Multiple Cavernous Hemangiomas in the Orbit. A Case Report

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¹Kaunas Medical University Hospital, Department of Neurosurgery ²Department of Ophthalmology, Kaunas, Lithuania Cavernous hemangiomas are relatively common benign lesions in the orbit producing proptosis and more rarely visual disturbances in adults. However, multiple lesions diagnosed as cavernous hemangiomas in the orbit have been described extremely rare in the ophthalmological and neurosurgical literature. A case of multiple cavernous hemangiomas appearing in a single orbit 19 years after removal of one cavernous hemangioma in the same orbit is analyzed. Diagnosis was determined by CT examination that revealed multiple lesions located superiorly, laterally, and medialy in the left orbit, displacing the optic nerve inferiorly and producing bulging of the medial wall of the orbit. The microsurgical left lateral orbitotomy approach was used. All five oval-shape lesions were totally removed employing sharp microdissection not damaging the optic nerve sheet. The patient recovered uneventfully, and a relatively good cosmetic result was achieved. Pathological examination revealed that all five lesions were cavernous hemangiomas. No disturbance of eye movements was detected in a month time. We suggest lateral orbitotomy approach in large multiple cavernous hemangiomas affecting one orbit.

Key words: cavernous hemangioma, orbital tumor, lateral approach

INTRODUCTION

Cavernous hemangiomas are relatively common benign lesions in the orbit, producing proptosis and more rarely visual disturbances in adults (1–7). However, multiple lesions diagnosed as cavernous hemangiomas in the orbit have been described extremely rarely in the ophthalmological and neurosurgical literature (8, 1, 2, 9, 5, 10). We describe one case of five cavernous hemangiomas appearing in a single orbit 19 years after removal of one cavernous hemangioma in the same orbit.

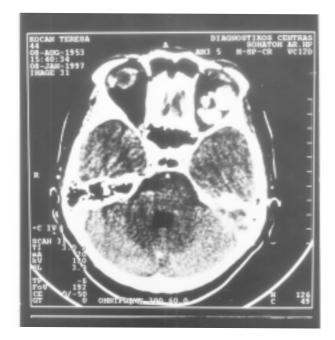
CASE REPORT

A 44-year-old female was admitted to our hospital with a 0.5-year history of impaired visual acuity and proptosis of the left eye. She had a past medical history of cavernous hemangioma in the left orbit, removed by ophthalmologist in another institution through the lateral transconjuctival approach 19

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years ago. Examination on this admission revealed impaired visual acuity of her left eye (20/400), concentric reduction of her left visual field and a marked relative proptosis (6 mm) of her left eye. Lateral movements of the left eye were also slightly impaired. Atrophy of the left optic disc was found during fundoscopy. No other neurological deficit was observed on admission. CT (computer tomography) examination of the orbits revealed multiple lesions located superiorly, laterally, and medially in the left orbit, displacing the optic nerve inferiorly and producing bulging of the medial wall of the orbit. Those lesions homogeneously enhanced suggesting multiple cavernous hemangiomas (Fig. 1).

The microsurgical left lateral orbitotomy approach was applied. Removal of the lateral wall of the orbit was performed through a 3-cm linear skin incision without damaging the external canthal ligament. Incision of the periorbit above the lateral rectus muscle exposed orbital fat and one blue-red encapsulated mass lesion. All five oval-shape lesions were removed one by one with a gentle retraction of the lateral rectus muscle inferiorly and using blunt the microdissecting technique (Fig. 2). No significant bleeding was noticed during tumor removal.



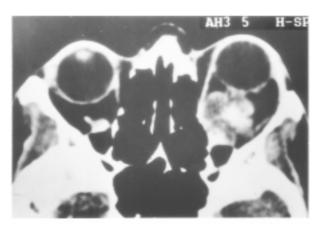


Fig. 1. Preoperative CT scan demonstrates homogeneously enhanced multiple lesions in the left orbit (a) and magnified CT showing the relation of the lesion to the medial compartment of the orbit (b)

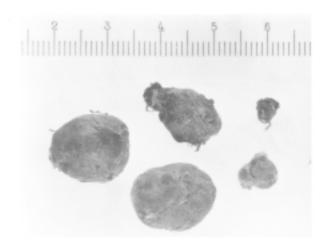


Fig. 2. The specimen of five cavernous hemangiomas after removal. Ruler's smallest gradation 1 mm

One lesion was adherent tightly to the anterior superior compartment of the optic nerve. It was totally removed by sharp microdissection sparing the optic nerve sheet. The skin incision was closed using intracutaneous 6–0 absorbable suture. The patient recovered uneventfully, and a relatively good cosmetic result was achieved. Pathological examination showed that all 5 lesions were cavernous hemangiomas (Fig. 3). CT showed slightly edematous lateral and medial rectus muscles one week after surgery (Fig. 4). One month after operation her visual acuity remained at the preoperative level (20/400), the visual field improved to normal and proptosis disappeared. No disturbance of eye movements was detected in a month time.

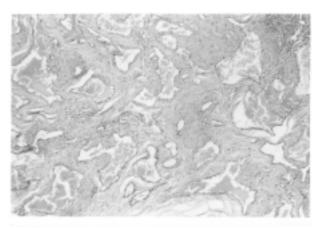


Fig. 3. Photomicrograph of cavernous hemangioma showing large irregular venous channels containing blood. H & E, \times 60

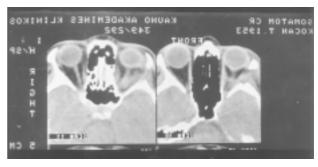


Fig. 4. CT scans 1 week after operation demonstrate slight swelling of lateral and medial rectus muscles and removed lateral wall of the left orbit (Pre- and postoperative scans are made by different CT scanners. For better comparison we put postoperative scan in inverse position)

DISCUSSION

Multiplicity of cavernous hemangiomas is an extremely rare presentation of those benign intraorbital lesions. Only seven cases are described in the lite-

rature available for us (8, 1, 2, 9, 5, 10). Four patients had two separate lesions in one orbit (1, 2, 5, 10). Another two patients had bilateral lesions in both orbits (8, 9). As far as we know, only one case of 5 separate cavernous hemangiomas in one orbit has been reported (1). Pertuiset (11) found 30% of multiple lesions in 48 cases of orbital angiomas, however, it was questioned by others, suggesting most of these lesions to be lymphangiomas (1, 10). Analysis of cases of verified multiple cavernous hemangiomas revealed female predominance 5 to 1 (in one case (8) gender is not stated) similar to single cavernous hemangiomas (8, 12, 1, 2, 13), supporting the theory that female hormones may play a role in the pathogenesis of this disease (9, 14). There are reports that even after partial removal of cavernous hemangiomas there have been no recurrences in ten to eighteen years (1, 15). In our case the patient developed five cavernous hemangiomas 19 years after removal of one lesion. It could not be attributed to regrowth of partially removed one cavernous hemangioma because of the multiplicity of the lesions. We think our patient developed five new lesions after removal of one. It could support the theory by Harris et al. that cavernous hemangiomas are acquired lesions (1). In our case the lesions were removed via lateral orbitotomy approach (16-18). It is still some debate among neurosurgeons and ophthalmologists as to the approach to cavernous hemangiomas located in orbit. Many neurosurgeons still prefer craniotomy (12, 19, 20, 13) while ophthalmologists favor various modifications of orbitotomies (1, 21, 2, 22). At present we prefer the lateral orbitotomy approach even for big cavernous hemangiomas (4 out of 5 cases in the last two years, unpublished data) because of fewer complications as compared to craniotomy. We suggest to employ the lateral orbitotomy approach in multiple cavernous hemangiomas affecting one orbit on the basis of our case. Even if the lesions are big in size when joined together, they are easily removed one by one even from the medial compartment of the orbit and proximal conus. Because the working space increases after removal of each subsequent lesion, it is possible to reach the less accessible areas. Also, when removing multiple lesions it does not produce significant bleeding, as it usually happens in single large lesion when removed piecemeal (20).

CONCLUSION

We report a rare case of multiple cavernous hemangiomas in a single orbit 19 years after removal of one cavernous hemangioma. We suggest a lateral orbitotomy approach in large multiple cavernous hemangiomas affecting one orbit.

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DAUGYBINĖ AKIDUOBĖS KAVERNINĖ HEMANGIOMA: KLINIKINIS ATVEJIS

Santrauka

Kaverninės hemangiomos yra dažna gėrybinė liga, pasireiškianti išverstakumu, rečiau – sutrikusiu matymu. Vis dėlto daugybinės akiduobės kaverninės hemangiomos yra labai reta liga, aprašoma oftalmologinėje bei neurochirurginėje literatūroje. Straipsnyje analizuojamas daugybinės recidyvavusios akiduobės kaverninės hemangiomos klinikinis atvejis. Diagnozė buvo nustatyta remiantis galvos kompiuterine tomografija, radus daugybinius ribotus židinius viršutinėje, išorinėje, vidinėje orbitos dalyje. Navikas buvo operuotas atliekant šoninę mikrochirurginę orbitotomiją. Pašalinti visi penki navikiniai mazgai. Po operacijos stebėtas neblogas kosmetinis efektas. Histologiškai patvirtintos penkios kaverninės hemangiomos. Akies judesiai visiškai atsistatė po mėnesio. Mes siūlome naudoti šoninę mikrochirurginę orbitotomiją šalinant akiduobės daugybines hemangiomas.

Raktažodžiai: kaverninė hemangioma, orbitos navikas, šoninė orbitotomija