Case Report
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대한영상의학회지

Malignant Oncocytoma of the Orbit: A Case Report[1]

안와의 악성 호산성 과립세포종: 증례 보고[1]

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Malignant oncocytoma of the orbit is extremely rare. A 76-year-old man presented with a 10-year history of a mass in the left orbit. Facial computed tomography revealed a large homogeneously enhancing mass. Magnetic resonance images showed a mass with isosignal intensity on a T1-weighted image and low signal intensity on a T2-weighted image. No restricted diffusion was observed on diffusion weighted imaging and apparent diffusion coefficient mapping. The patient underwent an incisional biopsy, and histopathological review revealed a malignant oncocytoma that likely originated in the caruncle of the left eye.

Index terms
Orbit
Adenoma, Oxyphilic
Tomography, X-Ray Computed
Magnetic Resonance Imaging

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INTRODUCTION

Oncocytoma of the ocular adnexa is a rare benign tumor that consists of transformed glandular epithelial cells with abundant eosinophilic granular cytoplasm (1).

Since Radnót (2) introduced the concept of ocular oncocytoma in 1941, > 60 cases of oncocytoma of the ocular adnexa have been reported (3). Oncocytomas account for 3.5% of all lacrimal sac neoplasms (4) and 3-8% of all biopsied caruncular masses (5). Oncocytoma of the ocular adnexa is reportedly prevalent in elderly women (6). Most cases of orbital oncocytoma arise from the caruncle, followed by the lacrimal sac (3). Among them, malignant oncocytoma of the orbit is especially rare.

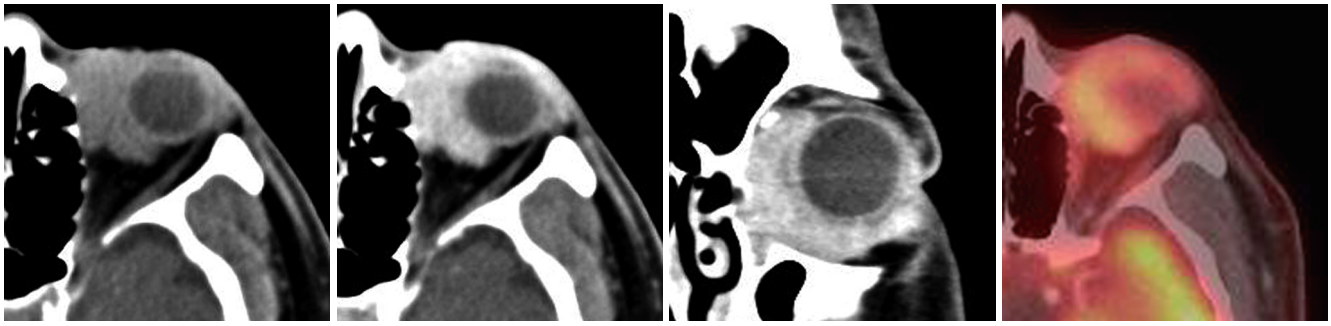
Here we report a case of a 76-year-old man who was diagnosed with a malignant oncocytoma.

CASE REPORT

A 76-year-old man presented with a 10-year history of a left orbital mass. The patient noticed the mass 10 years prior but did not seek treatment since it caused neither pain nor discomfort.

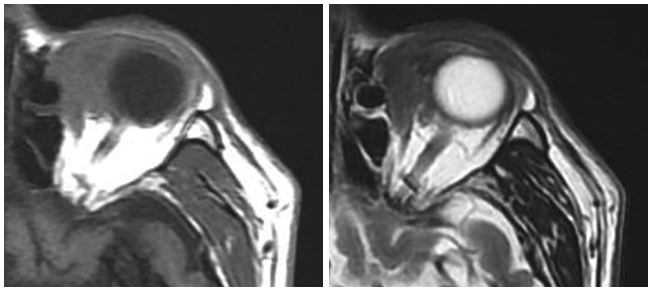
Facial computed tomography (CT) scans revealed a 4.0 × 3.0 × 3.3 cm relatively well-circumscribed, homogeneously enhancing mass in the left orbit. The mass showed high attenuation [77 Hounsfield unit (HU)] without calcification on a pre-contrast CT scan and good enhancement (140 HU) after contrast media administration (Fig. 1). It was located in the medial and inferior portions of the intraconal, conal, and extraconal spaces abutting the left eyeball.

Before the CT scans, he underwent brain magnetic resonance (MR) imaging due to two episodes of transient ischemic attacks. The intervals were 8 months between the initial and follow-up brain MR images and 2 months between the CT scans and the follow-up brain MR images. The size and signal intensity of the mass did not change significantly between either the initial and follow-up brain MR images or the CT scans and follow-up brain MR images. This mass showed isosignal intensity on a T1-weighted image (WI) and low signal intensity on a T2WI (Fig. 2). No



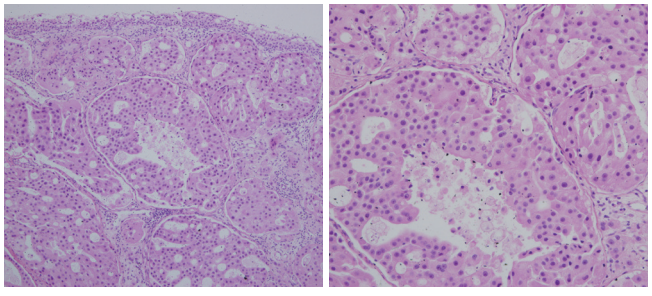
A B C D

Fig. 1. 76-year-old man with malignant oncocytoma of the left orbit.
A. Non-contrast axial CT scan shows a relatively well-circumscribed, high-attenuated mass (77 HU) in the left orbit.
B, C. Contrast-enhanced axial **(B)** and coronal **(C)** CT scans demonstrate strong enhancement (140 HU) in the left orbital mass.
D. PET-CT scan shows high FDG uptake in the left orbital mass.
Note.—FDG = fluorodeoxyglucose, HU = Hounsfield unit, PET = positron emission tomography



A B

Fig. 2. The mass shows isosignal intensity on T1WI **(A)** and low signal intensity on T2WI **(B)**.
Note.—WI = weighted image



A B

Fig. 3. Histopathologic examination (**A** and **B**; hematoxylin and eosin stain, × 100 and × 200) revealed the proliferation of atypical oncocytic neoplastic cells with stromal invasion, which is suggestive of malignant oncocytoma.

restricted diffusion was observed on diffusion weighted imaging (DWI) and apparent diffusion coefficient (ADC) mapping.

An incisional biopsy was performed under local anesthesia. Histopathological examination revealed the proliferation of atypical oncocytic neoplastic cells with stromal invasion, which is suggestive of malignant oncocytoma (Fig. 3).

Because of the malignant features of the lesion, exenteration was recommended, but the patient refused surgery and chose radiation therapy instead. There was no evidence of metastatic lesions on positron emission tomography-CT, which revealed high fluorodeoxyglucose uptake of the mass (Fig. 1). Radiation therapy was started. The mass decreased in size on inspection and follow-up CT scans obtained 1 month after the administration of radiation therapy (6750 cGy).

DISCUSSION

Oncocytic neoplasms are classified as hyperplasias, benign oncocytomas, and malignant oncocytomas (3). Benign and malignant oncocytomas occur in the ocular adnexa, including the caruncle, lacrimal sac, lacrimal gland, eyelid, conjunctiva, and plica semilunaris (3, 6, 7). In contrast to malignant oncocytoma of the salivary gland, especially the parotid gland, malignant oncocytoma of the ocular adnexa is extremely rare. To date, fewer than 10 cases of malignant oncocytoma in the orbit have been reported in the literature (3, 7, 8). Due to their rarity, few reports on CT and MR imaging characteristic findings of ocular oncocytoma are available in the literature. Yuen et al. (8) reported that malignant oncocytomas of the lacrimal sac appear as homogeneously hyperdense lesions on CT scans.

One study reported that the common CT finding of parotid oncocytomas is a well-circumscribed homogeneously enhancing mass with non-enhancing curvilinear cleft or cystic components (9). Parotid oncocytomas are hypointense on T1WI and isointense compared to the native parotid gland on fat-saturated T2WI and postcontrast T1WI (10).

Our case showed the following imaging findings: a relatively well-circumscribed, hyperdense mass without hemorrhage or calcification on pre-contrast CT scan; a homogeneously enhancing mass on post-contrast CT scan; isosignal intensity on T1WI; and low signal intensity on a T2WI mass without restricted diffusion on DWI and ADC mapping.

Although there are some discrepancies in findings among the current study and those of other studies, especially in signal intensity on MR imaging, our case may be helpful for the diagnosis of malignant oncocytoma of the ocular adnexa.

As in our case, oncocytomas of the caruncle usually present without clinical symptoms and are removed to enable a definite diagnosis and for cosmetic purposes (1). However, their complete removal is a curative method for both benign and malignant oncocytoma due to the possibility of local recurrence or malignant transformation (3). Although postoperative radiotherapy is sometimes recommended in cases of recurrence, subtotal resection, or metastases, its precise role remains unknown due to the rarity of cases (3, 8).

In conclusion, here we reported the extremely rare case of a 76-year-old man with a malignant oncocytoma that probably originated in the caruncle. Although the occurrence and location of this oncocytoma is very unusual, it should be kept in mind that malignant oncocytomas can occur in the orbit.

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안와의 악성 호산성 과립세포종: 증례 보고[1]

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안와에 발생한 악성 호산성 과립세포종은 매우 드문 질환이다. 76세 남자가 10년 전에 생긴 좌측 안와의 종괴를 주소로 내원하였다. 안면부 전산화단층촬영을 시행하여 좌측 안와의 균질하게 조영증강되는 커다란 종괴를 발견하였다. 이 종괴는 자기공명 T1 강조영상에서 등신호강도, T2 강조영상에서 저신호강도를 보였으며, 확산 제한은 보이지 않았다. 절개생검을 시행하여 좌안의 눈물언덕에서 기원한 것으로 추정되는 악성 호산성 과립세포종으로 확진되었다.

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