Intracranial capillary hemangioma

- Fatal intracranial haemorrhage shortly after belzutifan initiation in von Hippel-Lindau (VHL) disease-associated haemangioblastoma
- Belzutifan for von Hippel-Lindau disease-associated renal cell carcinoma and other neoplasms (LITESPARK-004): 50 months follow-up from a single-arm, phase 2 study
- Vascular malformation in head and neck: a clinicopathological analysis of 675 cases
- Genetic Insights Into Hemorrhagic Stroke and Vascular Malformations: Pathogenesis and Emerging Therapeutic Strategies
- An Unusual Case of Alarming Lactic Acidosis: Brain Metabolic Cross-Talk
- Immunohistochemical Expression of PAX8 in Central Nervous System Hemangioblastomas: A Potential Diagnostic Pitfall for Neuropathologists
- Retinal capillary hemangioma in a pthisical globe: Late sequelae in a case of Von Hippel-Lindau (VHL) disease presenting with endophthalmitis
- Convexity dural hemangioma: illustrative case

An intracranial capillary hemangioma refers to a benign vascular tumor composed of proliferating capillaries, which are the smallest blood vessels in the body. These tumors are typically characterized by a cluster of abnormal blood vessels and can occur within the brain or other parts of the body.

The mean age is 26 with slightly female prevalence (28 F, 25 M). [1)]

Clinical features

The most common presenting symptom was headache (21 cases, 40%) [2)]

Treatment

The surgical treatment consisted of biopsy in 7 cases (13%), partial resection in 10 cases (19%), gross total resection in 31 cases (58.5%), biopsy followed by total resection in 2 cases (3%), and partial resection followed by total resection in 1 case (1.5%), and the diagnosis was obtained from an autopsy sample in 1 case (1.5%). For symptomatic lesions, surgery is a valid option to obtain histological characterization, neurological improvement, and where possible a total resection. Stereotactic radiotherapy can be used if the lesion is not surgically approachable or as an adjuvant treatment in case of partial resection, having shown good results in terms of long-term disease control [3)]

Differential diagnosis

An intracranial capillary hemangioma is not the same as an intracranial cavernous malformation.

An intracranial cavernous malformation, also known as a cerebral cavernous malformation (CCM), is a vascular abnormality that consists of enlarged and irregularly shaped blood vessels called cavernous sinusoids. These sinusoids are thin-walled and lack the normal structure of arteries and veins. Cavernous malformations can occur in the brain and spinal cord, and they may cause symptoms such as seizures, headaches, or neurological deficits depending on their location and size.

While both conditions involve abnormal blood vessels in the brain, they differ in terms of their specific vascular structure and characteristics.

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an intracranial capillary hemangioma is not the same as an intracranial hemangioblastoma.

An intracranial hemangioblastoma is a benign tumor that arises from the cells called hemangioblasts. Hemangioblastomas are most commonly found in the central nervous system, particularly in the cerebellum, but they can also occur in the brainstem or spinal cord. These tumors are composed of two main cell types: stromal cells, which produce blood vessels, and neoplastic cells, which form the bulk of the tumor. Hemangioblastomas are typically highly vascularized and can cause symptoms such as headaches, dizziness, or problems with coordination.

Therefore, while both conditions involve abnormal blood vessels within the brain, they are distinct in terms of their cellular origin and the specific types of blood vessels involved. Capillary hemangiomas originate from abnormal capillaries, while hemangioblastomas arise from hemangioblast cells and exhibit a more complex vascular structure.

Intracranial capillary hemangiomas in adults are rare, and diagnosis can be challenging. Hemangiomas, in general (and particularly in the skin), are more often noted in the pediatric population. Due to the lack of imaging undertaken in the presymptomatic phase, the literature provides few clues on the rate of growth of these unusual tumors.

Literature review

Up to March 2020, the literature review identified 52 cases to which Santoro et al. added a case of personal experience. The mean age was 26 with slightly female prevalence (28 F, 25 M). The most common presenting symptom was headache (21 cases, 40%). The surgical treatment consisted of biopsy in 7 cases (13%), partial resection in 10 cases (19%), gross total resection in 31 cases (58.5%), biopsy followed by total resection in 2 cases (3%), and partial resection followed by total resection in 1 case (1.5%), and the diagnosis was obtained from an autopsy sample in 1 case (1.5%). For symptomatic lesions, surgery is a valid option to obtain histological characterization, neurological improvement, and where possible a total resection. Stereotactic radiotherapy can be used if the lesion is not surgically approachable or as an adjuvant treatment in case of partial resection, having shown good results in terms of long-term disease control [4)]

Case reports

2023

A 64-year-old man with a medical history of Lyme disease who presented with exhaustion and confusion. Imaging demonstrated an intraaxial lesion with vascularity in the posterior right temporal lobe, raising the possibility of a glioma. Imaging two years prior revealed a very small lesion in the same location. The patient underwent a craniectomy, total resection of the lesion was completed, and his symptoms of confusion resolved. Biopsy revealed a capillary hemangioma composed of small vascular channels lined by endothelial cells and pericytes without smooth muscle. Features of glioma, vascular neoplasms or neuroborreliosis (cerebral Lyme disease) were not identified. The case documents the growth over two years of a rare intracranial capillary hemangioma in an older adult male [5].

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a 32-year-old adult male with a capillary hemangioma, which developed within the left cerebellar parenchyma. The histopathological examination reveals a mass mostly formed by the proliferation of capillaries, lined by a layer of flat-plump endothelial cells, some branching and dilating large capillaries, forming a lobulated structure separated by fibrocollagenous connective tissue. Immunohistochemistry examination with CD31 and S100 was positive on the endothelial and stromal cells, respectively, and negative S100 on the endothelial cells. Although rare, capillary hemangioma should be one of the differential diagnoses for diagnosing intra-axial lesions in the cerebellar region. Confirmation of the histopathological characteristic is necessary to determine the diagnosis of capillary hemangioma and exclude other differential diagnoses [6].

2022

A rare case of capillary hemangioma (CH) in a 28-year-old woman suffering from gradual worsening diplopia at 28 weeks of pregnancy. Magnetic resonance imaging (MRI) showed a mass lesion (about 3 cm in diameter) in the right parasellar region. They decided to observe as she was pregnant, and had no symptoms other than right abducent nerve palsy. Fortunately, her symptoms did not worsen until delivery. Computed tomography, enhanced MRI, and angiography after delivery revealed that the lesion was highly calcified and vascularized. A dorsum sellae meningioma or highly calcified pituitary adenoma was suspected and the endoscopic transsphenoidal approach was used for tumor removal. The postoperative course was uneventful. The histological diagnosis was CH. Intracranial CHs or CHs of skull are rare vascular tumors. These tumors are reportedly more common in female patients and may change in size in adults according to menstrual cycle and pregnancy. Only six cases, including that of the present study, were diagnosed during the perinatal period. Some of them experienced rapid symptom progression and tumor growth in their course; thus, we should pay further attention to pregnant or peripartum patients with brain tumor, suspected hemangiomas [7].

2015

A 82-year-old woman presented with vomiting, reduced level of consciousness, and worsening mental state. Computed tomography showed a contrast-enhanced extra-axial lesion in the left frontal operculum, although no intracranial mass lesion was identifiable from magnetic resonance imaging taken 2 years earlier. Complete surgical excision was performed and histopathological examination diagnosed benign capillary hemangioma consisting of a variety of dilated capillary blood vessels lined

by endothelial cells.

This is the first description of rapid growth of an intracranial capillary hemangioma in an elderly woman. These lesions are exceedingly rare in the elderly population, but still show the capacity for rapid growth. Complete excision would prevent further recurrence [8].

[1] [2] [3] [4]
Santoro G, Piccirilli M, Chiarella V, Greco N, Berra LV, Santoro A. Intracranial capillary hemangiomas: literature review in pediatric and adult population. Neurosurg Rev. 2021 Aug;44(4):1977-1985. doi: 10.1007/s10143-020-01419-8. Epub 2020 Oct 20. PMID: 33079288.

[5]
MacLellan AD, Easton AS, Alubankudi R, Pickett GE. Documented growth of an intracranial capillary hemangioma: A case report. Neuropathology. 2023 Jul 4. doi: 10.1111/neup.12933. Epub ahead of print. PMID: 37403213.

[6]
Wardhana DPW, Awyono S, Lauren C, Muhammad Rosyidi R, Saputra H. Intraparenchymal cerebellar capillary hemangioma in a 32-year-old man: a case report. Front Surg. 2023 May 9;10:1141522. doi: 10.3389/fsurg.2023.1141522. PMID: 37228758; PMCID: PMC10203496.

[7]
Ishikawa T, Takeuchi K, Nagata Y, Ito K, Yamamoto T, Kabeya R. Case of a Pregnant Woman with Capillary Hemangioma of the Parasellar Region. NMC Case Rep J. 2022 Apr 21;9:77-82. doi: 10.2176/jns-nmc.2021-0326. PMID: 35646498; PMCID: PMC9107939.

[8]
Okamoto A, Nakagawa I, Matsuda R, Nishimura F, Motoyama Y, Park YS, Nakamura M, Nakase H. Intracranial capillary hemangioma in an elderly patient. Surg Neurol Int. 2015 Oct 23;6(Suppl 21):S539-S542. eCollection 2015. PubMed PMID: 26664868.