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Transarterial embolization with N-butyl 2-cyanoacrylate for the treatment of recurrent secondary post-tonsillectomy hemorrhage

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Abstract

Background: Postoperative hemorrhage is a potential complication of tonsillectomy, and early diagnosis and adequate management are mandatory to prevent hemorrhagic shock. The aim of this study was to assess the safety and efficacy of transcatheter embolization of recurrent post-tonsillectomy hemorrhage by using N-butyl 2-cyanoacrylate.

Results: We performed a retrospective analysis of the medical records, imaging findings, technical details, and clinical outcome of eight patients with delayed post-tonsillectomy hemorrhage who underwent endovascular embolization. All patients were followed up; technical and clinical successes of treatment were evaluated. All patients had arterial pseudoaneurysm as a source of bleeding. The injured artery was facial artery in four patients, lingual artery in three patients, and linguofacial trunk in one patient. All lesions were treated by endovascular embolization using NBCA glue. All patients were successfully embolized with no clinical complications. In one patient with pseudoaneurysm at the ostium of the facial artery, after filling the pseudoaneurysm with glue, inadvertently proximal reflux into the adjacent part of the external carotid artery (ECA) occurred leading to its occlusion with no related immediate or delayed complication.

Conclusion: Endovascular embolization is an effective and almost safe procedure in the management of recurrent post-tonsillectomy bleeding. In such cases, we can use NBCA glue with certain precautions as an effective appropriate embolizing agent.

Keywords: Endovascular embolization, NBCA, Post-tonsillectomy, Hemorrhage

Background

Among the otolaryngological procedures, tonsillectomy is one of the commonest surgeries performed representing about 20–40% [1]. It is considered simple but, in comparison with other similar operations, it has potential and sometimes serious complications [2].

Post-tonsillectomy hemorrhage represents the most common complication, reported in about 3–4% of cases [3]. Hemorrhage may be early within the first 24 h after surgery (primary hemorrhage) or later on (secondary hemorrhage), usually between 7 and 10 days after surgery. Early bleeding is related to operative technique or a bleeding diathesis. Delayed hemorrhage may be caused by a vascular injury, mostly pseudoaneurysm. The lingual artery is the most commonly injured artery. The facial artery and common linguofacial trunk injuries were less frequently reported [3–9]. Internal carotid artery pseudoaneurysm following tonsillectomy is a rare but potentially lethal complication [10].

The risk of potential arterial injury following tonsillectomy is due to variable course and close proximity of

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the great arteries to the tonsil, rich blood supply of the tonsillar region, and the possibility of an aberrant medialized course of the internal carotid artery [11]. Blunt or penetrating trauma during tonsil dissection and/or suturing may result in a periarterial hematoma which expands with time. Subsequently, fibrotic tissue and endothelial cells will encapsulate the hematoma. Liquefaction of the central part of the hematoma results in potential space that communicates to the injured artery, resulting in pseudoaneurysm [12].

Proper and early diagnosis depends on clinical suspicion. Delayed and repeated attacks of gushing hemorrhage with spontaneous stoppage appear to be a significant clinical marker [12].

Local maneuvers and/or surgical ligation of external carotid artery (ECA) branches were the usual ways of management. Surgical hemostasis can be very challenging for several reasons: (1) the bleeding may have temporarily stopped due to a decrease in blood pressure secondary to the hemorrhage or anesthesia; (2) difficultly to locate the bleeding vessel due to its retraction within inflammatory pharyngeal tissue, or because of the bleeding is diffuse within one or both tonsillar fossae [3]. Endovascular treatment with embolization of the bleeding source represents a promising alternative treatment and is increasing over the last years [4, 8]. Different embolic agents were used including coils, gelfoam, particles, detachable balloon, and Onyx [3–9]. NBCA glue with certain precautions is an effective appropriate embolizing agent in such cases [13].

To our knowledge, there are few studies in the literature discussing the use of NBCA as a first intention for embolization of post-tonsillectomy vascular injury. Therefore, in our retrospective study, we will evaluate the efficacy and safety of using NBCA in endovascular embolization of recurrent post-tonsillectomy hemorrhage.

Methods

The Institutional Review Board of our Radiology Department approved the design of the study and the use of clinical data. Written consent was obtained from the patients or their parents prior to the procedures.

We performed a retrospective analysis of the medical records, imaging findings, and technical details from patients with delayed post-tonsillectomy hemorrhage who underwent endovascular embolization. They were referred from the department of otorhinolaryngology to our interventional radiology unit between January 2008 and January 2020.

Demographic data, clinical symptoms, type and location of the vascular injury, technical success, clinical success, and any complications were recorded.

All procedures were performed under general anesthesia. Puncture of the right common femoral artery was done via the Selidenger technique followed by the insertion of 4–5 French femoral sheaths. First, we performed diagnostic angiogram of the external and internal carotid arteries by using 4–5 French vertebral catheter (Cordis, USA). Diagnostic images were carefully assessed for the presence and localization of any vascular injury. Pseudoaneurysm or active contrast extravasation was an indicator of the site of hemorrhage. Next, supraselective catheterization of the injured artery was performed using a coaxial Renegade microcatheter (Boston Scientific, USA). By using road-map control, transcatheter embolization by N-butyl 2-cyanoacrylate glue (NBCA, histoacryl) (B. Braun, Germany) diluted with ethiodized oil (Lipiodol® Ultra-Fluid, France) in 2:1 ratio was performed. Our target was to fill the pseudoaneurysm and the related part of the injured artery with glue as well as to avoid non-target embolization of the normal branches or reflux into the ECA. After that, removal of the femoral sheath was done followed by manual compression of the entry site for 10 min.

Results

The study included 8 patients. One adult female aged 22 years and 7 children, two females and five males ranging in age between 3 and 9 years old. All patients complained from repeated attacks of post-tonsillectomy bleeding started from 4 to 10 days after surgery.

We reported the demographic data, clinical symptoms, type and location of the vascular injury, technical success, clinical success, and any complications (Table 1).

The performed angiogram showed pseudoaneurysm of facial artery in four patients (Figs. 1 and 2), lingual artery in three patients, and linguofacial trunk in one patient. In one patient, the pseudoaneurysm was arising at ostium of the facial artery from the ECA (Fig. 3). Five of the lesions were on the right side while it was on the left side in three patients.

All patients were treated by embolization using NBCA (histoacryl) glue diluted with Lipiodol in 2:1 ratio.

In seven cases, the pseudoaneurysm was completely occluded with occlusion of the adjacent part of the injured artery. No non-target or distal branch embolization occurred.

In the patient with pseudoaneurysm at the ostium of the facial artery, after filling the pseudoaneurysm with the concentrated histoacryl glue, inadvertently proximal reflux into the adjacent part of the ECA occurred resulting in its occlusion. No immediate or delayed complication occurred up to 6 months. This is because of the presence of rich anastomosis and collaterals at this area.

Technical success was defined as complete occlusion of the targeted vessel with absence of the previously

Table 1 Demographic data and results

	Sex	Age (years)	Onset (days)	No of attacks	Side	Injured artery	Injury	Embolic agent	Outcome
1	F	22	7	3	R	Facial	Pseudoaneurysm	NBCA	No complication
2	M	5	4	2	R	Facial	Pseudoaneurysm	NBCA	No complication
3	F	7	6	3	R	Ostium of facial artery	Pseudoaneurysm	NBCA	Proximal reflux of NBCA into ECA with no related complications
4	M	3	10	4	L	Facial	Pseudoaneurysm	NBCA	No complication
5	M	9	5	4	L	Lingual	Pseudoaneurysm	NBCA	No complication
6	F	7	10	5	L	Lingual	Pseudoaneurysm	NBCA	No complication
7	M	6	5	4	R	Lingual	Pseudoaneurysm	NBCA	No complication
8	M	8	6	3	R	Linguo-facial trunk	Pseudoaneurysm	NBCA	No complication

viewed positive angiographic findings. Clinical success was defined as the improvement of signs and symptoms of bleeding after the procedure, with improvement of patient Hb level and absence for acute attacks for 30 days following the procedure. Technical and clinical success was reported in 100% of the cases

All patients were discharged after 2–3 days with no evidence of any recurrent bleeding. All patients were followed up 4 weeks after discharge. In addition, the patient with histoacryl proximal reflux was followed up every month for 6 months. In all patients, no recurrent bleeding or other complications occurred.

Discussion

Early diagnosis and adequate management of post-tonsillectomy hemorrhage is mandatory to prevent hemorrhagic shock and exsanguination [12]. Treatment options include local maneuvers, surgical ligation, and endovascular embolization. Endovascular embolization has some advantages. First, the initial diagnostic angiogram can be performed to detect the bleeding vessel followed by superselective occlusion of the injured artery in the same session. Second, it avoids the risk of injury of the vagal and accessory nerves [8]. In our study, the patients were sent to our unit after failure of the local maneuvers. No one of the patients underwent further surgery.

Van Cruijsen et al. [5] have stated that post-tonsillectomy pseudoaneurysm only involves children under the age of 10 years, due to the smaller anatomy and thinner pharyngeal muscles. However, in the current study, there was an adult patient. Studies by Pourhassan et al. [14] and Walshe et al. [15] have reported that the possibility of post-tonsillectomy pseudoaneurysm formation is not restricted by age.

The ascending pharyngeal artery, dorsal lingual artery, inferior tonsillar branch of facial artery, superior tonsillar branch of the descending palatine artery, and the ascending palatine artery supply the tonsils and are all at risk to develop a pseudoaneurysm [7]. In our study, the site of injury was facial artery in four patients, lingual artery in three patients, and linguofacial trunk in one patient. Most of the previous studies reported that lingual artery was most common site of injury in their studies [3, 4, 13]. To our knowledge, there are few published cases of facial artery injury as source of bleeding. Juszkat et al. [6] and Choi et al. [7] reported cases of recurrent post-tonsillectomy bleeding due to an iatrogenic facial artery pseudoaneurysm.

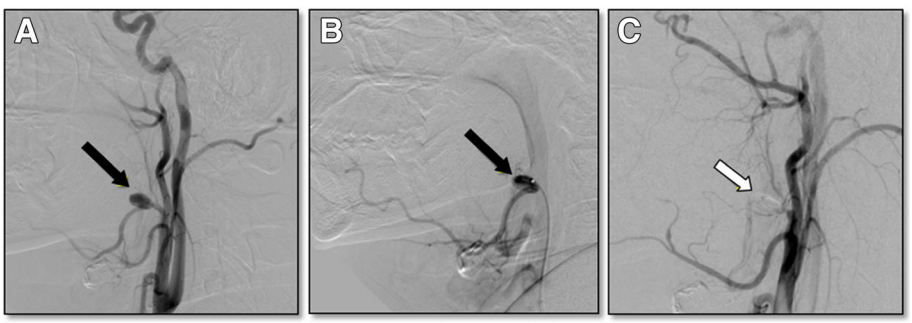

Fig.1 **a** ECA angiography shows a pseudoaneurysm (black arrow) arising at the primal part of the facial artery. **b** Superselective angiogram with the tip of microcatheter within the pseudoaneurysm (black arrow). **c** Post embolization ECA angiography, showing total occlusion of the pseudoaneurysm (white arrow)

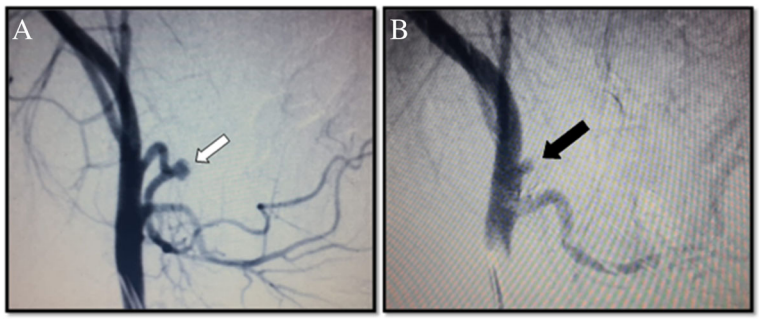

Fig. 2 a Common carotid artery angiography shows a pseudoaneurysm (white arrow) arising from the facial artery. **b** Post embolization angiography, showing occlusion of the pseudoaneurysm and facial artery (black arrow)

In our study, the post-operative onset of bleeding was from 4 to 10 days postoperatively. This corresponds with the study of Manzato et al. [8] that reported that cases of hemorrhage from pseudoaneurysm have presented in the range of 1–3 weeks and Hassan et al. [13] who stated that the bleeding started from 3 to 11 days after surgery. Moreover, there are some cases present over a month after surgery and so the possibility of pseudoaneurysm should remain even beyond the typical window of post-tonsillectomy hemorrhage [12].

In the current study, the eight patients were treated by endovascular embolization using concentrated NBCA glue diluted with Lipiodol. This matched with study done by Hassan et al. [13]. Manzato et al. [8] recommended trapping of the injured segment by microcoils to avoid possible distal migration of fluid or particulate embolic materials, such as glue or PVA particles. By using NBCA glue, we can achieve permanent occlusive effect, with a decrease possibility of recanalization, in contrast to possible delayed coil extrusion which may

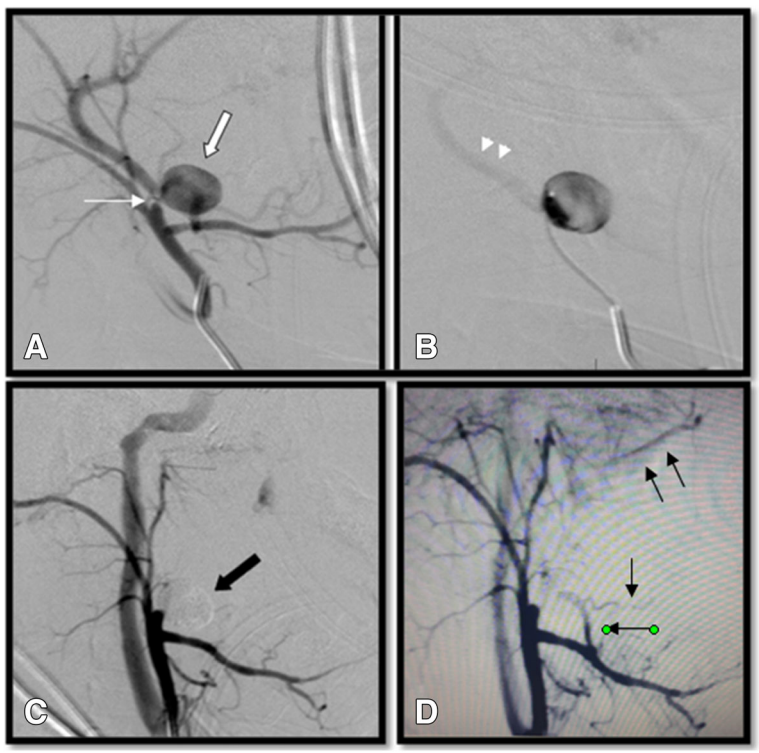

Fig. 3 a ECA angiography shows a pseudoaneurysm (thick white arrow) arising at the ostium of the facial artery (thin white arrow). **b** **S**uperselective angiography with the tip of microcatheter within the pseudoaneurysm with faint contrast reflux opacifying the main ECA (arrowheads). **c**, **d** Post embolization ECA angiography, showing occlusion of the pseudoaneurysm (black arrow) with proximal reflux of the glue and occlusion of adjacent part of the ECA. Few collaterals are refilling the internal maxillary and facial arteries in a retrograde manner (thin arrows)

occur with coils [3, 16]. Proximal occlusion of the injured artery by coils may lead to retrograde filling from collateral pathways resulting in recurrent bleeding; so, trapping of the injured segment should be done [7, 8]. For trapping of the pseudoaneurysm by microcoils, we should bypass the site of injury by the microcatheter. This will increase the risk of rupture of the pseudoaneurysm, and sometimes it could be technically difficult. The use of glue allows injection with the microcatheter situated just proximal to the pseudoaneurysm or slightly within it, infiltrating the whole segment without the need of pushing the microcatheter distally [13]. The glue has low cost as compared to microcoils and onyx. The histoacryl glue (NBCA) has a faster rate of polymerization than other liquid embolic agents as Onyx, so decreasing the possibility of distal migration and occlusion of the normal vessels [13].

Possible major complications of using histoacryl glue (NBCA) include inadvertent distal migration into the normal branches of the artery harboring the pseudoaneurysm. Also, proximal reflux into the main ECA or its other branches could occur. This can lead to local ischemic changes as tongue or lip necrosis [3]. In our study, we used concentrated histoacryl glue (NBCA) to avoid distal extension and non-target embolization. Another major possible complication is the extension of NBCA to the internal carotid artery either through retrograde reflux or through potentially dangerous anastomoses between the external carotid artery and internal carotid artery which may lead to cerebrovascular accidents and blindness. The possible existence of anastomoses determines the type of the embolizing agent that should be used [6, 7]. In all cases, we assess the pre embolization angiograms for any possible anastomosis with intracranial circulation. In our study, there was no major complication. In one patient with pseudoaneurysm at the ostium of the facial artery, inadvertently proximal reflux into the adjacent part of the ECA occurred leading to its occlusion, yet the patient did not experience any immediate or delayed complication due to the rich anastomosis and collaterals at this area. In the study done by Hassan et al. [13] in one patient, active bleeding occurred during the diagnostic angiogram followed by rapid injection of the glue leading to distal extension of glue into distal branches yet with no related complications.

The limitations of our study included the small number of patients and being a retrospective study.

Conclusion

Endovascular embolization by NBCA glue with certain precautions is considered an effective and safe therapeutic option in endovascular embolization of post-tonsillectomy pseudoaneurysm.

Abbreviations

ENT: Ear-Nose-Throat; NBCA: N-butyl 2-cyanoacrylate; PVA: Polyvinyl alcohol particles; ECA: External carotid artery.

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Authors' contributions

AS formulated the research goals, designed the study methodology, and supervised/actively participated in the research activity planning/execution. AN conducted/actively participated in the research process, performed the data collection/data analysis, and wrote the initial draft of the manuscript. SM assisted in data analysis, largely contributed in reviewing the manuscript, and contributed in the follow-up of the patients. All authors have read and approved the manuscript.

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Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

The Institutional Review Board of our Radiology Department approved the design of the study and the use of clinical data. Ethics Committee reference number is not available (was not provided). Written consent was obtained from the patients or their parents prior to the procedures.

Consent for publication

All patients (or their parents) included in this research gave written informed consent to publish the data contained within this study.

Competing interests

The authors declare that they have no competing interests.

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