Primary eosinophilic granuloma of the oculomotor nerve

Case report

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The authors report the occurrence of primary eosinophilic granuloma of the oculomotor nerve without osseous involvement in a 68-year-old man. Histopathological and neuroradiological findings are discussed. This case demonstrates that eosinophilic granuloma should be included in the differential diagnosis of tumor in which cranial nerves are involved.

KEY WORDS • brain neoplasm • eosinophilic granuloma • oculomotor nerve • histiocytosis • differential diagnosis

Primary eosinophilic granulomas in other cerebral regions are rare and have been reported most often in the frontal and temporal lobes. In the following study, we report the unusual case of primary eosinophilic granuloma in the oculomotor nerve.

Case Report

This 68-year-old man was admitted to our department with diplopia that first occurred 1 1/2 years earlier and showed rapid progression within 6 months prior to admission.

Examination. The patient was alert on admission, and physical examination was normal except for the following neurological findings. He presented with right-sided third nerve palsy with ptosis, divergent strabismus, and anisocoria, the right pupil being non-reactive to light stimulation. The right eye was unable to adduct, elevate, and depress, whereas the left eye showed normal ocular movement. There was no papillary edema, and further examination of the cranial nerves showed no abnormalities. Pareses or disturbances of sensitivity were not evident. Deep-tendon reflexes were symmetrical. The white blood cell count was 7.6 gm/L without eosinophilia in the differential blood count; all further blood parameters were within the normal ranges.
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**Neuroradiological Examination.** Magnetic resonance (MR) imaging revealed an enhancing tumor of 5 mm in diameter located between the right cerebral peduncle and dorsum sellae closely associated with the tentorium and the right oculomotor nerve (Fig. 1); this nerve was judged preoperatively to be either infiltrated or displaced. Computed tomography (CT) revealed an enhancing tumor on the right side of the tentorium extending into the interpeduncular cistern. Transfemoral angiography failed to demonstrate pathological vascularization of the tumor.

**Operation.** We performed a pterional osteoplastic craniotomy on the right side, divided the sylvian fissure extensively, then dissected the right-sided entrance into the dural sheath of the oculomotor nerve. The oculomotor nerve was found to be considerably distended by tumor tissue: the tumor measured approximately 13 × 5 mm (Fig. 2) and was tough, yellowish, and scarcely vascularized. Operative dissection showed that the continuity of all nerve fascicles was destroyed due to tumor infiltration. The tumor did not invade the skull base or underlying dura, and it was resected completely.

**Postoperative Course.** The patient’s postoperative course was entirely uneventful. A neurological examination revealed deficits identical to those found preoperatively. The granuloma was completely removed, and postoperative chemotherapy or radiation therapy was not applied. A follow-up MR investigation performed 1 1/2 years after the operation showed no recurrence of the tumor. A CT study confirmed the absence of tumor at 3 years postoperatively.

**Histopathological Findings.** On examination by microscope, a dense fibrous connective tissue was seen with mononuclear cells being scattered diffusely throughout the tissue or lying in groups with a preference to the perivascular area (Fig. 3a). These cells proved to be reactive with antibodies against T cells (Fig. 3c). Among them, many polymorphonuclear leukocytes, usually eosinophils (Fig. 3b), were found as were some plasma cells and α-1-antitrypsin-positive macrophages (Fig. 3d). The cytoplasm of the latter sometimes contained phagocytosed cells (arrow in Fig. 3d). Further immune reactions with antibodies against neuron-specific enolase and S-100 proteins (Fig. 3e) were positive only in the infiltrated nerve tissue.

**Discussion**
Eosinophilic granuloma has been described in various tissues and organs; most often it is found in the skeletal system, lungs, or gastrointestinal tract. There has been a small number of reports of eosinophilic granuloma in various parts of the brain, such as the frontal lobe,5,10 temporal lobe,12 hypothalamic region,2 and pituitary stalk.7 The occurrence of an eosinophilic granuloma infiltrating a cranial nerve without osseous involvement seems not to have been reported previously. In addition, the age of our patient is unusual for the primary occurrence of eosinophilic granuloma, because in previously reported cases, patients were not older than 44 years of age.7
The diagnosis of an eosinophilic granuloma was

![Fig. 1](image1.png) **Left:** Sagittal right paramedian magnetic resonance (MR) image after gadolinium-DTPA enhancement showing a hyperintense tumor anterior to the cerebral peduncle and posterior to dorsum sellae. **Right:** Axial MR image revealing a tumor with close spatial relation to the right-sided oculomotor nerve.

![Fig. 2](image2.png) Intraoperative photograph taken after a right-sided pterional craniotomy was performed, revealing considerable distention of the oculomotor nerve, seen here below the right optic nerve and carotid artery. The frontal lobe is retracted slightly by a spatula.
based on the granulomatous type of reaction that is present with an infiltration by lymphocytes, polymorphonuclear leukocytes, and eosinophils as well as by plasma cells and macrophages. There was no evidence of malignancy, a primary tumor of the oculomotor nerve or any other intraorbital nerve, or a vascular malformation. Although it has been reported that most histiocytes X cells in the skin are immunoreactive for antibodies against S-100,5,15 in our case only the infiltrated nerve showed a positive reaction, possibly due to a closer affinity of this antibody to Schwann cells than to histiocytes. (Transmission electron microscopy to demonstrate Birbeck’s granules was not performed because the tissue had been entirely fixed in formalin.) A comparable infiltration by eosinophils may also occur in eosinophilia-myalgia syndrome,1 or
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in eosinophilic polymyositis, which are two diseases that do not manifest as tumors of intracranial nerves, however.

Preoperative CT scans showed the lesion to be isodense, with marked enhancement after application of a contrast agent. There was no concurrent edema. In previously reported cases of solitary cerebral eosinophilic granuloma, the appearance of lesions on CT was either hypodense or isodense. Magnetic resonance imaging in our case revealed isointensity on T1-weighted images and high intensity on T2-weighted images, this being in agreement with previous reports. There was marked homogeneous enhancement of the lesion with gadolinium-diethylentriamine penta-acetic acid (Fig. 1); an occurrence that was not found by Penar, et al. These results may indicate that there is a high variability of intracranial presentation of eosinophilic granuloma; therefore, we believe that diagnosis of this type of lesion may not be based solely on radiological findings. Preoperative differential diagnoses in this case included metastasis, neurinoma, lymphoma, and an aneurysm of the posterior communicating artery.

References