American Journal of Otolaryngology and Head and Neck Surgery

Case Report
Published: 20 May, 2021

Internal Carotid Artery Pseudoaneurysm Presenting with Bloody Otorrhea and Horner's Syndrome in a Child

Fadhel Almolani[1], Ali Almomen[2], Jamal Alhamam[3], Hussain Alsheef[3], Amirah Aldhurais[3], Balsam Alawami[3], Salma Alhammad[3], Hussain Almulla[3], Zahra Almoumen[3], Kauthar Altawfiq[3] and Zainab Ali[3]*

[1]Department of Radiology, King Fahad Specialist Hospital, Saudi Arabia

[2]Department of ENT, King Fahad Specialist Hospital, Saudi Arabia

[3]King Fahad Specialist Hospital, Saudi Arabia

Abstract

Cervical internal carotid artery pseudoaneurysm is a rare disorder especially in children. The most common cause of acquired internal carotid artery pseudoaneurysm are trauma and infection. We present a case of a 10 months old girl who present with life threatening ear bleeding and ptosis after history of fever. We diagnosed her with retropharyngeal abscess complicated by ruptured internal carotid artery pseudoaneurysm. She was treated by endovascular carotid artery sacrifice with coiling after establishing her good collateral status of the intracranial circulation.

Keywords: Carotid artery pseudoaneurysm; Endovascular; Horner's syndrome

Introduction

Cervical internal carotid artery pseudoaneurysms are rare diseases in the children. The most common etiology in this age group includes trauma and infection. They can present as simple as non-specific neck swelling to profuse life threatening hemorrhage. We present a case of a 10-month-old girl who presented with profuse bloody otorrhea and left eye ptosis following an episode of fever and ear infection. We will discuss our endovascular approach and rational for this treatment.

Case Presentation

A 10-month-old girl presented to outside facility with history of fever for one week. She had left ear discharge that became bloody two days prior to evaluation. She was started on antibiotics ear drops. On the second day after starting the antibiotics, she had an episode of profuse bleeding from the ear. She was taken to the hospital. Her temperature was 38.2°C. Laboratory evaluation showed hemoglobin of 6 gm/dL. She underwent emergent CT which showed retropharyngeal/parapharyngeal abscess with variable density as well as pseudoaneurysm of the high cervical segment of the left internal carotid artery (Figure 1).

The patient was transferred to our facility under ENT on the same day. Evaluation at our hospital showed packing material in the ear with clotted blood. She has mild left eye ptosis. The patient was started on intravenous antibiotics. Multidisciplinary meeting including the ENT surgeon, pediatric intensivist and neurointerventional radiologist was conducted. The consensus was made to proceed with diagnostic angiogram and possible endovascular treatment. The procedure was done the next day. The diagnostic angiogram showed left internal carotid artery tapering with faint filling of the ICA pseudoaneurysm and petrous segment of the ICA (Figure 2A, 2B). The left anterior and middle cerebral arteries are filled through the anterior and left posterior communicating arteries respectively. Microcatheter angiogram of the left ICA pseudoaneurysm revealed no antegrade flow out of the pseudoaneurysm into the petrous segment of the ICA (Figure 2C). We have decided to proceed with coil embolization of the pseudoaneurysm non-thrombosed portion as well as the feeding segment of the left internal carotid artery. Post-embolization angiogram showed no filling of the pseudoaneurysm or feeding internal carotid artery with patent external carotid artery (Figure 3) with faint filling of the cavernous segment of the left internal carotid artery through collaterals.

The patient woke up from anesthesia intact with no new neurologic deficits. On the second post embolization day, the patient was noticed to have small amount of active fresh bleeding from the left ear. The patient was shifted for CT angiogram. The CT showed no expansion of the hematoma or

OPEN ACCESS

Correspondence:
Ali Almomen, Department of ENT, King Fahad Specialist Hospital, Dammam, MOH, Saudi Arabia,
E-mail: alihalmomen@yahoo.com

Received Date: *22 Apr 2021*
Accepted Date: *17 May 2021*
Published Date: *20 May 2021*

Citation:
Almolani F, Almomen A, Alhamam J, Alsheef H, Aldhurais, Alawami B, et al. Internal Carotid Artery Pseudoaneurysm Presenting with Bloody Otorrhea and Horner's Syndrome in a Child. Am J Otolaryngol Head Neck Surg. 2021; 4(4): 1133.

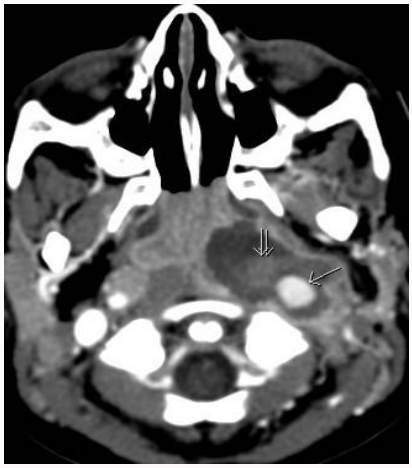

Figure 1: Contrast enhanced Tran's axial CT image of the upper cervical region showing the left retropharyngeal/parapharyngeal abscess with hyperdense/hypodense components (double arrows). The left internal carotid artery terminates in the collection as dilated sac (single arrow) consistent with pseudoaneurysm.

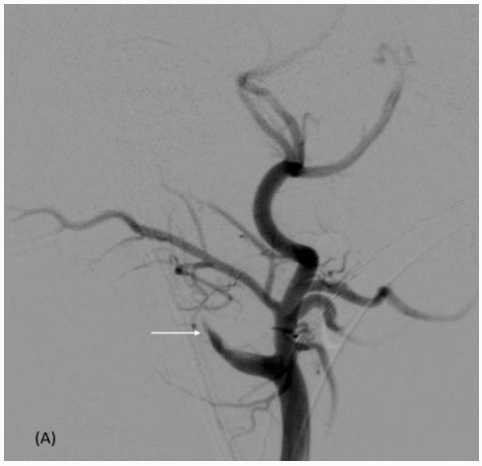

Figure 2A: Left common carotid artery angiogram in the lateral projection shows tapering of the internal carotid artery (single arrow).

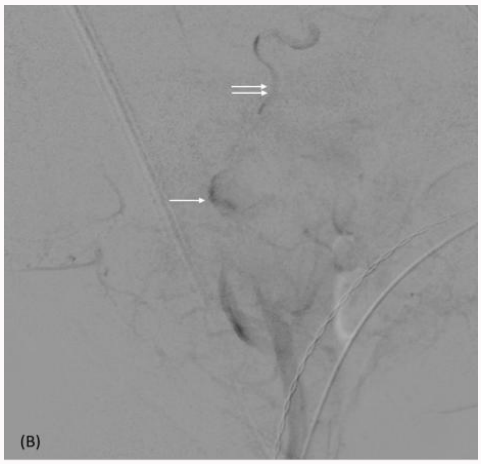

Figure 2B: Left common carotid artery angiogram in the lateral projection late arterial phase shows partial filling of the left ICA pseudoaneurysm (single arrow) with collaterals faintly filling the petrous segment of the left ICA (double arrows).

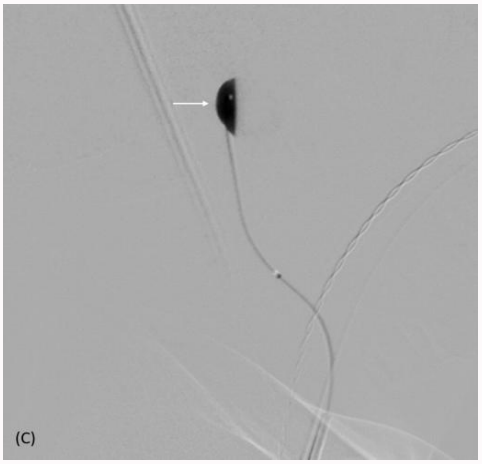

Figure 2C: Microcatheter angiogram within the left ICA pseudoaneurysm shows stagnation of contrast (single arrow) with no antegrade outflow into the intracranial ICA.

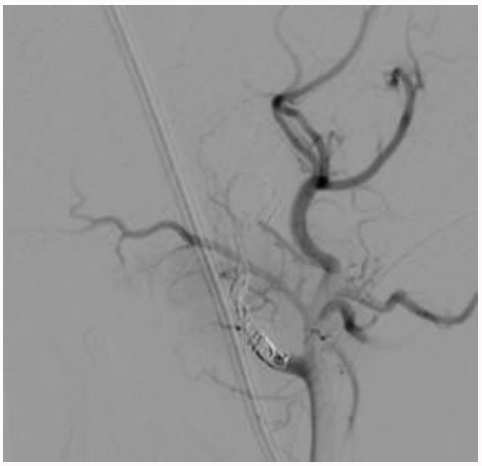

Figure 3: Left common carotid artery angiogram post embolization in lateral projection shows sacrifice of the ICA with coil filling the pseudoaneurysm with contrast filling.

filling of the pseudoaneurysm. The patient was shifted to angiography suite. Right internal carotid and left vertebral arteries cerebral angiogram showed filling of the left anterior and middle cerebral arteries with no retrograde filling of the pseudoaneurysm. Left common carotid artery angiogram showed occlusion of the aneurysm and ICA with delayed filling of the petrous ICA through collaterals (Figure 4A, 4B). The left external carotid artery and bifurcation were embolized with coils (Figure 4C). The patient woke up from anesthesia with no new neurologic deficit. The patient was discharged on day 14 post-embolization on oral antibiotics. The patient had no further attacks of bleeding.

Discussion

The cervical internal carotid artery aneurysms can be classified according to the histopathological findings into true or false aneurysm. The true aneurysms will contain the three layers of the artery into its wall. It can also be classified according to its pathogenesis into congenital or acquired. The internal carotid artery pseudoaneurysms, especially infectious are rare entities [1]. It was more common in the preantiobiotics era [2]. The most common causes of pseudoaneurysm

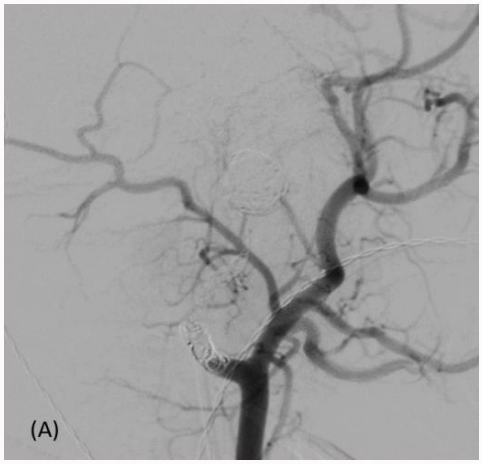

Figure 4A: Left common carotid angiogram lateral projection showed occlusion of the pseudoaneurysm with residual stump of the ICA origin.

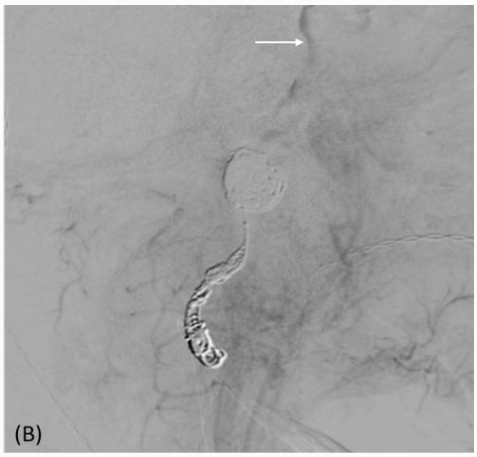

Figure 4B: Left common carotid angiogram lateral projection late arterial phase showed no filling of the pseudoanurysm with faint opacification of the petrous segment of the ICA (single arrow).

in children are from trauma and infections [3]. Localized infection results in higher rate of involvement in children secondary to the close proximity and direct infiltration and weakening of the walls [4]. Infection can also involve the internal jugular vein resulting in thrombophlebitis and Lemierre syndrome. Trauma results in disruption of the arterial wall which can be blunt or iatrogenic secondary to surgery or intervention [5-7].

The clinical presentation varies from benign appearance to florid life threatening. It might present as fever with neck swelling mimicking inflammatory disease [8,9]. If the mass increase in size, it will have local pressure effect on the adjacent structures and present as dysphagia or dyspnea [10]. The compression and stretching of the adjacent cranial nerves will result in hoarseness or Horner's syndrome like our patient [11]. Thrombus might form in the internal carotid artery pseudoaneurysm and migrate into the intracranial circulation which present as stroke [12]. Due to its proximity to the aerodigestive tract, patients can present as life threatening epistaxis, hematemesis or bloody otorrhea [13,14].

Imaging evaluation in children usually starts with ultrasound due to its availability and lack of radiation. It can be used as a screening tool. Further evaluation needs CT or MR imaging to evaluate areas

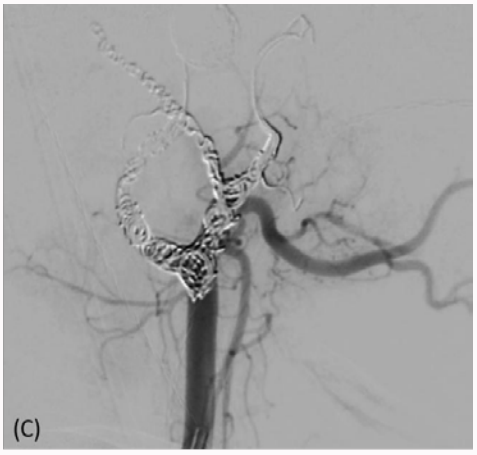

Figure 4C: Left common carotid angiogram lateral projection showed occlusion of the external carotid artery, stump of the ICA origin and bifurcation with residual filling proximal ECA branches.

cross to the skull base and collateral circulation. This can be combined with CT or MR angiography. The gold standard for arterial imaging is conventional angiogram which allows better evaluation of collaterals as well as simultaneous treatment.

The natural history of the disease without treatment shows high mortality rate without treatment reaching 77% [2]. The treatment includes surgical or endovascular options. The choice of treatment takes into the consideration the size of the pseudoaneurysm, its location, the status of collaterals and if the presenting symptom is life threatening hemorrhage. The surgical options include ligation, excision of the pseudoaneurysm with or without bypass. In children, the small size of the vessels makes vessel preserving surgery challenging with higher morbidity [15].

The endovascular option is gaining popularity due to its lower morbidity and mortality [16]. It has both reconstructive and destructive techniques depending on the patient's presentation and collaterals status. The reconstructive endovascular techniques include the use of stent-grafts across the neck of the pseudoaneurysm or stent-assisted coil embolization of the pseudoaneurysm [17]. If the patient has sufficient collaterals and presenting with life threatening bleeding, parent vessel sacrifices with proximal and distal trapping if feasible is taken as the treatment of choice [17]. Even if the patient tolerates the parent vessel occlusion acutely, the long-term effect is not without risk. Changing the flow dynamics with increase flow across the anterior and posterior communicating arteries that might result in flow related aneurysms [18].

Considering all these issues, we have chosen for our patient parent vessel sacrifice with coil embolization including the pseudoaneurysm. Backdoor embolization was not needed due to absence of outflow from the pseudoaneurysm. Although, the patient tolerated the occlusion acutely due to the good collaterals, long term follow up is advised to screen for flow related *de novo* aneurysm from the hyperdynamic circulation.

In conclusion, although cervical internal carotid artery pseudoaneurysm is a rare entity, having it in the differential diagnosis in children who presents with neck infection along with swelling pulsatile or not or bleeding is warranted for prompt recognition and treatment to save patient's life.

References

1. Beningfield A, Nehus E, Chen A, Yellin S. Pseudoaneurysm of the internal carotid artery after retropharyngeal abscess. Otolaryngol Head Neck Surg. 2006;134(2):338-9.
2. Salinger S, Pearlman S. Hemorrhage from pharyngeal and peritonsillar abscesses: report of cases, resume of the literature and discussion of ligation of the carotid artery. Arch Otolaryngol. 1933;18(4):464-509.
3. Gralla J, Brekenfeld C, Schmidli J, Caversaccio M, Do DD, Schroth G. Internal carotid artery aneurysm with life-threatening hemorrhages in a pediatric patient: Endovascular treatment options. J Endovasc Ther. 2004;11(6):734-8.
4. Pourhassan S, Grotemeyer D, Fokou M, Heinen W, Balzer K, Ramp U, et al. Extracranial carotid arteries aneurysms in children: Single-center experiences in 4 patients and review of the literature. J Pediatr Surg. 2007;42(11):1961-8.
5. Lago DM, Koning JL, Naheedy J, Leuin S. Carotid artery pseudoaneurysm: A rare complication following tonsillectomy. Int J Pediatr Otorhinolaryngol. 2015;79(12):2428-32.
6. Singhal M, Gupta V, Singh P, Lal A, Behra A, Khandelwal N. Iatrogenic life threatening pseudoaneurysms of extracranial internal carotid artery after fine-needle aspiration of cervical lymph nodes: Report of two cases in children. Cardiovasc Interv Radiol. 2009;32(6):1260-3.
7. DeFatta RJ, Verret DJ, Bauer P. Extracranial internal carotid artery pseudoaneurysm. Int J Pediatr Otorhinolaryngol. 2005;69(8):1135-9.
8. Lee NN, Long G, Ngai S, Sahrir S, Parker A, Lamont AC. Right internal carotid pseudoaneurysm mimicking a retropharyngeal abscess in a child. Med J Malaysia. 2004;59(5):685-7.
9. Gupta R, Patro SK, Chauhan N, Kumar A. A giant pseudoaneurysm mimicking retropharyngeal abscess in a child. Pediatr Emerg Care. 2019;35(5):e79-83.
10. Tannuri U, Mendes de Almeida N, Piske R, Matsumoto T. Giant pseudoaneurysm of the internal carotid artery causing upper airway obstruction in a 10-month-old infant treated by endovascular occlusion and surgical drainage. J Pediatr Surg. 2003;38(9):1393-5.
11. Flaherty PM, Flynn JM. Horner syndrome due to carotid dissection. J Emerg Med. 2011;41(1):43-6.
12. Ruff MW, Nasr DM, Klaas JP, Renaud DL. Internal carotid artery pseudoaneurysm and ischemic stroke secondary to retropharyngeal and parapharyngeal abscess. J Child Neurol. 2017;32(2):230-6.
13. da Silva PS, Waisberg DR. Internal carotid artery pseudoaneurysm with life-threatening epistaxis as a complication of deep neck space infection. Pediatr Emerg Care. 2011;27(5):422-4.
14. Gralla J, Brekenfeld C, Schmidli J, Caversaccio M, Do D-D, Schroth G. Internal carotid artery aneurysm with life-threatening hemorrhages in a pediatric patient: Endovascular treatment options. J Endovasc Ther. 2004;11(6):734-8.
15. Pourhassan S, Grotemeyer D, Fokou M, Heinen W, Balzer K, Ramp U, et al. Extracranial carotid arteries aneurysms in children: Single-center experiences in 4 patients and review of the literature. J Pediatr Surg. 2007;42(11):1961-8.
16. DuBose J, Recinos G, Teixeira PG, Inaba K, Demetriades D. Endovascular stenting for the treatment of traumatic internal carotid injuries: Expanding experience. J Trauma. 2008;65(6):1561-6.
17. Balasundaram P, Sebastian LJD, Jain N, Prabhakar A, Garg A, Gaikwad S. Management of arterial pseudoaneurysms of the neck in a pediatric population: An endovascular case series and review of literature. World Neurosurg. 2019;125:e273-81.
18. Briganti F, Cirillo S, Caranci F, Esposito F, Maiuri F. Development of "*de novo*" aneurysms following endovascular procedures. Neuroradiology. 2002;44(7):604-9.