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ILLUSTRATIVE CASE OF CAPILLARY HEMANGIOMA OF THE OPTIC NERVE

Capillary hemangiomas (CH) are benign proliferative vascular neoplasms commonly present in skin and soft tissues but rarely found intracranially. We describe a case of a 14-year-old male patient with histologically proven CH of the optic nerve, who underwent surgical resection of the lesion due to progressive visual loss. Imaging studies revealed a cystic-solid formation in the suprasellar region lateralized to the right and located above the pituitary gland. This case shows that CH may originate from the optic nerve, leading to its gradual compression and causing optic neuropathy. While the correct differential diagnosis on the MRI may be difficult, surgical treatment is warranted in cases of progressive visual decline.

Keywords: capillary hemangioma, optic nerve, optic nerve chiasm, surgical removal.

Capillary hemangiomas (CH) are benign vascular neoplasms, often found on skin and well known as infantile hemangiomas or strawberry birthmarks [1, 2]. They are usually diagnosed at birth and may exhibit rapid growth during the first year of life [1, 3]. Most CH regress and disappear spontaneously before reaching puberty. However, there are only anecdotal cases in the literature describing the intracranial CH (ICH) [3—6].

An accurate assessment of the true prevalence of CHs in the central nervous system is challenging because their maximum size is typically observed by 6 months of age, with subsequent tumor regression [1, 2, 7]. According to the literature, spontaneous involution of hemangiomas occurs in 30% of patients before the age of 4 and in 80% before the age of 7 [1]. The average prevalence of CH among newborns ranges from 0.05% to 5% [3, 8, 9]. However, most lesions are found on the skin surface, with visceral, intraspinal, and intracranial hemangiomas described in sporadic cases [6, 7, 10, 11]. These lesions are often diagnosed at birth and may grow rapidly in the first year of life [1]. Most hemangiomas regress spontaneously before puberty, distinguishing them from capillary malformations, which grow slowly and persist throughout life [3].

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Clinical features of CH of the optic nerve may include progressive visual loss, proptosis, and, in some cases, local pain due to pressure on surrounding structures. Importantly, the so-called CH of the optic nerve head, or juxtapapillary CH are morphologically distinct from the true CH. They are often described in the context of the von Hippel — Lindau disease and represent hemangioblastomas [12—14]. In children, the initial signs of the juxtapapillary hemangiomas often include decreased vision in one eye and strabismus [12, 15].

Hemangiomas are classified as benign vascular tumors, differentiating them from vascular malformations [16—18]. Structurally, they can be classified as a capillary, cavernous, or mixed type. CH typically consists of densely packed neoplastic capillaries separated by a thin layer of connective tissue [16, 17].

Furthermore, CH of the optic nerve requires careful histological and radiological differential diagnosis with other lesions affecting the optic nerve, such as optic nerve gliomas, meningiomas, cavernous hemangiomas, arteriovenous malformations, as well as other lesions of the optic nerve apparatus and optic chiasm [13, 14, 19, 20].

Considering the rarity of the condition, there is no consensus regarding the optimal treatment strategy for ICH [2— 4, 15, 21]. Surgical treatment is described in many cases with symptomatic lesions [2, 5, 6, 22, 23]. We describe a unique case of a successful surgical treatment of a patient with histologically confirmed CH of the optic nerve.

Case report

The study was performed according to the ethical requirements of the 1964 Helsinki declaration, International Conference of Harmonization and Good Clinical Practice (1996), EU Directive No. 609 (from April 24, 1986), and the applicable orders of the Ministry of Health of Ukraine. The Ethics Committee of the State Institution "Romodanov Neurosurgery Institute" of the National Academy of Medical Sciences of Ukraine detected no violations of ethical, moral, and legal standards during the research and approved it. Informed consent was obtained from the patient and his parents prior to publishing this report

A 14-year-old male patient presented to our department with complaints of decreased vision in the right eye, a sensation of a black spot in front of the right eye, narrowing of the temporal visual field in the left eye, and headache. These symptoms had developed suddenly two weeks before admission.

MRI scans revealed a partly cystic, partly solid suprasellar lesion lateralized to the right and located above the pituitary gland, measuring 1.67 × × 1.8 × 1.48 cm (Fig. 1).

It showed a heterogeneous signal intensity on the T2-weighted images and was unevenly hyperintense on the T1-weighted and FLAIR images, with no change in signal characteristics after contrast administration. The cerebral CT-angiography excluded an arterial aneurysm. The lesion exhibited a density of +49 to +59 Hounsfield units in the suprasellar region and was lateralized to the right (Fig. 2).

To rule out an embryonic tumor, blood and cerebrospinal fluid samples were tested for alpha-fetoprotein and human chorionic gonadotropin, both of which were within the normal range.

Ophthalmological examination revealed a reduced visual acuity in the right eye (20/200) compared to the normal vision in the left eye (20/20). Visual field testing demonstrated a large central scotoma in the right eye with temporal deviation and temporal hemianopia in the left eye (Fig. 3). Fundoscopy showed pale pink optic nerve discs with clear margins and narrowed vessels, consistent with the chiasmatic syndrome.

Due to the unclear nature of the lesion, we postulated it to be either an atypical optic glioma with hemorrhagic foci or, less probably, a craniopharyngioma. Given the progressive visual deterioration due to the space-occupying lesion in the optic chiasm, and following an interdisciplinary discussion, we opted for surgical treatment through the endoscopic endonasal approach.

After the incision in the nasal mucosa and opening of the sellar floor, suprasellar cisterns above the tuberculum sellae were exposed. During subsequent arachnoid dissection, the significantly enlarged right optic nerve was found (Fig. 4).

A small incision was made in the altered area (area of discoloration), and the cyst, with a volume of up to 0.5 ml, was drained. A gross total resection of all abnormal tissue from the walls of the cystic cavity was performed. Upon examining the cavity with an angled endoscope, a lesion resembling a cavernous malformation — a node with hemorrhagic changes — was identified and completely re-

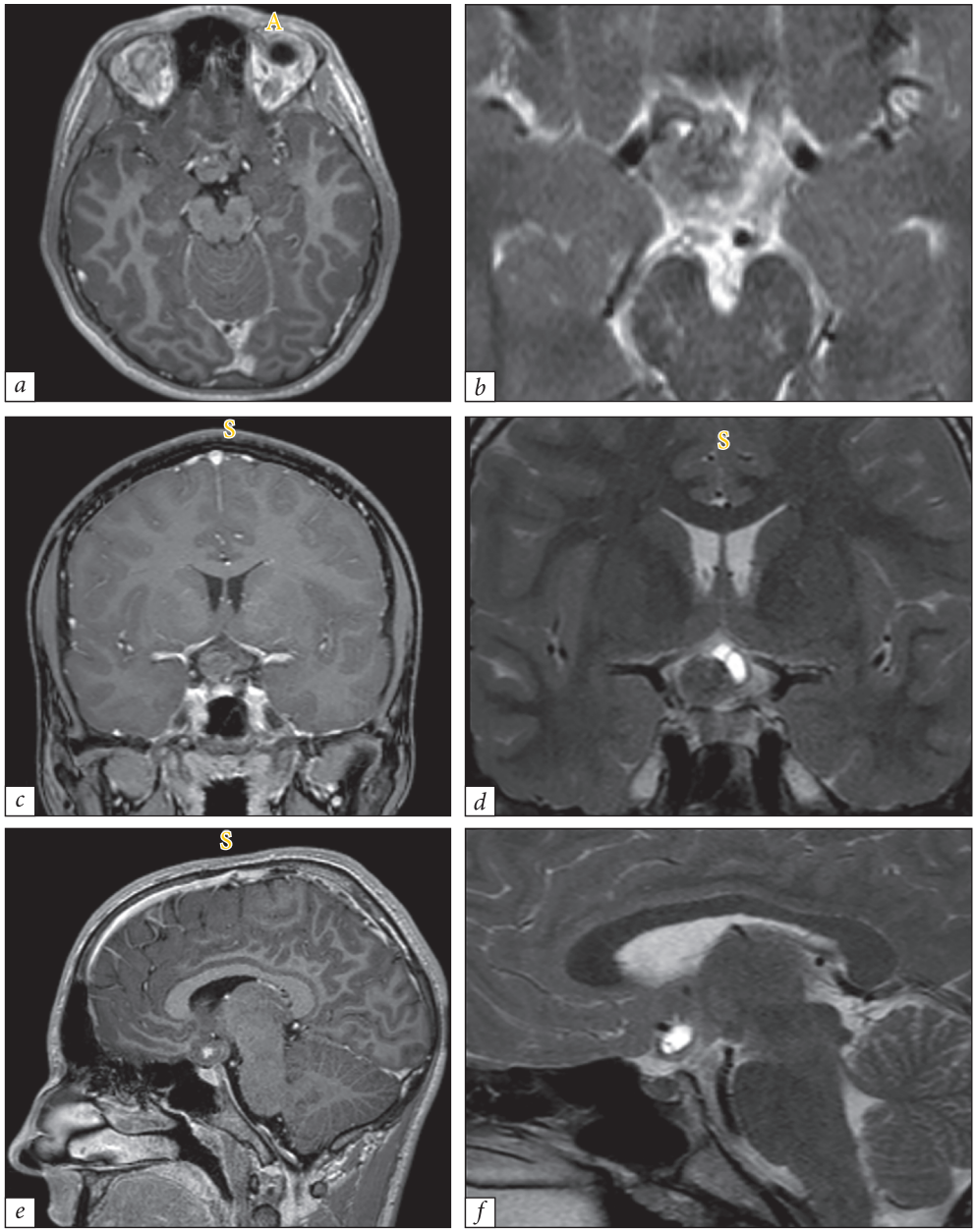

Fig. 1. Characteristics of capillary hemangiomas. There is a heterogeneous signal on T2-weighted images (right column, *b*, *d*, *f*), which remains unchanged after contrast administration, and a heterogeneous hyperintense signal on T1-weighted images (left column, *a*, *c*, *e*; white arrowheads). From above to below, *a* and *b* represent an axial, *c* and *d* — coronal, *e* and *f* — sagittal view, respectively

moved. The sellar defect was reconstructed using a multilayered closure with a gentamicin-infused sponge, oxidized cellulose preparations, artificial dura, fascia lata, and nasoseptal flap.

In the early postoperative period, the patient experienced subjective worsening of the visual function in the right eye. Laboratory monitoring showed the normal levels of pituitary hormones.

A postoperative ophthalmological examination before discharge revealed unchanged visual acuity in both eyes. Additionally, a slight enlargement of the visual field defect in the right eye was noted, while the defect size decreased in the left eye (Fig. 5, *a, b*). The fundoscopic picture was unchanged.

The postoperative MRI confirmed gross total resection of the lesion (Fig. 6).

The pathology report indicated morphological features consistent with CH, accompanied by reactive gliosis, thrombosis, and both fresh and old hemorrhages (Fig. 7).

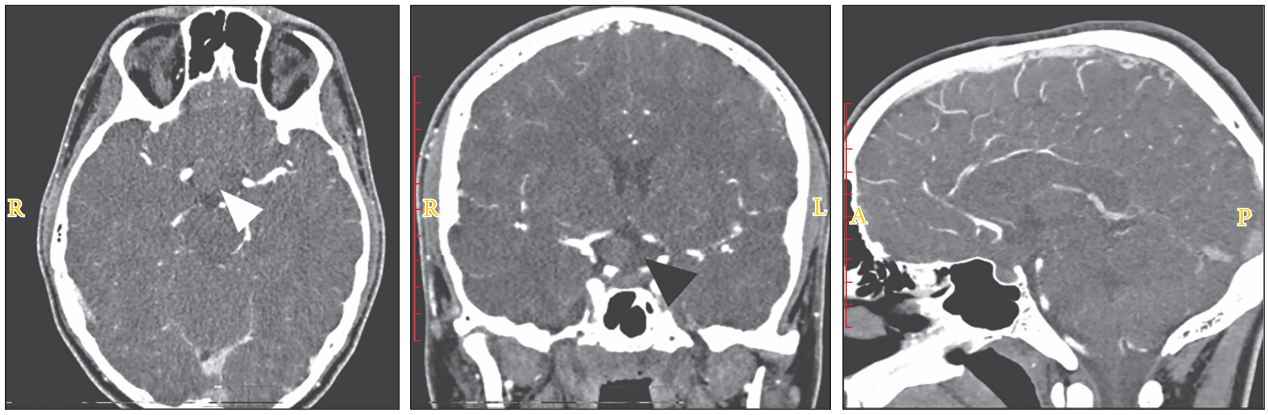

Fig. 2. CT angiography shows a suprasellar lesion with a density of +49 to +59 Hounsfield units lateralized to the right (white and black arrowheads, respectively)

Molecular analysis identified non-neoplastic glial tissue in the initial specimen, along with vascular proliferation and thrombosis in both capillaries and larger vessels, confirming the diagnosis of CH. Subsequent results from methylation profiling and molecular analysis of the glial tissue showed no numerical alterations or mutations, effectively excluding a glial tumor.

After the four-month postoperative follow-up, the ophthalmological examination showed improvement in visual acuity in the right eye (20/100), as well as in the visual field of both eyes compared to the preoperative and direct postoperative data (Fig. 5, *c, d*). A temporal hemianopia was still noted in the right eye, whereas the left eye showed a normal visual field. However, fundoscopy of the right eye revealed subtle signs of descending primary optic nerve atrophy.

Discussion

Intracranial CH (ICH) is an exceedingly rare benign congenital tumor predominantly observed in children, which typically regresses spontaneously at an early age [3, 4, 21, 24]. Sasaki et al. [6] reported a total of 48 cases of intracranial ICH in both pediatric and adult populations. Xia et al. [23] found 4 cases of ICH in adolescents (13—16 years old). 32 cases of adult ICH were summarized by MacLellan et al. [25], most of them being located on the convexities or extradurally near the sinuses. To our best knowledge, no cases of ICH located in the optic nerve have been reported yet. This was corroborated by our extensive search in the databases Pubmed, Web of Science Core Collection, and Google Scholar.

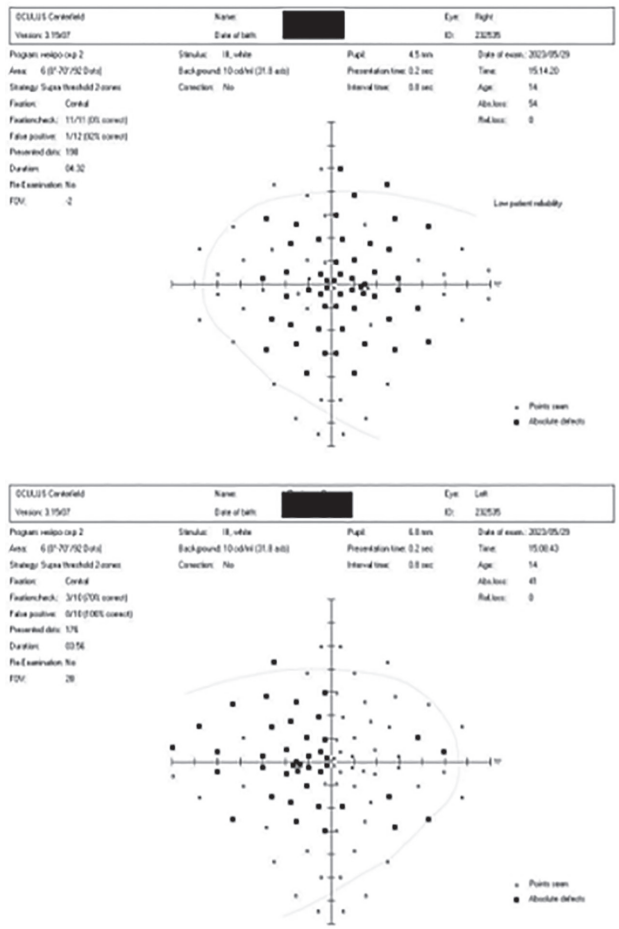

Fig. 3. Automatic static perimetry (screening method) before surgery (above — the right eye, below — the left eye).

According to the literature, ICH demonstrates a slight female predominance and may be associated with hormonal changes, notably during pregnancy [3, 5, 26, 27]. Due to various anatomic locations, ICHs seem to have an unpredictable natural history, ranging from an asymptomatic course to profound

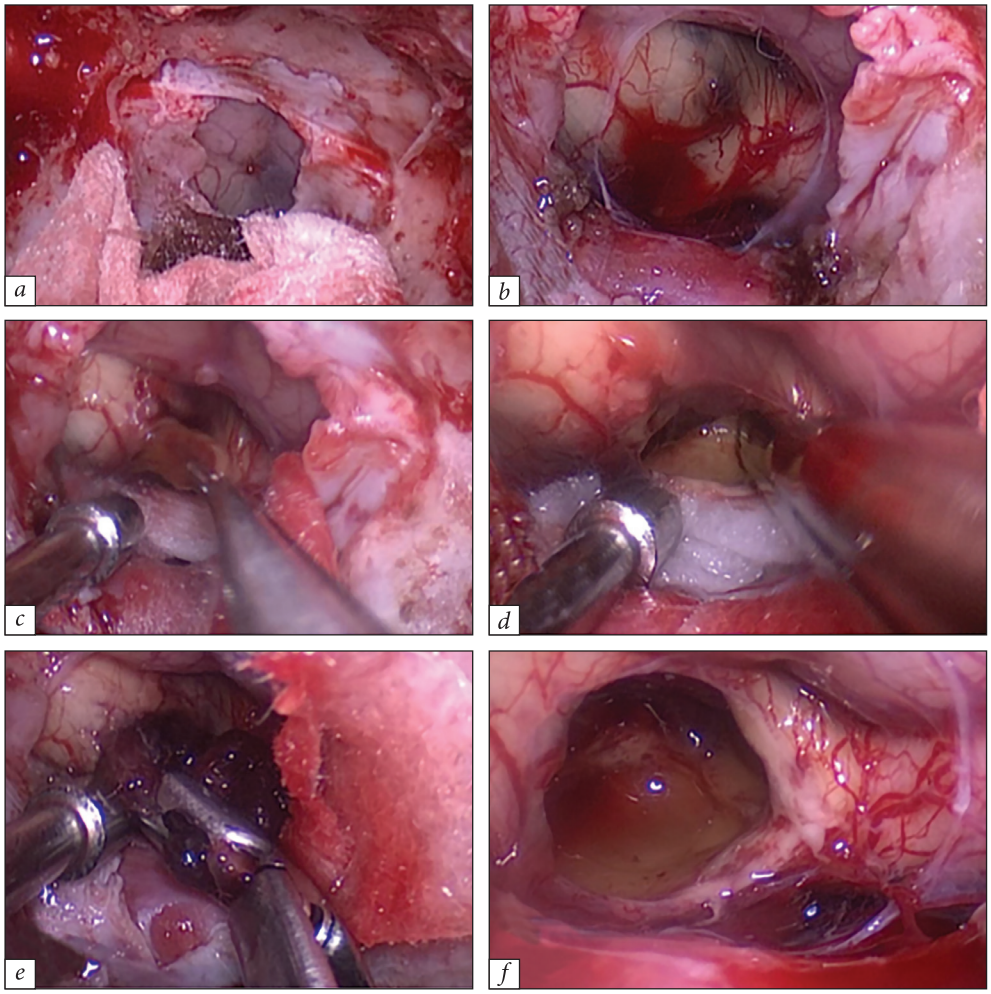

Fig. 4. The stages of the surgical procedure: *a* — Incision of the sellar diaphragm and drainage of the perineural cyst. *b* — Incision of the affected nerve area (purplish discoloration). *c* — Biopsy of the altered neural tissue. *d* — Visualization of a moderately hemorrhagic soft reddish-purple circumscribed vascular lesion, which is not adherent to the surrounding structures. *e* — Removal of the tumor. *f* — Resection cavity

clinical manifestation, on the one side, to spontaneous involution, on the other side, the latter being more characteristic of pediatric patients [3, 4, 21, 25, 28]. However, most cases of ICH are symptomatic and require proper evaluation and treatment.

From a diagnostic standpoint, imaging characteristics of CH present significant challenges. A typical CT scan may show an enhanced homogeneous signal, sometimes with signs of hemorrhage as in other vascular lesions [10, 23, 29]. On MRI, CH are usually hypointense on T1-weighted images and iso- or hyperintense on T2-weighted images [3, 8, 9, 26]. Gadolinium enhancement allows for better visualization of the tumor vessels, especially thin septae between them. The dural-based lesions may homogeneously enhance, mimicking a dural-tail sign typical for meningiomas [23, 25, 30]. However, these features are not pathognomonic for CH, complicating diagnosis based solely on radiological criteria. Cystic changes are typically described in cases of intraparenchymal ICH on MRI but not found intraoperatively as in our case.

A histological diagnosis is also difficult. According to the latest WHO classification of tumors, two groups of vascular lesions can be distinguished: *vascular malformations* and *angiomas*, which are considered developmental anomalies, and *hemangiomas*, true proliferating vascular neoplasms classified as mesenchymal non-meningothelial tumors [16, 17]. Morphological criteria for defining a *hemangioma* include the presence of mesenchymal stroma with fibroblasts, absence of foamy stromal cells, and densely packed capillary and cavernous vessels beneath a single layer of benign endo-

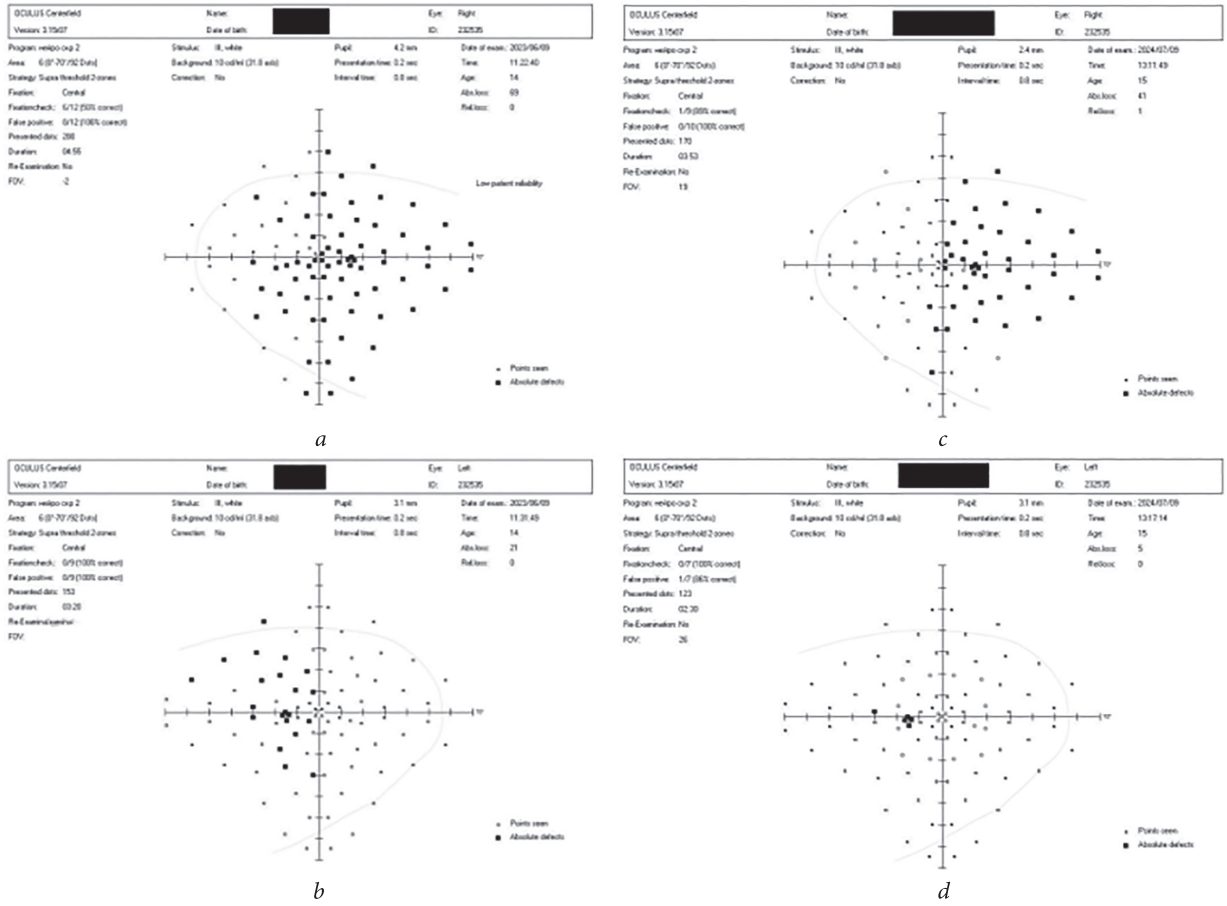

Fig. 5. Automatic static perimetry (screening method) immediately (left, *a, b*) and 4 months after (right, *c, d*) surgery demonstrates enlargement of the visual field (*a, c* — the right eye, *b, d* — the left eye)

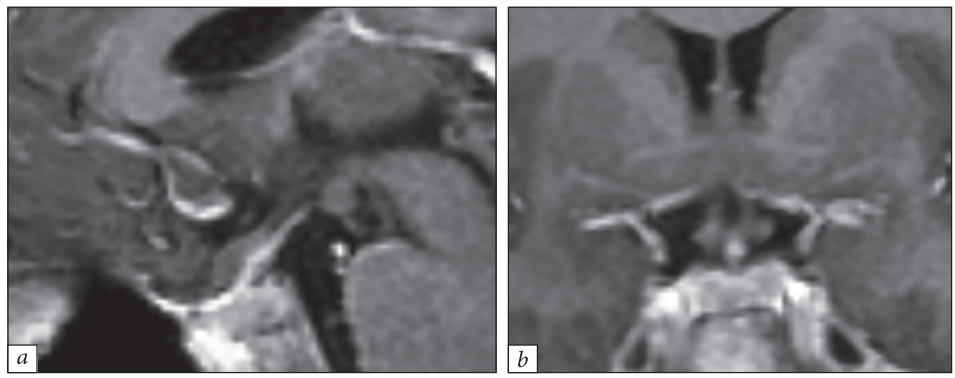

Fig. 6. Postoperative MRI: (*a*) sagittal and (*b*) coronal T1-weighted images showing no signs of residual tumor

thelial cells [17]. These features are believed to be consistent with all ICH regardless of their location [11, 23, 25].

Nonetheless, a confusion regarding the definition of the CH and overall nomenclature of the intracranial vascular lesions still exists. As a consequence, some cases may go misdiagnosed as cavernous malformations, cavernous hemangiomas, and hemangioblastomas or, vice versa, where these lesions are described as CH [13, 21, 28]. Given the scarcity of documented cases of CH, it is likely that many cases remain undiagnosed [26, 30, 31].

Significant questions persist regarding the treatment strategy for CH. The rarity of the condition, the heterogeneous nature and variability of clinical presentation, depending on the tumor location and age, make a standardized treatment algorithm virtually impossible.

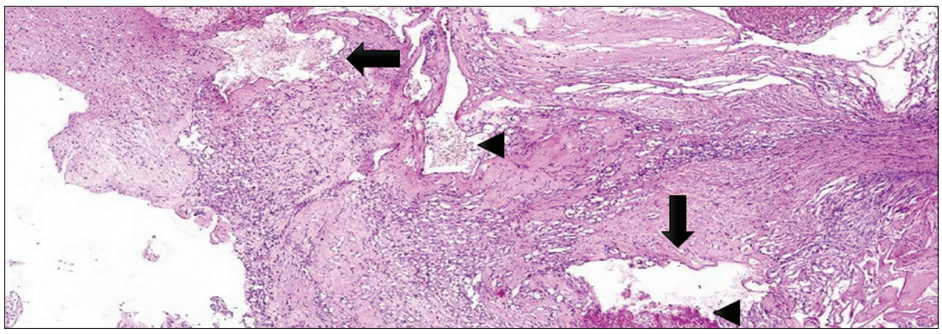

Fig. 7. Histopathological examination revealed tissue morphology consistent with capillary hemangioma (arrows show partially thrombosed vascular channels, arrowheads — hemorrhages). 40×

Pharmacological approaches, including steroids, interferon, and beta-blockers, have been proposed to reduce the lesion volume and to treat associated edema [3, 6, 21, 32]. Recent publications suggest monotherapy with propranolol as an initial treatment strategy for infants and young children [28, 32]. Other medical treatment modalities involving thalidomide, bevacizumab, and temozolomide have also been discussed [2, 9]. Due to the potential for spontaneous regression in pediatric patients, a "watch-and-wait" approach may be recommended in cases with mild to moderate symptoms [7, 28].

Radiotherapy was described for extradural ICH as a stand-alone treatment or as an adjuvant modality after partial resection of the lesion [23].

In symptomatic cases, surgical removal of the tumor is often performed, occasionally preceded by preoperative embolization. In our case, ICH was diagnosed in a 14-year-old boy and was highly symptomatic. The visual decline prompted us to proceed with surgical treatment.

CH of the optic nerve may be reached through intracranial and endonasal routes like the other lesions of the optic pathways, e.g., cavernous malformations, which share similar anatomic features [19, 20, 33, 34]. A classic pterional, mini-pterional, minimally invasive trans-eyebrow, or even an eyelid approaches were described for cavernous malformations, mostly with good outcomes [11]. Alternatively, an endonasal approach may be utilized [33]. In the systematic review of optochiasmatic cavernomas, Ruparelia et al. [34] reported that the endoscopic endonasal approach provided the best visual outcomes, which correlates with the complete surgical removal.

In our case, it was difficult to estimate the accurate position of the chiasma relative to the lesion, although it seemed to be compressed from below. Therefore, we decided that the endonasal approach would offer a straightforward trajectory to the lesion (without unnecessary manipulation on the optic apparatus) and the possibility of complete resection in the case the lesion turns out to be a craniopharyngioma. Th total resection of the hemangioma resulted eventually in an acceptable visual outcome. Although rapid growth and recurrence of ICH were described in the literature, we did not see any signs of lesion during follow-up.

We presented the diagnostic features, clinical course, and treatment strategy for the capillary hemangioma of the optic nerve in a 14-year-old boy. The rarity of the condition, coupled with nonspecific MRI and CT features, and the lack of a typical clinical course limit the possibility of an early and accurate diagnosis of CH. In case of visual decline, surgical removal with decompression of the optic nerve presents a reasonable treatment option. Other strategies may include surgical, interventional, and pharmacological approaches, as well as radio- and chemotherapy, tailored to individual patients depending on their age, location of the lesion, and symptoms.

Statements and Declarations

All authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest, or non-financial interest (such as personal or professional relationships, affiliations, knowledge or beliefs) in the subject matter or materials discussed in this manuscript.

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ВИПАДОК КАПІЛЯРНОЇ ГЕМАНГІОМИ ЗОРОВОГО НЕРВА

Капілярні гемангіоми (КГ) є доброякісними проліферативними судинними новоутвореннями, які зазвичай локалізуються на шкірі та в м'яких тканинах, проте рідко зустрічаються інтракраніально. Більшість із них самостійно регресують у ранньому дитинстві. У роботі описано клінічний випадок 14-річного пацієнта чоловічої статі з гістологічно підтвердженою капілярною гемангіомою оболонки зорового нерва. Візуалізаційні дослідження виявили кістозно-солідну формацію в супраселярній ділянці, зміщену праворуч та розташовану над гіпофізом. Враховуючи прогресивне зниження зору, пацієнту було проведено хірургічне видалення пухлини. Рідкість цієї патології, разом із неспецифічними ознаками на МРТ та КТ, а також відсутністю типової клінічної картини, ускладнюють ранню точну діагностику КГ. У випадках зниження зору хірургічне видалення із декомпресією зорового нерва є доцільним варіантом лікування. Інші стратегії можуть включати хірургічні, інтервенційні, фармакологічні підходи, а також радіо- та хіміотерапію, які підбираються індивідуально залежно від віку пацієнта, локалізації ураження та симптоматики.

Ключові слова: капілярна гемангіома, зоровий нерв, хіазма зорового нерва, хірургічне видалення, клінічний випадок.