CASE REPORT

Imaging of Unilateral Meningo-ophthalmic Artery Anomaly in a Patient with Bilateral Nasopharyngeal Angiofibroma

Louise Louw, Johan Steyl[1], Eugene Loggenberg[2]

Departments of Otorhinolaryngology, [1]Basic Medical Sciences and [2]Clinical Imaging Sciences, University of the Free State, Bloemfontein, South Africa

Address for correspondence:
Prof. Louise Louw,
Department Otorhinolaryngology,
Faculty of Health Sciences, University of the Free State, Bloemfontein, South Africa.
E-mail: gnanll@ufs.ac.za

ABSTRACT

A 12-year-old boy with epistaxis presented with a rare midline nasopharyngeal angiofibroma that extended lateral into the pterygoid and infratemporal fossae. Pre-operative angiography revealed bilateral prominent feeder arteries and two major anastomotic connections, and a rare left meningo-ophthalmic artery (M-OA) anomaly that was the sole path of supply to the eye. A literature search using Pubmed and Medline was conducted. For imaging, a six-vessel study (i.e. external and internal carotid and vertebral arteries on both sides) was selected. Embolization of prominent tumor feeder arteries was unsafe for tumor extirpation, but super-selective embolization of both sphenopalatine arteries was performed to control epistaxis. The M-OA anomaly that originated from the maxillary artery (MA) was marked by an ophthalmic artery (OA) variant with orbital and ocular divisions that coursed through the superior orbital fissure and optic foramen, respectively, each with distinct branching patterns, a middle meningeal artery (MMA) with normal branches (i.e. anterior and posterior branches), and two branch variations (i.e. lacrimal and meningeal branches) that originated from the anterior branch of the MMA. The lacrimal branch coursed through a cranio-orbital foramen, but the meningeal branch remained outside the orbit. The anatomy of the right OA was normal. The left M-OA anomaly was considered incidental and not tumor-related since the tumor was more prominent on the right side, and no intra-orbital infiltrations occurred. Of clinical significance is that proximal embolization of MA or MMA carries a high risk of visual impairment in cases where M-OA anomalies are the sole mode of supply to the eye.

Key words: Dangerous extracranial–intracranial anastomoses, dangerous intracranial–extracranial connections, meningo-ophthalmic artery anomaly, nasopharyngeal angiofibroma, pre-operative angiography

INTRODUCTION

Upon reviewing the literature, it was found that nasopharyngeal angiofibroma and meningo-ophthalmic artery (M-OA) anomaly are relatively rare phenomena. In Denmark, the incidence of nasopharyngeal angiofibroma

is reported to be 3.7 per million males per year.[1] Tumor extirpation is the mainstay treatment, but the recurrence rate (20%, ranging between 5 and 50%) remains a challenge for the surgeon.[2] In the case of M-OA anomalies, the ophthalmic artery (OA) variant may supply the orbit completely or partially, in the absence or presence of a normal OA with internal carotid artery (ICA) origin, respectively.[3] Retinal supply from the middle meningeal artery (MMA) was also encountered.[4] The following incidences for OA origin from the MMA have been reported: 1.18% among 170 specimens, based on dissection studies in the USA;[5] and 1.45% among 1652 OAs, based on magnetic resonance imaging (MRI) studies in Japan.[3] To the best of our knowledge, an M-OA anomaly has not been previously reported in a patient with nasopharyngeal angiofibroma.

Awareness of vascular collateral routes with routine angiography can effectively contribute to strategic planning and surgical interventions for tumor extirpations. Embolization of the external carotid artery (ECA) is a useful tool in the devascularization of nasopharyngeal angiofibroma and in the treatment of epistaxis. However, the surgical interventionist needs to be aware of dangerous ECA–ICA connections to avoid neurological complications such as embolic stroke and palsies.[6] Among the potential orbital collateral routes the M-OA Is the most frequent[7] followed by a route from the anterior branch of the MMA.[4] and a route from the anterior branch of the MMA.[4] For embolization procedures, it is imperative to identify the source of the retinal supply to avoid visual complications.

This paper reports on unilateral M-OA and MMA orbital collateral routes as the sole supply of the left orbit in a patient with nasopharyngeal angiofibroma. Questions pertaining to the orbital collateral routes are the following: Why do they exist? Are they incidental or tumor-related?

CASE REPORT

A 12-year-old boy was admitted to our academic hospital and diagnosed with nasopharyngeal angiofibroma. Computed tomography (CT) scanning revealed a midline nasopharyngeal angiofibroma with prominent right-side infratemporal fossa infiltration [Figure 1]. Magnetic resonance imaging (MRI) confirmed exclusion of intra-orbital infiltrations [Figure 2]. A lateral rhinotomy to remove the tumor was required. Pre-operative angiography with the transfemoral approach was performed. Access to the right common transfemoral artery was gained with a 22 G Jelco, and a micro-puncture guide wire was advanced into the right common femoral artery. A 4 F pediatric sheath was advanced over the micro-puncture guide wire into the right common femoral artery. A headhunter catheter was used and 50 ml of
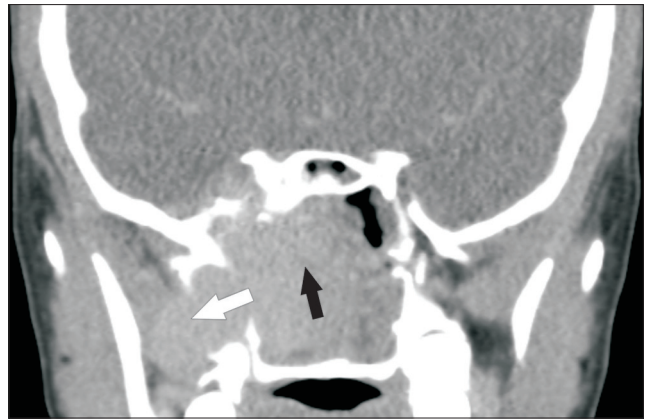

Figure 1: Twelve-year-old boy presented with nasopharyngeal angiofibroma diagnosed with unilateral meningo-ophthalmic artery anomaly. Computed tomography scan shows midline nasopharyngeal angiofibroma (black arrow), with prominent right-side infiltration into the infratemporal fossa (white arrow).

Jopamiron 300 was administered intra-arterially without any side effects. A six-vessel study, i.e. ECA, ICA, and vertebral artery (VA) on both sides, was selected to determine the blood flow routes of the angiofibroma for optimal tumor extirpation. Dangerous ECA–ICA connections rendered embolization of prominent feeder arteries unsafe, but super-selective fibrin glue embolization of both sphenopalatine arteries with a micro-catheter was performed in an attempt to control epistaxis. Intra-operatively, severe bleeding occurred and blood transfusion was required. Bleeding was attributed to the presence of multiple feeder arteries and extensive anastomotic networks. Post-operatively, the child still suffered from epistaxis because of the extensive nasal anastomotic networks encountered. The child had a difficult recovery, but was eventually discharged and scheduled for follow-up visits.

The pre-operative angiographic study revealed the following prominent feeder arteries: Ascending pharyngeal artery with multiple pharyngeal branches from the pharyngeal trunkus (bilateral), proximal maxillary artery (MA) with branches from the stem of the M-OA (left side) and the MMA and accessory meningeal artery (AMA) (right side), distal MA with sphenopalatine artery (i.e. the nasopharyngeal artery) (bilateral), and distal facial artery with nasal branches (bilateral). According to expectation, ethmoidal and nasal branches via the OA (i.e. the left OA variant and the right OA with ICA origin) and nasal branches via the distal facial artery (unilateral) nourished the midline nasopharyngeal angiofibroma. For the purpose of this paper, prominent feeder arteries and two major vascular networks are given in Table 1. Pre- and post-embolization angiograms of tumor vessels are presented in Figure 3.

Left angiograms revealed no OA with ICA origin, and an M-OA anomaly that originated from the MA of the ECA with an OA variant and MMA branch variations [Figures 4-6]. Of

particular interest in this case was that the OA variant had two intra-orbital parts, namely an orbital division with a distinct branching pattern to supply the nasal cavity and nose (i.e. ethmoidal and nasal branches) and an ocular division with a distinct branching pattern to supply the

| Table 1: Tumor feeder arteries, branches, and vascular networks |
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| Prominent feeder arteries |
| Ascending pharyngeal artery: Pharyngeal trunkus with branches (bilateral) |
| Proximal maxillary artery: Meningo-ophthalmic artery (left side), middle and accessory |
| Meningeal arteries (right side) |
| Distal maxillary artery: Sphenopalatine artery (bilateral) |
| Contributing branches |
| Ethmoidal and nasal branches via ophthalmic artery variant (left side) and normal |
| Ophthalmic artery (right side) |
| Nasal branches via distal facial artery (bilateral) |
| Major vascular networks |
| In pterygopalatine fossa (bilateral) |
| On pharyngeal mucosa (bilateral) |
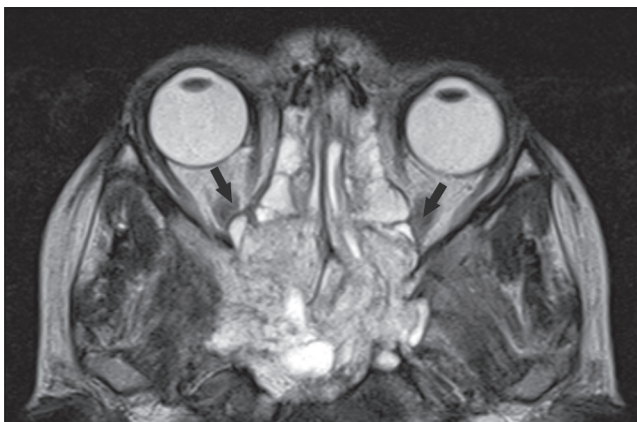
Figure 2: Twelve-year-old boy presented with nasopharyngeal angiofibroma diagnosed with unilateral meningo-ophthalmic artery anomaly. Magnetic resonance imaging shows no intra-orbital infiltrations (black arrows).
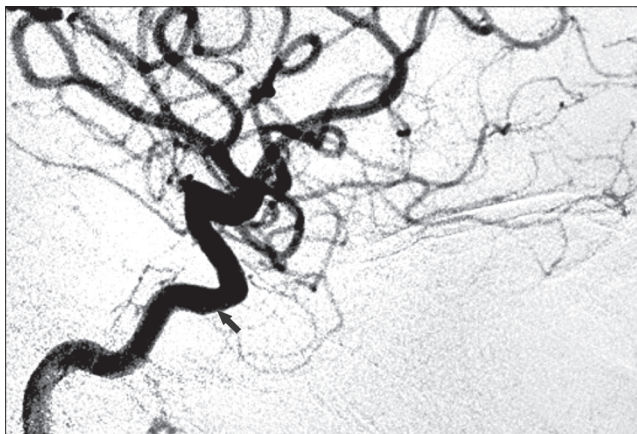

Figure 4: Twelve-year-old boy presented with nasopharyngeal angiofibroma diagnosed with unilateral meningo-ophthalmic artery anomaly. Left lateral view angiogram of the head shows internal carotid artery (black arrow) with no ophthalmic artery.

eyeball (i.e. ciliary and muscular branches) and the retina (i.e. the central retinal artery as witnessed by choroidal blush). The orbital and ocular divisions coursed through the superior orbital fissure and optic foramen, respectively, according to expectation. The MMA presented with two branch variations that originated from the anterior branch of the MMA, namely a lacrimal branch that coursed through a cranio-orbital foramen and a meningeal branch that remained outside the orbit.

DISCUSSION

Nasopharyngeal angiofibroma often originates in the vicinity of the sphenopalatine foramen and epistaxis is a symptom to be reckoned with. Severe intra-operative
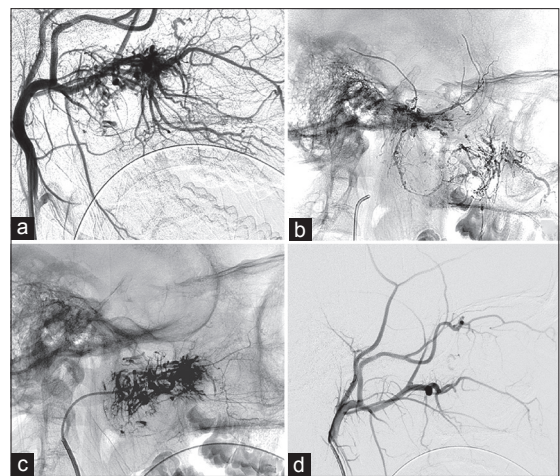

Figure 3: Twelve-year-old boy presented with nasopharyngeal angiofibroma diagnosed with unilateral meningo-ophthalmic artery anomaly. Right lateral view angiograms of head show a) pre-embolization of tumor vessels and b) post-embolization of tumor vessels. Left lateral view angiograms of head show c) pre-embolization of tumor vessels and d) post-embolization of tumor vessels.
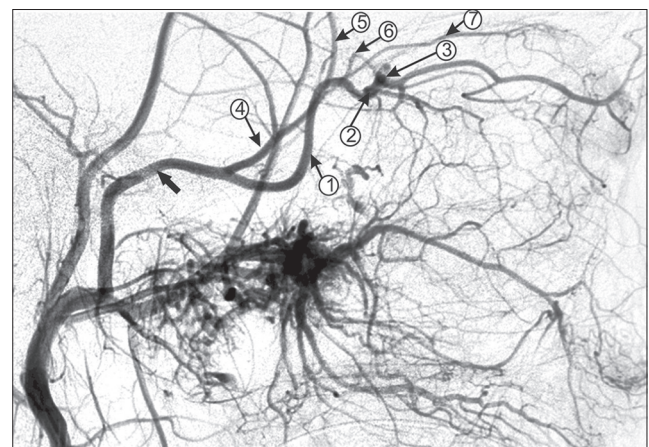

Figure 5: Twelve-year-old boy presented with nasopharyngeal angiofibroma diagnosed with unilateral meningo-ophthalmic artery anomaly. Left antero-posterior view angiogram of head shows meningo-ophthalmic artery anomaly (black arrow). (1: Ophthalmic artery variant; 2: Ocular division; 3: Orbital division; 4: Middle meningeal artery; 5: Anterior branch; 6: Meningeal branch; 7: Lacrimal branch).
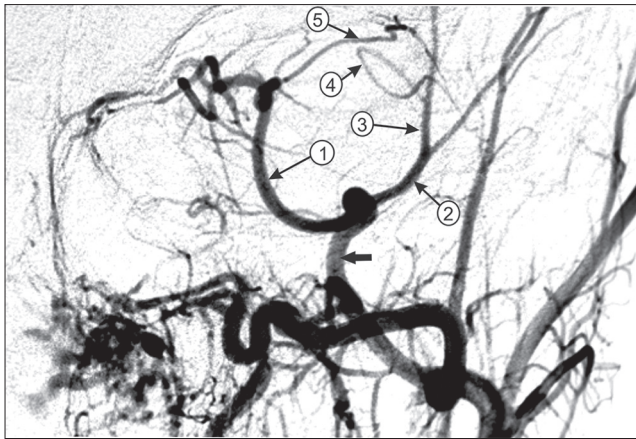
Figure 6: Twelve-year-old boy presented with nasopharyngeal angiofibroma diagnosed with unilateral meningo-ophthalmic artery anomaly. Left view angiogram of head shows meningo-ophthalmic artery anomaly (black arrow). (1: Ophthalmic artery variant; 2: Middle meningeal artery; 3: Anterior branch; 4: Meningeal branch; 5: Lacrimal branch).

Figure 7: Schematic presentations. a) Diagram shows normal development of the ophthalmic artery. b) Diagram indicates probable development of the meningo-ophthalmic artery anomaly. (1: Internal carotid artery; 2: Ophthalmic artery; 3: External carotid artery; 4: Middle meningeal artery; 5: Orbital branch; 6: Stapedial artery; 7: Maxillary artery; 8: Meningo-ophthalmic artery; 9: Meningeal branch; 10: Lacrimal branch).

bleeding in the current case is attributed to the presence of a rare Vidian artery that is often encountered in nasopharyngeal angiofibroma. A Vidian artery (i.e. the artery of the pterygoid canal) usually arises from the distal MA, or may have origin from the petrous ICA (30% incidence), or may serve as anastomosis between the ECA and the ICA.[8] Regardless of origin, a Vidian artery participates in two major vascular networks through anastomoses in the pterygopalatine fossa (with pharyngeal, ethmoidal, and sphenopalatine arteries) and on the pharyngeal mucosa (with ascending pharyngeal and meningeal arteries),[8] as witnessed in this patient. The presence of an inferolateral trunkus (ILT) with origin from the cavernous ICA (C4 segment) and its anastomotic connections with the MA, MMA, and OA must also be taken into consideration.[9] An ILT is often identifiable in the dissection specimen, but is less frequently seen on imaging and was not witnessed in this case.

Of particular interest to this paper is the M-OA anomaly. Embryological explanations for M-OA and MMA orbital routes are fetal anastomotic connections between the ICA and ECA and enlargement of fetal anastomosis between branches of the MMA and OA, respectively. In principle, the stapedial system plays a pivotal role in the development of M-OA and MMA orbital routes.[7,10] Suggested embryonic evolutionary changes for the orbital collateral routes (M-OA and MMA) encountered in this case are as follows: Regression of the proximal parts of the stapedial artery (SA) (origin: ICA) and primitive ophthalmic artery (POA) (origin: ICA) occurred, and an annex between the distal parts of the SA and POA formed, whereby the M-OA gained ECA origin. All orbital branches of the MMA are remnants of the SA.[10] See Figure 7 for schematic embryological presentations.
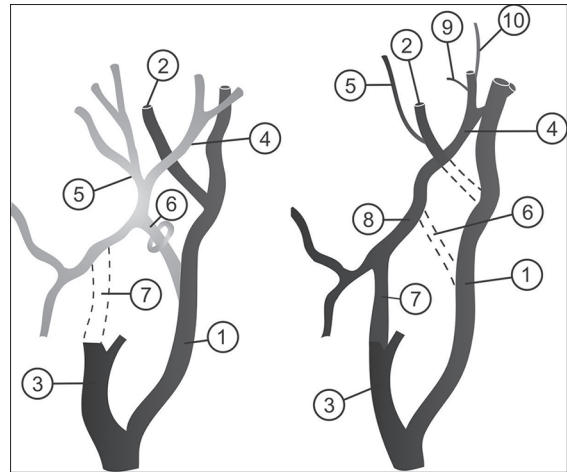
Upon decision-making whether the M-OA and MMA routes were tumor-related or incidental, it was evident that the latter was applicable in this case. In favor of the incidental presence of the M-OA and MMA routes were their unilateral (left side) presence, the prominent right-side occurrence of the tumor, and no intra-orbital infiltrations. Of clinical significance is that in cases where the M-OA is the sole supply to the eye, proximal embolization of the MA or MMA carries a high risk of visual impairment and must be avoided.

CONCLUSION

A pre-operative routine angiographic study in a patient who presented with a nasopharyngeal angiofibroma revealed an M-OA anomaly. The M-OA anomaly followed distinct patterns of orbital supply and was the sole supply to the eye. In this particular case, the M-OA anomaly was not tumor related and was considered incidental. Proximal embolization of the MA was considered unsafe for tumor extirpation, but embolization of both sphenopalatine arteries was a useful tool in the management of epistaxis. Documentation of this tumor entity and M-OA anomaly, both relatively rare, remains a valuable contribution to the literature.

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