysm, associated with retropharyngeal abscess, complicated by ischemic cerebral infarction is described and the previously reported cases are reviewed.

Case

A 2-year-old boy with a history of tonsilloadenoidectomy and bilateral tympanostomy, several months prior, developed 2 days of nausea and vomiting followed by sore throat with neck stiffness, decreased oral intake, irritability, and increased drooling. His parents noted decreased hand coordination. He presented to medical attention 3 days after the onset of symptoms. At presentation, he was found to have a temperature of 38.0° C, leukocytosis of $24~000 \times 10^{9}$ /L (normal 5-12), and a C-reactive protein of 138~mg/L (normal <8). His examination was significant for neck discomfort with movement and

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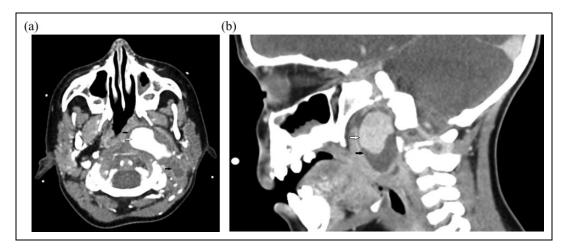


Figure 1. Axial (A) and sagittal (B) images of computed tomography with contrast revealing an internal carotid artery pseudoaneurysm (periarterial encapsulated hematoma) (white arrow) within the parapharyngeal and retropharyngeal abscess (black arrows) in a 2-year-old boy.

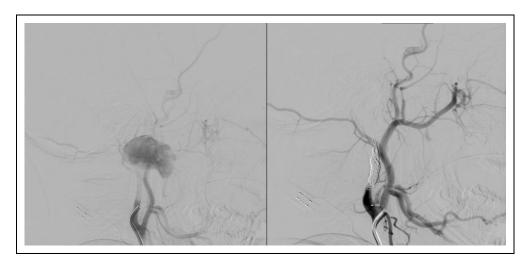


Figure 2. Conventional angiography images demonstrate a large left internal carotid artery pseudoaneurysm measuring 1.3×3.6 cm, with no anterograde flow distal to the pseudoaneurysm, pre (left) and post (right) internal carotid artery endovascular coiling.

pharyngeal erythema. The neurologic examination in the emergency room was documented as normal with no focal deficits. Coordination and hand strength were specifically documented as being normal. Broad-spectrum antimicrobials were started because of concern for meningitis or sepsis. A lumbar puncture obtained before administration of antimicrobials was negative and revealed colorless fluid with 0 nucleated cells/UL, 7 red blood cells/UL, protein of 11 mg/dL, glucose 76 mg/dL. Blood, urine, and cerebrospinal fluid cultures were negative. Both influenza swab and rapid streptococcal antigen testing was negative. However, throat cultures grew group A streptococcus. He was observed for 2 days and then discharged on oral cefdinir with a diagnosis of streptococcal pharyngitis. His C-reactive protein at discharge was 174 mg/L, after a peak value of 186 mg/L.

Following discharge, the patient's neck pain persisted and gradually worsened. He developed "muffled and gargled" speech and an antalgic gait. On reassessment, cervical adenopathy was noted on examination. The documented

examination did not specifically mention the speech or gait but noted "no focal deficits." Additional testing, including a Monospot and Epstein-Barr virus (EBV) titer, was negative. Repeat C-reactive protein and white blood cell count had normalized (1.10 mg/L and 9200 \times 10 9 /L, respectively). Observation was recommended.

He was reevaluated again 3 days later after developing reduced range of neck motion as a result of swelling at the base of his left jaw and anterior neck. He was sent to the nearest emergency department because of these clinical findings, where a computed tomography scan with contrast demonstrated a large retropharyngeal abscess and left internal carotid artery pseudoaneurysm (Figure 1).

The patient was emergently transferred to our institution. Upon arrival, a 4-vessel angiogram was performed. The left internal carotid artery injection confirmed the presence of a large pseudoaneurysm measuring $1.3~\rm cm \times 3.6$ with no anterograde flow distal to the pseudoaneurysm (Figure 2A). Injection of the right internal carotid artery and bilateral vertebral

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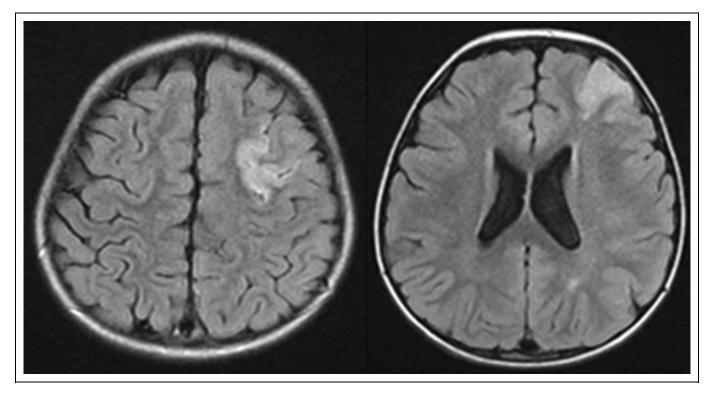


Figure 3. Magnetic resonance imaging (MRI) brain axial fluid-attenuated inversion recovery images demonstrate ischemic infarction in the left frontal and parietal regions.

arteries demonstrated substantial collateral flow across the circle of Willis, with filling of the left anterior and middle cerebral arteries via the anterior and posterior communicating arteries. Presumably as a result of the left internal carotid artery occlusion, the patient had established collateral flow, with the anterior cerebral circulation being provided by the right internal carotid artery and vertebral as well as intracranial collaterals including the artery of rotundum originating from the left external carotid artery.

In order to treat the pseudoaneurysm, and prevent clot propagation and distal embolization, the internal carotid artery was surgically occluded. This was done by placing platinum coils within the proximal left internal carotid artery between the carotid bifurcation and the pseudoaneurysm. Sacrificing the internal carotid artery was an option because the vessel was functionally occluded, and collateral flow had been established. An angiogram status post endovascular surgical occlusion demonstrated non-filling of the left internal carotid artery, pseudoaneurysm and left internal jugular vein at the base of the skull (Figure 2B). The retropharyngeal and parapharyngeal abscess remained unchanged in size. The patient's mean arterial blood pressure was measured by arterial catheter as 86-62 mmHg during the procedure.

Following the procedure, the patient was noted to have weakness of the right arm, though he appeared to move both legs without difficulty. Magnetic resonance imaging (MRI) demonstrated restricted diffusion in the left frontal lobe consistent with an acute/subacute ischemic infarct. There was also a small area of T2/fluid-attenuated inversion recovery

hyperintensity in the white matter of the left parietal lobe, which was also consistent with ischemic stroke (Figure 3). The location of the strokes was most suggestive of a watershed distribution infarction. The lesions may have occurred during the procedure; however, his parents did note clumsiness prior to his evaluation at our institution, which may have been secondary to a mild right upper extremity monoplegia. Unfortunately, his right arm was immobilized for the intravenous line prior to the procedure and therefore the exact timing of the right arm weakness cannot be definitively determined.

Broad-spectrum intravenous antibiotics were continued throughout hospitalization. On the seventh day of hospitalization (28 days after onset of symptoms), the patient underwent ultrasound-guided aspiration of the retropharyngeal abscess. The aspirated fluid was sterile. His C-reactive protein and white blood cell count were normal at time of hospital discharge, and his neurologic examination was improved.

Discussion

Infectious carotid pseudoaneurysm is an uncommon complication of deep neck infections. Although much more common in the preantibiotic era,¹ they are rarely encountered today. In adults, this may be a complication of dental infections or septic emboli from intravenous drug use, while in children it is more commonly seen in the setting of pharyngeal and retropharyngeal infections. Osler² initially felt that this was secondary to direct contiguous spread of the infection to the carotid artery sheath resulting in arteritis and erosion of all 3 layers of the

Table 1. All Cases of Extracranial Infectious Carotid Pseudoaneurysm in Pediatrics (<18 Years Old).^a

Author, year	Age/sex	Symptoms	Medical management of abscess	Treatment of pseudoaneurysm	Neurologic complication
Alexander et al, 1968 ⁵	a) 3 y/ F b) 4 y/ Μ	 a) Pharyngomaxillary space abscess, peritonsillar abscess b) Neck swelling, sore throat, parotiditis, hematemesis 	 a) Drained: Yes Antibiotics: tetracycline for presumed mumps. b) Drained: yes. Antibiotics: achromycin and 2 injections of penicillin for presumed parotiditis 	a) Surgical common carotid artery ligation b) Surgical internal carotid artery exploration and ligation	None
Garino and Ryan 1987 ³	15 y/M	Peritonsillar abscess, upper airway	Unknown	Surgical internal carotid artery ligation	None
Tovi et al, 1987 ⁶	14 y/F	oreeung Epistaxis, pharyngeal bleed	None?	Surgical internal carotid artery ligation with endovascular coil occlusion	None
Stevens, 1990 ⁷	15 y/M	Epistaxis, Group A streptococcal deep neck infection	Keflex, cefazolin, penicillin, cloxacillin	Endovascular internal carotid artery coil occlusion	CN X, XII palsy
Gonda et al, 1990 ⁸	I8 mo/M	Neck mass, respiratory distress, strep throat	Antibiotics : oral penicillin failed. Penicillin and clindamycin IV $ imes$ 10 d, continued oral 3 wk	Surgical external carotid artery ligation and resection	Horner syndrome
Watson et al, 1991	7 y/M	Epistaxis, tonsillitis	Antibiotics: IV benzylpenicillin, failed oral antibiotics. Drainage: no	Surgical internal carotid artery ligation	None
Wells and Sty, 199110	4 y/F	Dysphagia, cervical lymphadenitis	No drainage, no antibiotics mentioned	Surgical exploration	٧Z
Lueg et al, 1995 ¹¹ Karas et al, 1997 ¹²	3 y/F 6 y/M	Dysphagia, bloody sputum Posttonsillectomy hematemesis, 5-cm pulsatile mass	Cloxacillin preoperatively None	Surgical common carotid artery ligation Surgical external carotid artery ligation	None None
Willemsen et al, 1997 ¹³	Μ/λ 6	ALL, neck mass, angioinvasive aspergillus infection	Drained, plus broad-spectrum antibiotics, then antifungal	Surgical common carotid artery ligation	Right vocal cord paralysis- resolved
Reisner et al, 1999 ¹⁴	16 y/F	Pulsatile neck mass, hemoptysis, deep neck space infection, pharyngitis, Stathblococus	Parapharyngeal abscess drained. Antibiotic treatment for 6 wk	Endovascular internal carotid artery coil occlusion	Horner syndrome- resolving
Ferguson et al, 2002 ¹⁵	8 y/M	parapharyngeal	IV antibiotics	Surgical external carotid artery ligation and endovascular internal carotid artery balloon occlusion	None
Glaiberman et al, 2003 ^{l6}	M/om II	Neck swelling, necrosis of third distal phalanx- strep A parapharyngeal abscess	IV antibiotics	Endovascular carotid artery balloon occlusion	None
Tannuri et al, 2003 ¹⁷	I0 mo/NA	Left neck swelling: Retropharyngeal abscess I mo before, bruit	Broad-spectrum antibiotics for 10 d	Endovascular internal carotid artery coil occlusion and surgical drainage	None
Singh et al, 2003 ¹⁸	4 y/M	Trismus, dysphagia, face and neck swelling, parapharyngeal infection	Drained	Surgical common carotid artery ligation	Facial nerve palsy, resolved
Brochu et al, 2004 ¹⁹	6 y/F	Neck pains, dysphagia, dysphonia,	IV antibiotics for 6 wk and tonsillectomy	Endovascular internal carotid artery coil	Horner syndrome, vocal
Gralla et al, 2004 ²⁰	Μ/γ	parapuar yrışcar absecss Epistaxis, pharyngeal hemorrhage	Not stated	Endovascular internal carotid artery stent exclusion	None
Mordekar et al, 2005 ²¹	8 y/F	Odynophagia, trismus, peritonsillar swelling	IV antibiotics for 4 wk	Endovascular internal carotid artery coil occlusion	None

rable I. (continued)					
Author, year	Age/sex	Symptoms	Medical management of abscess	Treatment of pseudoaneurysm	Neurologic complication
Semple et al, 2005 ²²	I5 y/M	Pulsatile neck mass, deep neck infection	Broad-spectrum antibiotics and amphotericin B	Endovascular internal carotid artery coil occlusion	None
Hoff et al, 2005 ²³	7 y/M	Posttonsillectomy hemorrhage 2 wk	Not stated	Endovascular internal carotid artery coil	None
Pearson and Choi, $2005^{24} + 4 \text{ y/F}$	4 y/F	Dysphonia, parapharyngeal infection	IV antibiotics for 3 wk Hematoma aspirated	occusion fortower by surgical igation Endovascular internal carotid artery coil occlusion	Deficits of IX-XII cranial nerves, vocal cord paralysis, Horner
DeFatta et al, 2005 ²⁵	8 y/F	Throat pain, peritonsillar abscess, neck IV and oral antibiotics swelling and pain	IV and oral antibiotics	Endovascular internal carotid artery coil	syndrome Horner syndrome, resolved
Meher et al, 2006 ²⁶	8 mo/M	Swelling and palli Neck swelling, parapharyngeal and retropharyngeal space infection	Drained	Surgical internal carotid artery ligation	None
Beningfield et al, 2006 ²⁷	I0 mo /NA	Δ	Drained	Endovascular internal carotid artery coil	None
Pourhassan et al, 2007 ²⁸	17 y/M	retro- and parapnaryngear abscess Tonsillitis s/p tonsillectomy 10 d prior,	Oral antibiotics Not stated	occiusion Surgical external carotid artery ligation and resertion	Transient dysphagia
Watanabe et al, 2009 ²⁹	15 y/NA	suprifationari swelling, uyspitagia s/p BMT allogenic, otalgia, aspergillus	Drainage of middle ear and decompressed CN VII IV antibiotics, amphotericin B, and micafungin	and resection Endovascular internal carotid artery coil occlusion	Hemorrhagic cerebral infarctions, right hemiplegia- resolved, persistent left facial nerve palsy
Makeieff et al, 2010 ³⁰	16 y/F	Tonsillitis, retropharyngeal abscess,	Antibiotics	Endovascular internal carotid artery saccular coiling occlusion with covered	(preprocedure) None
da Silva et al, 2011 ³¹	3 y/M	Epistaxis, deep neck space infection,	Drained	Stent Endovascular internal carotid artery	Mild left hemiparesis (post
Sankararaman et al, 2012 ³² 6 mo/F	6 mo/F	peritorisillar abscess, cardiac arrest Fever, pharyngitis, dysphagia, retropharyngeal abscess, neck mass	Airtiblotics 101 4 Wk Drained IV antibiotics for 6 wk	bandon occiusion Conservative No surgical or endovascular intervention	procedure), resolved None
Requejo et al, 2013³³	4 y/M	Strep throat, pharyngitis, pulsatile neck mass	Not stated	Endovascular internal carotid artery stent occlusion	None
Current Case	2 y/M	Sore throat with neck stiffness, decreased oral intake, irritability, and increased drooling	Drained IV antibiotics	Endovascular internal carotid artery coil occlusion	Ischemic infarction with mild right hemiparesis resolved
		D			

Abbreviations: ALL, acute lymphoblastic leukemia; BMT, bone marrow transplant; CN, cranial nerve; IV, intravenous; NA, not available; s/p, Status post; strep, streptococcal.
^aExcluded cases: Age >18 y, true internal carotid artery aneurysms, intracranial aneurysms, and traumatic or noninfectious pseudoaneurysms.

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vessel wall. The extravasated blood is then contained within the surrounding fibrous tissues, creating a pseudoaneurysm or infectious aneurysm. The very limited descriptions of the pathology of infectious pseudoaneurysms throughout the body suggest an involvement of inflammation or direct injury to one or more layers of the vessel wall in the pathophysiology of pseudoaneurysm but have not definitively shown the presence of direct spread of the infection to the carotid artery sheath.

Missed or delayed diagnosis of this entity can result in devastating complications including septic cerebral emboli, carotid rupture and death. If untreated, infectious pseudoaneurysms have mortality rates of up to 77%. Treatment involves both antibiotic therapy for 4 to 6 weeks and either surgical ligation, resection, and grafting or endovascular occlusion of the affected vessel. Conservative management with antibiotics alone has not been favored because of the perceived risk of clot propagation and distal embolization and hemorrhage in untreated pseudoaneurysm. Although there are very limited data regarding endovascular occlusion, it is currently the favored method as it is felt to have less operative risk. Surgical internal carotid artery ligation has a reported mortality rate higher than 17%.

The main risk of these procedures is a large ischemic stroke, necessitating delineation of collateral flow through the Circle of Willis prior to carotid sacrifice. In the endovascular approach, coils or balloons are placed proximal and distal to the pseudoaneurysm to exclude blood flow within the pseudoaneurysm. In our case, only the proximal occlusion with coils was performed. Distal occlusion was not felt to be required and would have been technically difficult, as the preoperative cerebral angiogram demonstrated no antegrade flow in past the pseudoaneurysm. Furthermore, contralateral internal carotid artery and vertebral artery injections demonstrated occlusion of the internal carotid artery immediately distal to the pseudoaneurysm.

In review of the literature, there are only 31 published pediatric cases of infectious extracranial carotid pseudoaneurysms described since 1968 in 30 case reports. The characteristics of the individual cases are summarized in Table 1. Previous cases have involved, on average, older patients than ours (range 6 months to 17 years, average age 14.1 years). Most previously reported cases have been male (18/31, 58%). The presentation of our case was in many ways similar to prior cases. Sore throat with pharyngitis was present in 15/31 patients. Dysphagia was mentioned in 10/31 cases and respiratory distress in 5/31. Hemorrhage during the disease course occurred in the majority (17/31) representing a serious, potentially fatal complication. An external neck mass, similar to our patient, was noted in 20/31 cases and intraoral mass was noted in 12/30. Prior tonsillectomy or tonsillitis was described in 9/31 cases although timing relative to the presentation was not consistently mentioned. Treatment of the retropharyngeal/parapharyngeal abscess included oral or intravenous antibiotics in 22/31 cases reported. Two cases did not receive antibiotics, and for 7 cases treatment of the abscess was not described. Eleven patients also had drainage of the abscess performed. Our patient received

both intravenous antibiotics and drainage of the retropharyngeal abscess.

The method for treating patients has evolved since the first case report in our cohort, as endovascular technology and techniques have progressed. The majority of recent cases have been performed with endovascular coils and stents. Of the 19 cases reported since the millennium, 12 cases have been entirely endovascular, 3 have been purely surgical, 3 have been a combined approach and 1 patient was treated conservatively with antibiotics and drainage of the retropharyngeal abscess. Endovascular coiling was planned in the latter patient but was not performed when a decrease in size of the pseudoaneurysm was observed with aggressive management of the abscess.

The majority of cases reported no neurologic sequelae (17/31, 55%). Horner's syndrome was the most common persistent neurologic deficit affecting 9/31 cases. Documented ischemic infarction, as seen in our case, is uncommon. Only 2 patients previously reported had hemiparesis (presumably secondary to infarction), which resolved. The right arm weakness seen in our patient also completely resolved. It was not possible to determine whether the infarctions noted on our patient's MRI occurred before the endovascular procedure or as a result of the procedure since the right arm had been immobilized for an intravenous line on admission to the hospital, prior to the procedure. The remaining deficits described in the literature included vocal cord paralysis, dysphagia, as well as hypoglossal, and facial nerve, palsies.

Internal carotid artery pseudoaneurysm is an uncommon but potentially life-threatening complication of retropharyngeal/ parapharyngeal abscess in children. Appropriate imaging studies to evaluate for this complication is recommended.

Author Contributions

MR, DN, and DR were responsible for drafting and preparing the manuscript. JK provided critical review and editing. DR is the senior and corresponding author.

Declaration of Conflicting Interests

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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Ethical Approval

Requirement for informed consent was waived by the IRB given minimal risk to the subjects from retrospective review of records.

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