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Case Report

A case series of preoperative endovascular embolization of nasopharyngeal angiofibroma ☆,☆☆

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ABSTRACT

Nasopharyngeal angiofibroma (NA) is a relatively rare, noncancerous, extremely vascular tumor, and it is only found in males. NA receives blood supply from various sources and can lead to major blood loss after surgical excision. Nowadays, endovascular embolization has the potential to reduce intraoperative bleeding and lower the recurrence rate. This case series aimed to describe the preoperative endovascular embolization of nasopharyngeal angiofibroma. In this study, we reported 3 cases of adolescents with NA diagnosed using CT angiography. All subjects had a successful embolization, which led to minimal postoperative bleeding and good outcome.

Introduction

Nasopharyngeal angiofibroma (NA) is a relatively rare, noncancerous, extremely vascular tumor, and it is only found in males. This tumor originates in the sphenopalatine foramen and may extend to the pterygopalatine fossa, paranasal sinuses, and nasal cavities, and may cause erosive changes [1].

Surgical excision of the tumor is the preferred and standard of care therapy. However, due to its location, NA received blood supply from various sources. Therefore, a preoperative angiographic procedure is needed to address the feeding artery and to describe tumor size and location [1]. Endovascular embolization has serious limitations that prevent it from being effective in the total obliteration and devascularization of nasopharyngeal angiofibroma before surgery. The most common technological issues involve catheter navigation, artery selection, and particle injection. Nonetheless, endovascular embolization is now a highly important procedure and is essential to devascularize nasopharyngeal angiofibroma before surgical excision to reduce intraoperative bleeding [2]. We reported 3 cases of NA treated with preoperative endovascular embolization before tumor removal.

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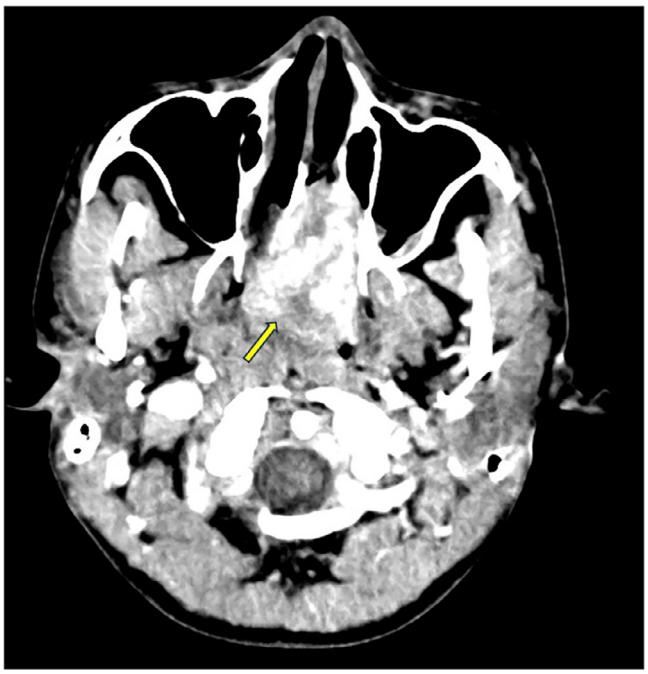

Fig. 1 – Head CT scan of the first patient. A solid mass arising from bilateral nasal cavity, especially the left one. The mass extended to the nasopharyngeal meatal, mucosal pharyngeal space bilateral, choana bilateral, obliterating the bilateral retropharyngeal space (yellow arrow).

Case presentation

The first case was a 15-year-old boy who presented with nasal congestion for the last 6 months. The patient also has 1 episode of nosebleeds that stopped on its own and a decrease in the ability to smell. Complaints of fullness in the face, lump on the palate, lump in the neck, blurred vision, shortness of breath, swelling of the face, facial pain, history of allergies, and history of similar complaints in the family were denied. Local status examination revealed that the right and left ear, oropharynx, maxillofacial, and neck were within normal limits. Inspection on both sides of the nasal cavity showed calm mucosa, eutrophic inferior turbinate, and a reddish mass at the level of the inferior turbinate. CT angiography (Fig. 1) showed a solid mass originating from both sides of nasal cavity extending to the nasopharyngeal meatus, bilateral mucosal pharyngeal space, bilateral choana which obliterated the bilateral retropharyngeal space and torus tubarius. The patient was diagnosed with Radkowski IA Juvenile Nasopharyngeal Angiofibroma.

The patient underwent digital subtraction angiography (DSA) and embolization followed by extirpation of the mass using a naso-endoscopic approach. The DSA procedure in super-selective catheterization was successfully carried out using a 2.7 Fr microcatheter on bilateral maxillary artery branches (Fig. 2). Embolization was carried out using histoacryl and lipiodol on the bilateral maxillary artery branches until the flow slowed down. Postembolization angiography showed that the feeding artery was no longer visible in the tumor stain in that area. Subsequent surgery resulted in a bluish mass with a smooth, spongy surface attached to the superior wall of the left nasopharynx. Then, extirpation of the mass was carried out using a guiding endoscope. The mass is saved as a biopsy sample and taken to the laboratory for histopathological examination. The DSA procedure and preoperative embolism succeeded in minimizing bleeding in the patient, so he only experienced 400 cc of bleeding. Residual bleeding is controlled by placing anterior Ollie tampons and Bellocq tampons. Postoperatively, the patient was treated with intravenous fluid Ringer Lactate 1500 cc/24 hours, Ceftriaxone 1 gram/12 hours, Paracetamol 1 gram/6 hours, and Tranexamic acid 500 mg/8 hours. Postoperative observations of the oropharynx showed no bleeding, blood seepage, or accumulation of bleeding in the installed drain. Intraoperative and postoperative observations showed no complications in the patient. The anatomical pathology results confirmed the patient's diagnosis as nasopharyngeal angiofibroma.

The second case was a 14-year-old boy with recurrent episodes of nosebleed for the last 6 months, impaired sense of smell and nasal congestion. Complaints of fullness in the ear, ringing in the ear, lump on the palate, lump in the neck,

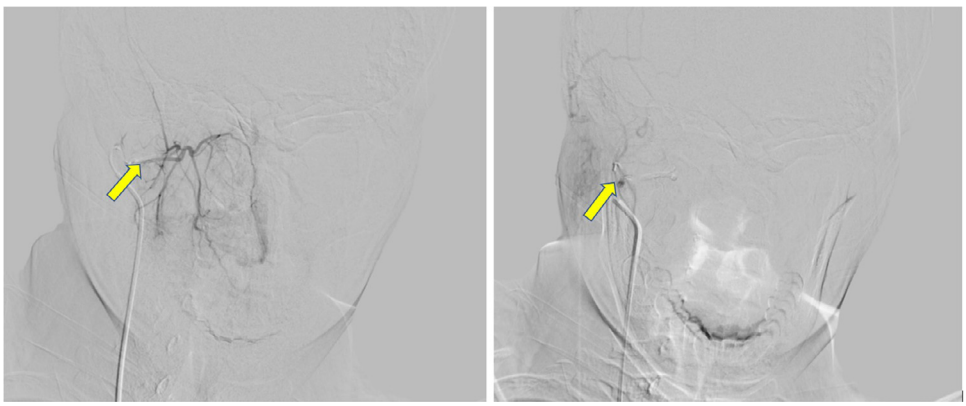

Fig. 2 – DSA anteroposterior view of the second patient: before (left) coiling and after (right) coiling. Embolization was performed on the right maxillary artery branches, which showed no feeding artery on the tumor stain (yellow arrow).

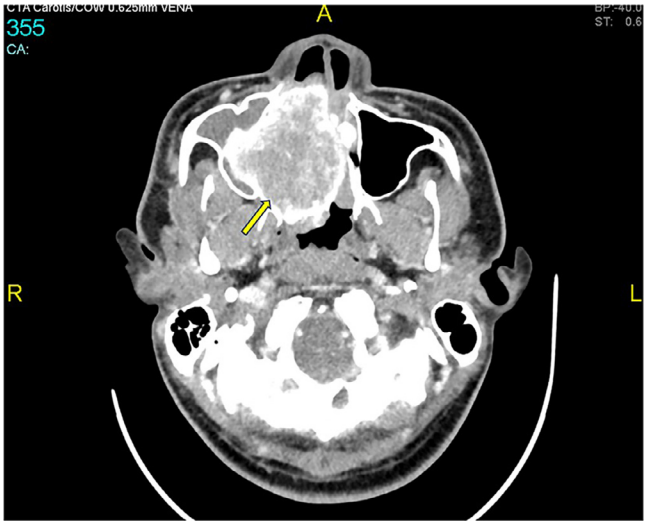

Fig. 3 – Head CT scan of the second patient. A hypervascular mass in the right nasal cavity region attaching to the right pterygoid process, superiorly obliterating the right sphenoidal sinus, laterally eroding the medial right maxillary sinus extending to the right maxillary sinus (yellow arrow).

blurred vision, shortness of breath, swelling of the face, facial pain, history of allergies, and history of similar complaints in the family were denied. An inspection of the right nasal cavity found calm mucosa, mucoserous secret, and a reddish mass with a smooth surface at the level of the inferior turbinate, filling up to the anterior nasal cavity. Meanwhile, in the left nasal cavity, there was a deviation of the septum to the left, narrow nasal cavity, eutrophic inferior turbinate, and serous secretion. Examination of the oropharynx revealed a mucous-colored mass in the posteromedial glossus region, with a stem measuring 2 × 1 × 1 cm. CT angiography also showed a hypervascular mass in the right nasal cavity area attached to the right pterygoid process, superiorly obliterating the right sphenoid sinus, right frontal sinus, and right sphenoid sinus. It extends to the right maxillary sinus; medially, it appears to be eroding and pushing the nasal septum to the left, eroding the medial orbital wall, and pushing into the right retrobulbar space. The mass appeared to receive arterial feeding from the bifurcation of the right external carotid artery (Fig. 3). From the results of this examination, the patient was diagnosed with Radkowski IIB Juvenile Nasopharyngeal Angiofibroma.

The patient was planned to undergo digital subtraction angiography (DSA) and embolization followed by extirpation of the mass using a midfacial degloving approach. Angiography of the right and left external carotid and its branches was performed using an approach via the right femoral artery using a 5 Fr vertebral macro-catheter. Super-selective catheterization was performed with a 2.7 Fr microcatheter on bilateral maxillary artery branches. Embolization was performed using a detachable coil on the bilateral internal maxillary arteries. Postembolization angiography (Fig. 4) shows that the feeding artery is no longer visible in the tumor stain in that area. Intraoperative findings showed a reddish mass with a smooth surface at the level of the inferior turbinate, filling up to the anterior nasal cavity. Then, extirpation of the mass was carried out using a midfacial degloving approach. The mass is saved as a biopsy sample and taken to the laboratory for histopathological examination. The DSA procedure and preoperative embolism succeeded in minimizing bleeding in the patient, so he only experienced 700 cc of bleeding. Residual bleeding is controlled by placing anterior Ollie tampons and Bellocq tampons. Postoperatively, the patient was treated with intravenous fluid Ringer Lactate 1500 cc/24 hours, Ceftriaxone 1 gr/12 hours, Paracetamol 1 gr/6 hours, Tranexamic acid 500 mg/8 hours. Postoperative observations of the oropharynx showed no bleeding, blood seepage, or accumulation of bleeding in the installed drain. Intraoperative and postopera-

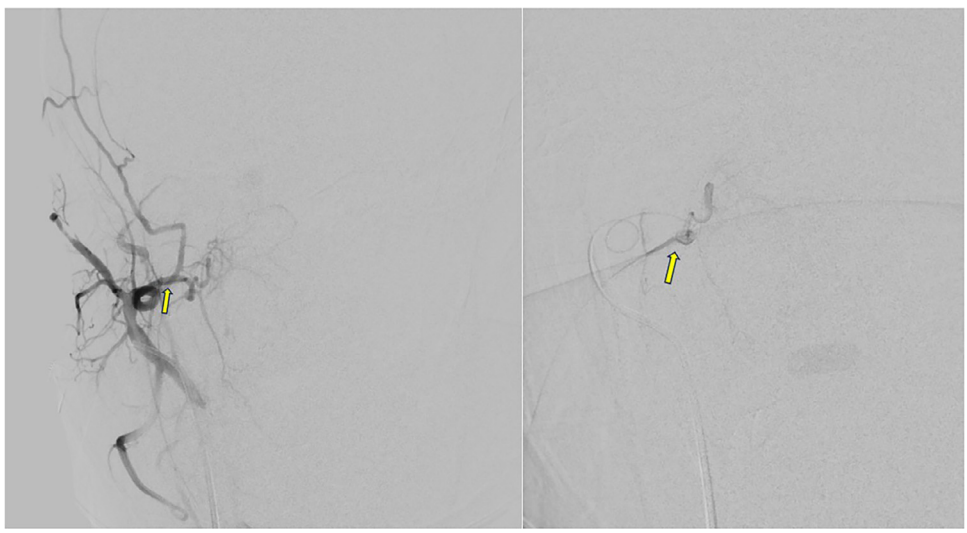

Fig. 4 – DSA anteroposterior view of the second patient: before (left) coiling and after (right) coiling. Embolization was performed on the right maxillary artery branches, which showed no feeding artery on the tumor stain (yellow arrow).

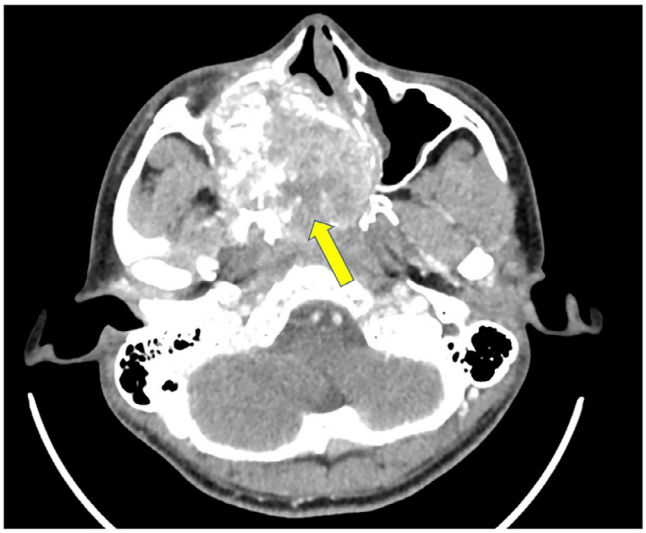

Fig. 5 – Head CT scan of the third patient. A hypervascular mass in the right nasal cavity region, superiorly obliterating the right sphenoidal sinus, laterally obliterating the right maxillary sinus, medially eroding and pushing the nasal septum to the left (yellow arrow).

tive observations showed no complications in the patient. The anatomical pathology results confirmed the patient's diagnosis as sinonasal ossifying fibroma.

The third case was a 14-year-old boy with recurrent episodes of nosebleed for the last 3 months. The patient also reported nasal congestion and an impaired sense of smell persisting for the past year. There were no associated symptoms such as lumps in the mouth, difficulty breathing, facial swelling, pain, blurred vision, double vision, headaches, neck lumps, or unexplained weight loss. The patient's eating and drinking habits were normal. There was no history of allergies, other medical conditions, familial malignancies, or smoking. The patient had no known exposure to dust or asbestos. There was no history of hypertension or diabetes mellitus. Physical examination showed a bluish mass with a smooth surface at the level of the inferior turbinate. In the left nasal cavity, physical examination showed calm mucosa, septum deviated to the left, eutrophic inferior turbinate, mucoserous secretion and narrow nasal cavity. CT angiography (Fig. 5) showed a hypervascular mass in the right medial nasal cavity adherent to the right pterygoid process. The lesion appeared to receive blood supply from branches of bilateral external carotid arteries, consistent with Juvenile Nasopharyngeal Angiofibroma. There was also a sinonasal mass. The patient was diagnosed with Radkowski IIA Juvenile Nasopharyngeal Angiofibroma. Subsequent DSA and embolization were conducted until the blood flow slowed down using histoacryl and lipiodol. Postembolization angiography (Fig. 6) did not reveal any feeding arteries supplying the tumor, indicating successful embolization. The impression was a vascular mass in the right nasopharynx post-DSA embolization, suggesting Nasopharyngeal Angiofibroma. The patient subsequently underwent surgical excision via lateral rhinotomy approach. Intraoperative findings showed A bluish-red mass was found in the right nasal cavity, adherent to the right nasopharyngeal wall. The mass was smooth, elastic, partly

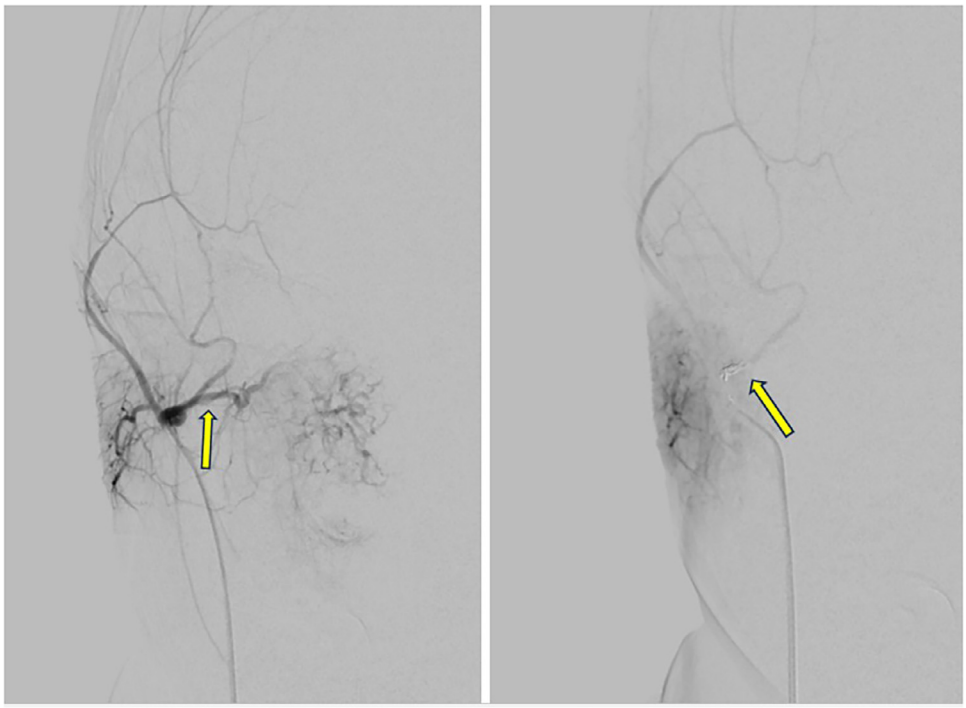

Fig. 6 – DSA anteroposterior view of the third patient: before (left) coiling and after (right) coiling. Embolization was conducted on the bilateral maxillary arterial branches. No feeding artery shown in the tumor stain of the area (yellow arrow).

fragile, and bled easily. Anterior ollie and Bellocq tampons were applied in the right nasal cavity, with no active bleeding noted. Postoperatively, the patient was treated with intravenous fluid Ringer Lactate 1500 cc/24 hours, ceftriaxone 1 gr/12 hours, paracetamol 1 gr/6 hours, and tranexamic acid 500 mg/8 hours. The anatomical pathology results showed a nasopharyngeal angiofibroma with a differential diagnosis of fibrosarcoma.

In conclusion, the 3 patients underwent digital subtraction angiography (DSA) and embolization using histoacryl and lipiodol until the blood flow slowed down. Post embolization angiography did not reveal any feeding arteries supplying the tumor, and tumor stain was no longer visible in the area, indicating successful embolization. Subsequently, surgical excision was carried out in all patients, which showed minimal intraoperative bleeding of approximately 400-700 cc. Postoperative follow-ups revealed no further complications or major bleeding in all patients. Further histopathological examinations confirmed the diagnosis of nasopharyngeal angiofibroma in all patients. This study has been approved by the IRB and all procedures and reports were in accordance with the Declaration of Helsinki and all patients have signed an informed consent to be included in the study.

Discussion

Angiofibroma is a rare type of tumor. Angiofibroma is a benign tumor that commonly develops in the superoposterior region of the sphenopalatine foramen. Although these tumors are histopathologically nonmalignant, they can cause injury to surrounding tissue. The most prevalent symptoms are 1-sided nasal blockage, a nasopharyngeal bulge, and recurrent nasal hemorrhage. This tumor has the potential to cause facial deformities, proptosis, migraines, and hearing loss [3–5]. Angiofibroma is typically diagnosed via radiological tests such as CT scans and MRIs. A radiological examination aims to determine tumor growth, bone damage, and the extent of angiofibroma. Furthermore, angiography may be conducted before the procedure to identify the arteries and characterize the size and position of the tumor [3,6]. In this study, 3 patients aged 13 to 15 were included. All the patients in this case were men, reported recurrent nasal bleeding and had chronic recurrent nasal bleeding. Other symptoms observed included nasal congestion and reduced nasal function. Only 2 patients reported persistent cold symptoms. All patients were diagnosed with angiofibroma using CT Angiography, which revealed a hypervascular solid mass in the nasal cavity.

In patients with nasopharyngeal angiofibroma, surgery is the primary treatment option; however, since angiofibroma is a hypervascularized and fragile mass, it is prone to bleeding. Embolization is currently one of the methods used before surgery. Embolization is a minimally invasive procedure used to revascularize the tumors. A catheter is inserted through the femoral artery and guided until the tip reaches the target blood vessel. The artery that vascularizes the tumor tissue is the target blood vessel in the case of angiofibroma. In order to revascularize the tumor, the embolization agent is then expelled through the catheter tip [7].

Studies have shown conflicting results regarding embolization procedure before tissue removal. Several studies showed no significant differences between tumor tissue removal procedures with embolization and those without [7]. Other studies, however, showed that patients without preoperative embolization are at risk for tumor recurrence [7]. Furthermore, if embolization is carried out before surgery, the cure rate may be higher. Several studies also showed that embolization 24 to 48 hours in advance improves bleeding control. By shrinking the size of the tumor and making it simpler to remove the tumor tissue, embolization can stop intraoperative bleeding. As a result, the recurrence rate of angiofibroma is low [8]. All patients in our case series had embolization, with intraoperative bleeding amounts of below 1000 mL.

The study by Chataut et al. demonstrated that the angiofibroma's stage affects intraoperative bleeding. Every patients in their study had embolization and had intraoperative bleeding amounts below 1000 mL. According to the study, patients with Radkowski stage I–III angiofibroma experienced intraoperative bleeding of less than 1000 mL, whereas patients with stage IV angiofibroma experienced intraoperative bleeding of more than 1000 mL [9]. Another study by Tan et al. also showed that embolization during preoperative management of Angiofibroma Radkowski Stage II/III significantly decreased intraoperative bleeding; however, this effect was not statistically significant for other stages [10].

In conclusion, the role of endovascular embolization may be beneficial in reducing intraoperative bleeding in patients with nasopharyngeal angiofibroma by devascularizing the tumor. All subjects in our study who underwent endovascular embolization had minimal intraoperative bleeding.

Ethics approval

This study has been approved by the Ethics Committee of University of Padjadjaran with an IRB approval number of DP.04.03/D.XIV.6.5/114/2024.

Patient consent

We confirm that an informed consent was obtained from patients reported in this study for publication of their case.

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