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Case Report

Bilateral infraoptic A1 arteries in association with a craniopharyngioma: Case report and review of the literature

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Abstract

Background: While variation within the anterior cerebrovascular circulation is common, an infraoptic course of the proximal anterior cerebral artery (ACA), or infraoptic A1, is a relatively rare cerebrovascular anomaly. Associations with suprasellar neoplasms may occur, and accurate identification of this aberrant vessel during dissection is crucial to preventing vascular injury or stroke.

Case Description: We present the first reported case of surgically confirmed bilateral infraoptic A1 arteries associated with a craniopharyngioma. We review the relevant magnetic resonance imaging (MRI), angiographic, and intraoperative anatomic features of the infraoptic A1 to emphasize the importance of these variables when planning and performing surgery in the region of the anterior communicating artery (AComm) complex.

Conclusions: Awareness of the existence and clinical significance of this unusual anomaly can facilitate its recognition on preoperative studies and during dissection in the suprasellar space, allowing neurosurgeons to adjust operative plans accordingly.

Key Words: Anterior cerebral artery, cerebrovascular anomaly, craniopharyngioma, infraoptic A1, magnetic resonance angiography

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INTRODUCTION

Neurosurgeons must always be keenly aware of the anatomic variability of the cerebral vasculature. First described from an anatomic dissection by Robinson in 1959, an infraoptic course of the proximal anterior cerebral artery (ACA), or infraoptic A1, is a rare vascular anomaly.[1] The infraoptic A1 typically takes its origin from the proximal internal carotid artery (ICA) at or near the origin of the ophthalmic artery (OphA) before passing beneath the ipsilateral optic nerve and pursuing an aberrant interoptic course in the pre-chiasmatic cistern.[1-38] In the vast majority of reported cases, the anomaly was found in association with saccular aneurysms, generally at or near the ACA/anterior communicating artery (AComm) complex.

More rarely, this unusual anatomic variant may occur bilaterally. To date, the presence of bilateral infraoptic A1s has been reported only nine times in the literature, and in eight of these cases, the bilateral anomaly was associated with one or more aneurysms.[3,13,15,16,18,21,22,26,37]

To our knowledge, we present the first report of bilateral infraoptic A1 arteries associated with a suprasellar neoplasm.

CASE REPORT

Presentation

A previously healthy 37-year-old man presented to an outside hospital with a 2-month history of progressive headache, nausea, and intermittent blurred vision. A computed tomography (CT) scan of the head revealed a large suprasellar mass and obstructive hydrocephalus, and the patient was transferred to our facility for further evaluation.

Examination and imaging studies

Neurologic examination revealed moderate gait ataxia, but no focal deficits. Magnetic resonance imaging (MRI) demonstrated a 4-cm mass with solid and cystic components filling the suprasellar region and causing considerable compression of the optic apparatus [Figure 1a, b]. The enhancing solid component originated in the suprasellar cistern, while the large cystic portion of the mass extended into the third ventricle superiorly. The lateral ventricles were enlarged bilaterally with evidence of transependymal flow of cerebral spinal fluid (CSF). A diagnosis of craniopharyngioma was considered very likely based on these imaging findings.

Operation

Shortly after his initial evaluation, the patient was taken for resection of the mass via a left-sided pterional craniotomy. Initial microdissection revealed two large-caliber vessels ascending between the optic nerves and coursing posteriorly over the chiasm [Figure 2a]. Further investigation demonstrated the vessels to be bilateral infraoptic A1 segments. Care was taken to preserve these

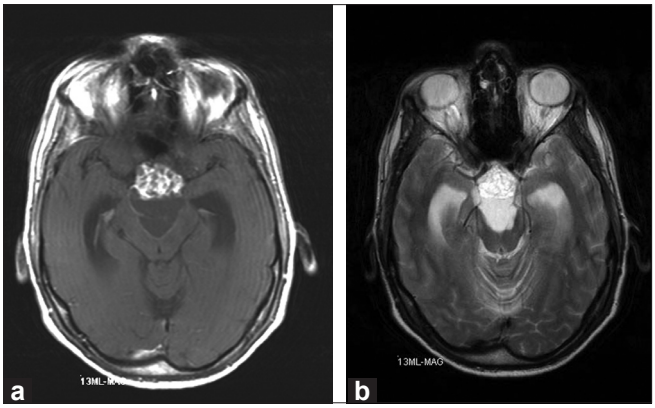

Figure 1: Preoperative (a) axial T1 post-contrast and (b) T2 images demonstrating a large suprasellar mass with a heterogeneously enhancing nodular component and enhancing cystic component. Marked ventriculomegaly of the temporal horns is evident, reflecting obstructive hydrocephalus secondary to the large tumor. The bilateral A1s are not well visualized due to the presence of the mass lesion

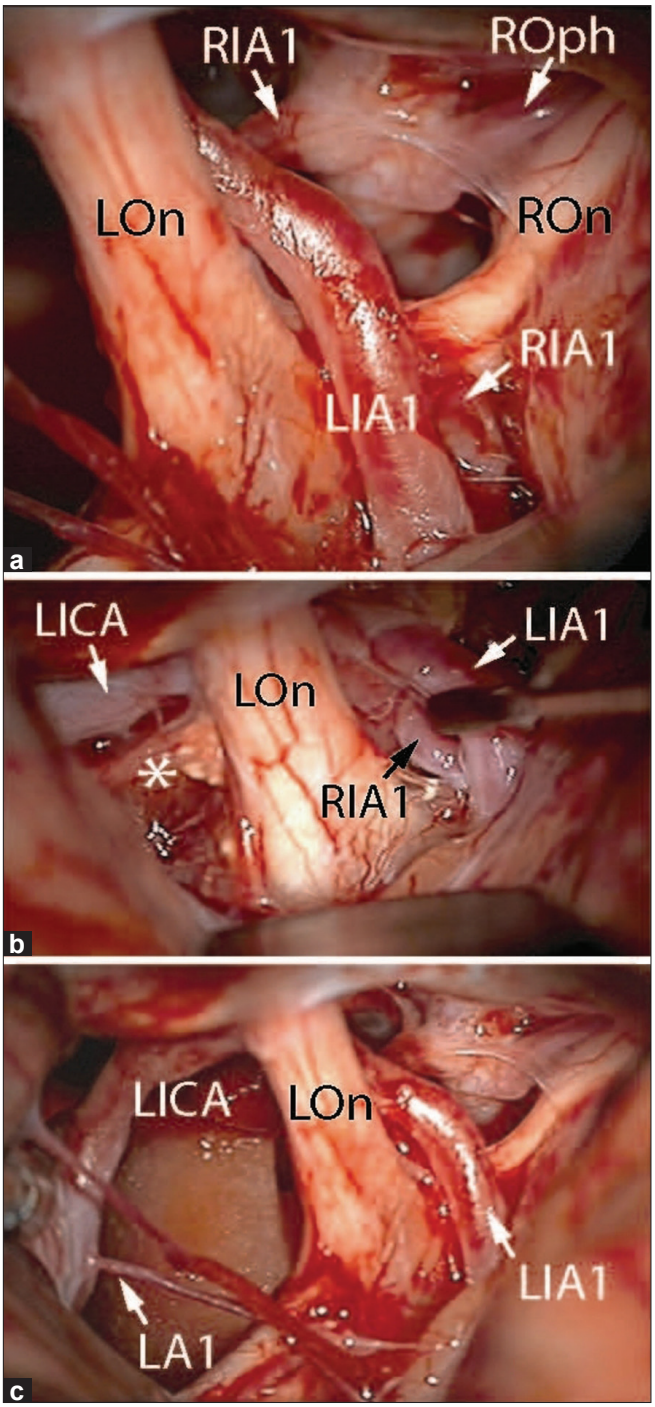

Figure 2: (a) View of the left-sided (*LIA1*) and right-sided (*RIA1*) infraoptic A1s coursing posteriorly in the pre-chiasmatic cistern and over the optic chiasm. The origin of the right infraoptic A1 and right ophthalmic artery (*ROph*) from the proximal right ICA are also visible. *LOn* and *ROn* signify left and right optic nerves, respectively. (b) A Rhoton dissector is utilized to elevate the left infraoptic A1 (*LIA1*) and reveal the right-sided infraoptic A1 (*RIA1*) in the pre-chiasmatic cistern. Calcified tumor (*) can be seen filling the optico-carotid cistern between the left internal carotid artery (*LICA*) and left optic nerve (*LOn*). (c) Lower magnification view of the left infraoptic A1 (*LIA1*) taking its origin from the proximal left ICA (*LICA*) and passing underneath the left optic nerve (*LOn*). The hypoplastic, supraoptic A1 (*LA1*) is also seen taking its origin from the left ICA in the more customary location before coursing above the optic apparatus toward the AComm complex (not pictured)

vessels, as well as the left-sided hypoplastic A1 branch that was found coursing over the chiasm [Figure 2b]. Intraoperative frozen section confirmed the diagnosis of craniopharyngioma, adamantinomatous type. Following gross total resection of the lesion, the infraoptic course of the A1s could be easily visualized [Figure 2c]. The anatomy of the region is illustrated in Figure 3.

Postoperative course

The patient remained neurologically stable throughout his hospitalization. He developed diabetes insipidus postoperatively which was controlled with vasopressin. Routine MR examination on the first postoperative day revealed image-complete resection of the craniopharyngioma, with evidence of resolving hydrocephalus. In the absence of the large mass, the A1 segments could be seen clearly taking their origin near the level of the OphA and passing beneath the optic nerves bilaterally [Figure 4a, b]. Because of the relatively strong association of cerebral aneurysms with an infraoptic A1 anomaly, an MR angiogram was obtained [Figure 5a, b]. This did not demonstrate the presence of any aneurysms.

DISCUSSION

The ACA typically arises as the medial component of the internal cerebral artery (ICA) bifurcation and courses over the superior surface of the optic chiasm (70%) or nerves (30%) before joining the AComm complex.[39] However, the ACA may be associated with a very proximal origin from the ICA as it passes through the internal dural ring. Commonly referred to as an infraoptic A1, this anomalous vessel bifurcates from the proximal ICA at or near the origin of the OphA before passing beneath the ipsilateral optic nerve and pursuing an interoptic course in the pre-chiasmatic cistern to reach the ACA–AComm complex.[10,30,32]

Surgically verified cases and cadaver studies alike have demonstrated that the infraoptic A1 is almost always accompanied by additional structural anomalies of the circle of Willis. The presence of a plexiform AComm artery,[20] agenesis/hypoplasia of the contralateral ICA,[35] and azygous pericallosal arteries[32] have all been reported. In the most commonly reported anatomic variation, the infraoptic A1 takes its origin from the right ICA and continues as a large-caliber vessel that directly supplies the distal ACA territories bilaterally, with associated hypoplasia or agenesis of the contralateral A1.[5,6,9,25,30,32,34] Closely correlated with this altered hemodynamic state is an overall increased frequency of cerebral aneurysms reported in association with the infraoptic A1, with the ACA–AComm complex representing the single most common site of aneurysm formation.[9,14,18,24,25,28-30,32,40]

Recently, Wong and colleagues proposed a classification scheme for the various anatomic configurations of the

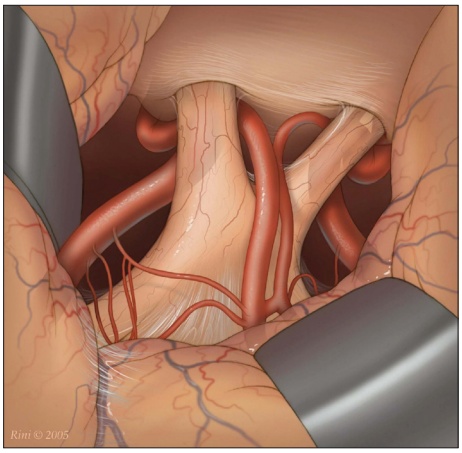

Figure 3: Illustration of the operative field after resection of the tumor demonstrating the course of the bilateral infraoptic A1 arteries

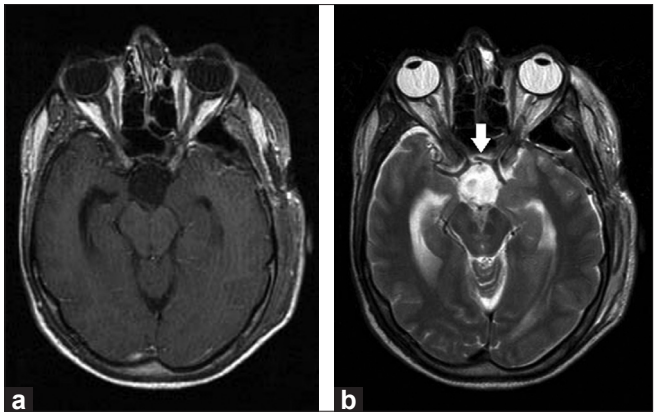

Figure 4: Postoperative (a) axial T1 post-contrast image demonstrating gross total resection of the tumor and (b) axial T2 image illustrating the bilateral infraoptic A1s (arrow) arising from the proximal ICA bilaterally

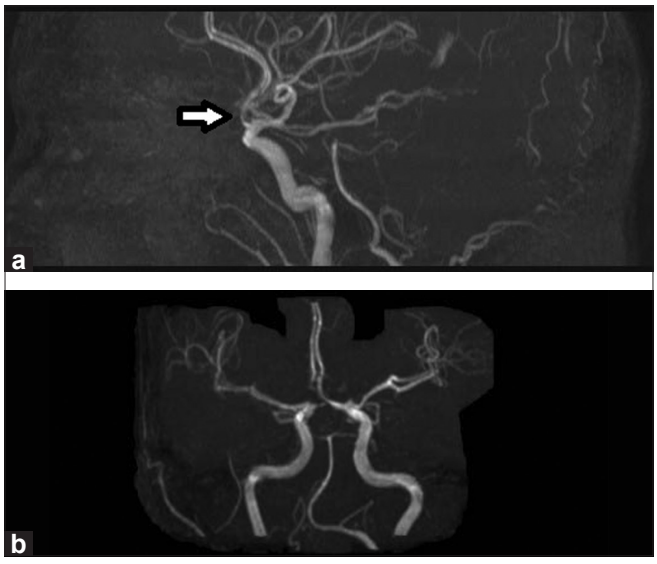

Figure 5: (a) Lateral and (b) A-P reconstruction of a 3D time-of-flight MR angiogram obtained postoperatively, revealing the hallmark appearance of the infraoptic A1s (arrow) taking a very proximal origin off the bilateral ICAs just distal to the cavernous segment

infraoptic A1.[37] Drawing upon all previously reported cases of infraoptic A1, four distinct classes of the anomaly were identified based on the presence or absence of an ipsilateral supraoptic A1 and contralateral A1. Based upon this scheme, the case described here would be classified as a bilateral type 2 infraoptic A1, of which only six cases have been previously described.[13,16,18,21,26,37] Interestingly, an exhaustive literature search failed to identify any previous cases of bilateral infraoptic A1 arteries associated with a suprasellar neoplastic lesion.

Although the infraoptic A1 seemingly functions in the capacity of a "normal" proximal ACA, it is often associated with an additional ipsilateral hypoplastic A1 segment taking origin from the ICA in the more usual location and pursuing a more characteristic supraoptic course.[20,24,31,35] Cases reporting the presence of this additional supraoptic A1 branch consistently document that the associated infraoptic A1 maintains hemodynamic predominance; indeed, the hypoplastic supraoptic A1 is often not readily visible by digital subtraction angiography. Because these supraoptic A1 and infraoptic A1 segments exist together in many cases, some authors have postulated that the infraoptic A1 is not simply a misplaced A1 segment, but rather a persistent embryological vessel. They prefer to describe the infraoptic A1 as an anomalous "carotid–anterior cerebral artery anastomosis" to more accurately reflect the origin of this anatomic variant.[24,25] However, the exact embryological basis of this anomaly remains unknown. Current theories regarding its embryogenesis include a persistent embryonic anastomosis between the primitive maxillary artery and the ACA,[6] as well as error in the development of the definitive OphA,[13] and have been reviewed in detail previously.[24,25,32]

The infraoptic A1 has a characteristic appearance on magnetic resonance angiography (MRA) [Figure 5a, b]. Lateral views reveal a rather robust vessel taking its origin off the ICA just as it becomes intradural, near the level of the OphA, and offer perhaps the most easily appreciable and reliable view of the infraoptic A1 [Figure 4a]. Some reports describe the infraoptic A1 sharing a common origin with the OphA, a fact which should be considered when interpreting preoperative imaging studies.[32] A-P and oblique projections are less ideally suited to recognize an infraoptic A1; yet, on close inspection, they do demonstrate the abnormally low, medial course of the proximal ACA required for it to pass under the ipsilateral optic nerve before ascending in the prechiasmatic cistern [Figure 5b]. However, the precise relationship of the proximal A1 to the optic nerve can be difficult to appreciate with angiography or MRA alone. In fact, the majority of the surgically verified cases in the literature describe the infraoptic A1 as an unexpected intraoperative finding not initially appreciated on preoperative studies.[19,25] MRI can be particularly helpful in demonstrating the ACA's spatial relationship with the optic nerves and chiasm in such cases, and as such it should be considered as an adjunct imaging study to help facilitate diagnosis when anatomic relationships are unclear.[10] Source images from MRA as well as routine T_2-weighted MR images can be used to reliably visualize the A1's course and relationship to the optic apparatus.[10] In addition, MRI may suggest vascular compression of the optic nerve or chiasm by the infraoptic A1 in patients experiencing visual symptoms, as has been reported previously.[30]

The case presented here is unique in that it documents the first report of bilateral infraoptic A1s in association with a suprasellar tumor. Interestingly, we discovered and recognized this rare anomaly during surgery as it was not appreciated on preoperative MR imaging. Looking retrospectively at the preoperative images, neither of the infraoptic A1s is readily visible as they were both displaced and effectively concealed by the large craniopharyngioma. However, both infraoptic vessels can be easily appreciated on routine MRI sequences following resection of the tumor [Figure 4b]. Recognition of the anomalous vessels and proper interpretation of the vascular anatomy at the time of surgery were critical in the case. In the setting of a large, complex mass lesion distorting local anatomy and obstructing the surgeon's view, it is possible that one or both of the infraoptic A1s could have been inadvertently injured or even erroneously sacrificed.

CONCLUSIONS

While extensive variability in the microsurgical anatomy of the ACA–AComm complex is certainly the rule and not the exception, the infraoptic A1 is a rare variation of the anterior circulation with important clinical and surgical implications for neurosurgeons operating in the region. The infraoptic A1 may be unilateral or bilateral and may be associated with sellar/suprasellar neoplasms or aneurysms of the anterior circulation. Awareness of the existence and clinical significance of this unique anomaly can help facilitate its recognition, thereby allowing neurosurgeons to adjust operative planning accordingly.

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