

Coincident craniopharyngioma and unruptured aneurysm at the right M1- M2 segment of the middle cerebral artery. Case report

Craniofaringioma coincidente com aneurisma não-roto no segmento M1-M2 à direita da artéria cerebral média. Relato de caso

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ABSTRACT

We report a case of coincident craniopharyngioma with an unruptured aneurysm at the right M1- M2 segment of the middle cerebral artery; and discuss the surgical approach to two different pathologies treated at same surgical time, as well as current theories to explain the coincidence or association between tumor and vascular lesions at the sellar and suprasellar regions. The patient was a 59-year-old woman; with a history of visual impairment and headache. Both lesions were identified by magnetic resonance imaging (MRI) and cerebral angiography, and were treated surgically. We suggest individualized treatment for each case, choosing the best neurosurgical approach to achieve an appropriate treatment of both pathologies in the same operative session.

Key-words: Craniopharyngiomas; Middle cerebral artery aneurysms; Supracaloid aneurysms

RESUMO

Relatamos um caso de craniofaringioma com aneurisma não roto no segmento M1-M2 da artéria cerebral média à direita; e discutimos a abordagem cirúrgica a duas patologias diferentes, porém tratadas no mesmo tempo cirúrgico, assim também como as teorias atuais para explicar a coincidência ou associação entre os tumores e as lesões vasculares na região selar e supresselar. Paciente mulher de 59 anos, com história de deficiência visual e cefaleia. Ambas as lesões foram identificadas, por ressonância magnética (RM) e angiografia cerebral, e tratadas cirurgicamente. Sugerimos tratamento individualizado para cada caso, escolhendo a melhor abordagem neurocirúrgica para o tratamento adequado de ambas patologias no mesmo ato cirúrgico.

Palavras-chave: Craniofaringiomas; Aneurismas da artéria cerebral média; Aneurismas supracaloides

INTRODUCTION

Craniopharyngiomas are benign tumors that occur at the skull base, above the pituitary gland and behind the optic chiasm, in the suprasellar area.

Craniopharyngiomas derive from the Rathke cleft rather than the squamous cell crests along the craniopharyngeal duct and are classified as World Health Organization (WHO) grade I. These tumors, may affect people of any age and have two

peaks of incidence: in children aged 5-14 years old and in adults aged over 50-60 years old¹. These tumors have a similar incidence in both males and females. There are two subtypes of craniopharyngiomas: adamantinomatous (found in pediatric and adult patients) and papillary (found only in adults)².

The vascular supply of the tumor is through the anterior circulation; via small perforating branches from the A1 segment; of the anterior cerebral artery; and the intracavernous meningohypophyseal arteries branches, without a direct vascular supply of the tumor from the M1-M2 segment of the

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middle cerebral artery. The craniopharyngioma and the middle cerebral artery do not have any direct vascular relationship. Therefore, the exact pathophysiology of the coincident craniopharyngioma and the unruptured aneurysm that we described here remains unknown.

CASE REPORT

A 59-year-old woman had presented with progressive binasal visual field loss, turbidity, frontal headache with an intensity of 5/10, phonophobia and photophobia, which worsened with exertion for the prior 2 years. Three months earlier, the clinical status of the patient worsened in frequency and intensity, but without a loss of awareness, nausea or vomiting, motor or sensitivity deficits or other signs of neurological alarm. Her past medical and surgical history was: high blood pressure, non-insulin-dependent diabetes mellitus, appendectomy, uterine myomas (with a hysterectomy 5 years ago), and no relevant nonmedical personal or social history. Family history: her brother and sister had ruptured cerebral aneurysms previously, but there was no association with polycystic kidney disease. The neurological examination revealed that the patient presented with binasal hemianopsia, a decline in visual acuity to 20/200, a preoperative mini-mental state examination score of 24/30, and no other neurological deficits. Magnetic resonance imaging (MRI) revealed a suprasellar mass with high intensity on T1 (Figure 1) and on the T2-weighted sequence the solid portion was heterogeneous, whereas the cystic part was hyperintense, measuring approximately 3.0 mm × 2.5 mm × 2.0 mm (Figure 2). MRI and Angio-resonance also showed a flow void mass at the right Sylvian fissure in the way of the middle cerebral artery (Figures 3, 4) and due to this factor and the previous family history, a cerebral angiography was performed and confirmed the presence of a saccular aneurysm (measuring approximately 5.0 mm × 3.5 mm at the neck) (Figure 5 a-c). Laboratory tests for hormone levels were normal. The patient was operated on using the fronto-temporo-orbito-zygomatic approach (FTOZa). The first step was the clipping of the middle cerebral artery aneurysm, and the second step was to perform the resection of the suprasellar tumor. The patient's fluid and electrolyte levels were closely monitored postoperatively; these parameters stayed within the normal ranges, and no additional drug treatment was necessary.



Figure 1. Sagittal view of T1-weighted magnetic resonance imaging (MRI) showing a suprasellar mass with high intensity.

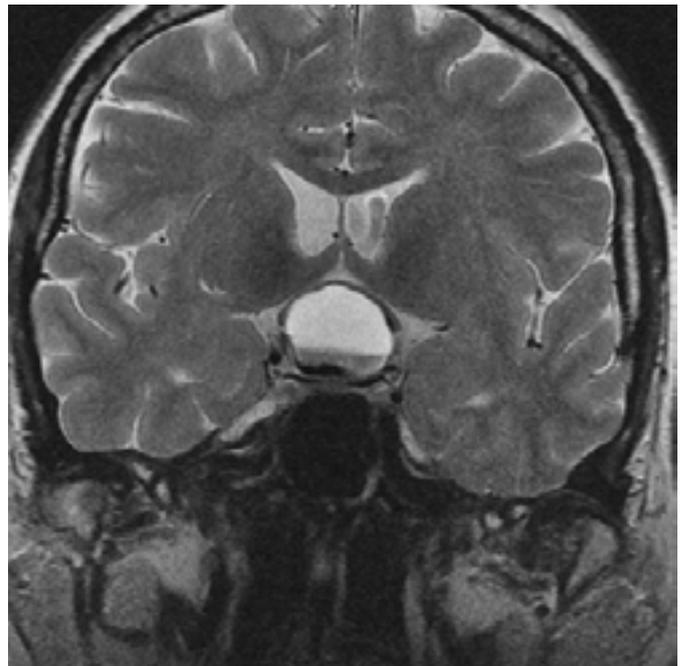


Figure 2. Coronal view of a T2-weighted sequence showing a solid portion with heterogeneous image, whereas the cystic part was hyperintense.

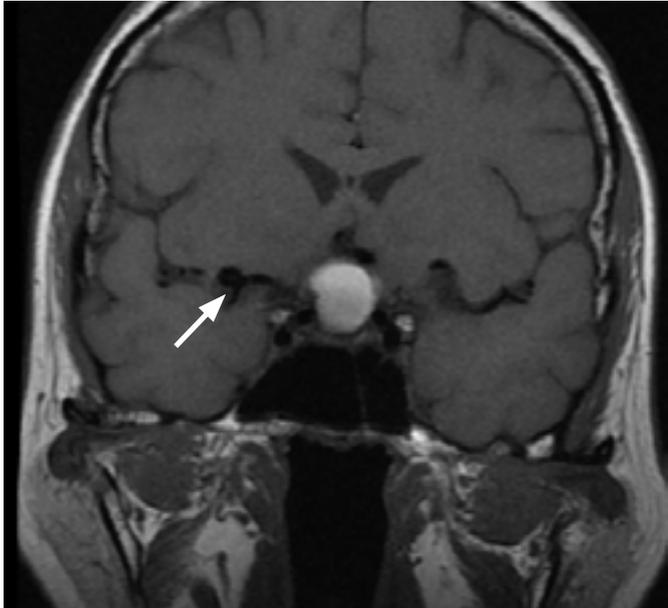


Figure 3. Coronal view of MRI showing a flow void mass at the right Sylvian fissure in the way of the middle cerebral artery (arrow).



Figure 4. Angio-resonance showing a suprasellar mass (white arrowhead) and flow void mass at the right Sylvian fissure in the way of the middle cerebral artery (white arrow).

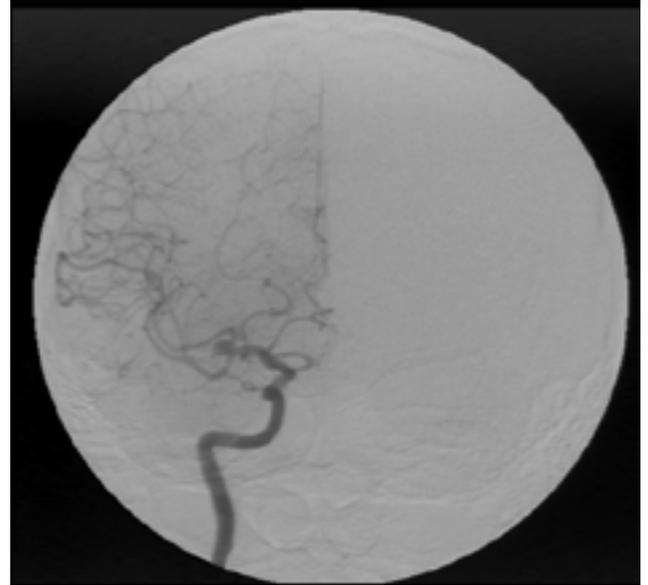


Figure 5 (a). Frontal view of the right internal carotid angiogram showing a saccular-type aneurysm at the M1-M2 middle cerebral artery.

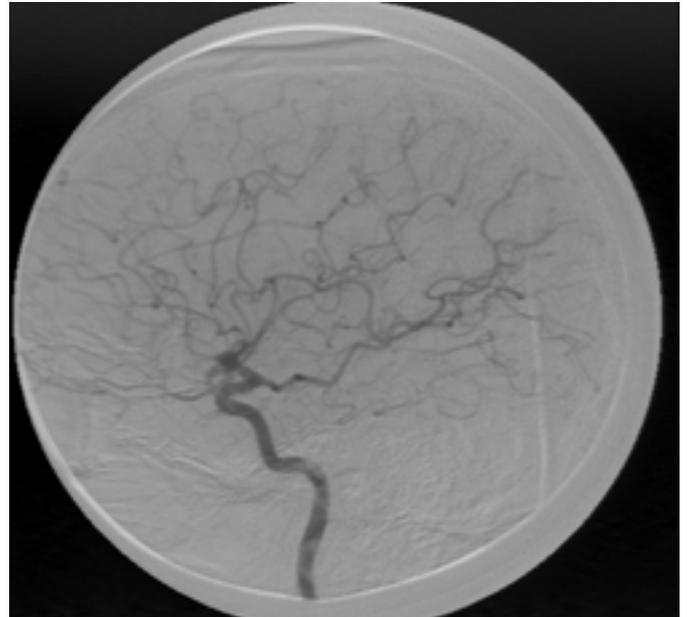


Figure 5 (b). Lateral view of the right internal carotid angiogram showing a saccular-type aneurysm at the M1-M2 middle cerebral artery.

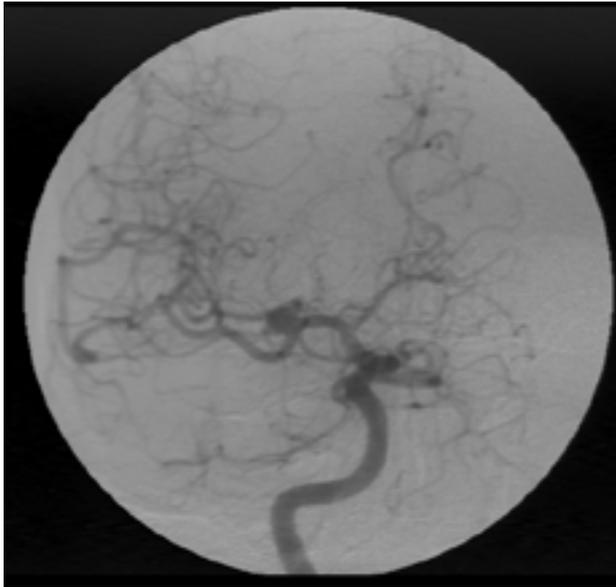


Figure 5 (c). Oblique view of the right internal carotid angiogram showing a saccular-type aneurysm at the M1-M2 Middle cerebral artery.

DISCUSSION

We present an uncommon coincident association between a craniopharyngioma without pituitary dysfunction and a right middle cerebral artery aneurysm (M1-M2 segment). The patient was operated on using the FTOZa. We clipped the middle cerebral artery aneurysm and then resected the suprasellar tumor.

Currently, several approaches for the removal of craniopharyngiomas have been proposed, including pterional, subfrontal, subfrontopterional³, bifrontal interhemispheric through the lamina terminalis⁴, and more recently transsphenoidal⁵.

Due to the hidden and deep position of retrochiasmatic craniopharyngiomas behind the optic chiasm with an upward extension into the third ventricle, surgical exposure of these tumors requires an approach from the inferior to superior and the anterior to posterior directions. Since our patient had an unruptured aneurysm at the right middle cerebral artery, the senior surgeon chose the FTOZ approach. This route provides access to the orbit, the anterior cranial base, the suprasellar area, the cavernous sinus, the anterior communicating artery, the lateral fronto-temporal region and the Sylvian fissure, allowing a wide exposure of the surgical area. Moreover, we

were able to expose both the bifurcation of the middle cerebral artery and the suprasellar surface without unnecessary brain retraction.

After searching the neurosurgical literature, the authors found some case reports describing the relationship among pituitary adenomas and aneurysms involving the supraclinoid segment and the anterior and posterior communicating segments of the internal carotid artery⁶⁻⁸. Several theories have been proposed to explain the development of the aforementioned association. These theories suggest three possible mechanisms: a mechanical stimulus (increased the blood flow to supply the tumor), microcirculatory alterations and hormonal factors (high levels of growth hormone)⁹. However, the presence of a craniopharyngioma and a middle cerebral artery aneurysm have not been linked, and, to our knowledge, no case was reported in the literature.

CONCLUSION

We described a very interesting and rare case. The theories of the coincidence or association of these two lesions remain a matter of debate. We suggest an individualized treatment for each case, choosing the best neurosurgical approach to achieve the appropriate treatment of both pathologies in the same operative session.

REFERENCES

1. Larkin, S.J., Ansorge, O. Pathology and pathogenesis of craniopharyngiomas. *Pituitary*. 2013;16(1):9-17. doi: 10.1007/s11102-012-0418-4.
2. Osborn AG. *Osborn's Brain: Imaging, Pathology, and Anatomy*. Wolters Kluwer Lippincott Williams & Wilkins; 2013; pg 707
3. Ammirati M, Samii M, Sephernia A. Surgery of large retrochiasmatic, craniopharyngiomas in children. *Childs Nerv Syst*. 1990;6(1):13-17
4. Kanno T, Kasama A, Shoda M, Yamaguchi C, Kato Y. A pitfall in interhemispheric translamina terminalis approach for the removal of a craniopharyngioma. Significance of preserving draining veins. Part I. Clinical study. *Surg Neurol* 1989;32:111-115 doi: 10.1016/0090-3019(89)90197-3
5. Liu JK, Christiano LD, Gupta G, Carmel PW. Surgical nuances for removal of retrochiasmatic craniopharyngiomas via the transbasal subfrontal translamina terminalis approach. *Neurosurg*

Focus. 2010 Apr;28(4):E6. doi: 10.3171/2010.1.FOCUS09309

6. Bulsara KR, Karavadia SS, Powers CJ, Paullus WC. Association between pituitary adenomas and intracranial aneurysms: An illustrative case and review of the literature. *Neurol India* 2007;55:410-412
7. Almeida Silva JM, Campos RR, Souza RR, Sette Dos Santos ME, Aguiar GB. Spontaneous subarachnoid haemorrhage from rupture of an anterior communicating artery aneurysm in a patient with pituitary macroadenoma. *Neurocirurgia (Astur)*. 2014 Mar-Apr;25(2):81-5. doi: 10.1016/j.neucir.2013.03.005
8. Pritz MB (2002) Ruptured true posterior communicating artery aneurysm and cystic craniopharyngioma. *Acta Neurochir (Wien)*. 2002;144(9):937-9; doi: 10.1007/s00701-002-0943-4
9. Acqui M, Ferrante L, Fraioli B, et al. Association between intracranial aneurysms and pituitary adenomas. Aetiopathogenetic hypotheses. *Neurochirurgia (Stuttg)*, 1987,30(6):177-181. doi: 10.1055/s-2008-1054091

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