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Oncocytoma of the lacrimal gland- a rare nodular lesion: A literature review

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ABSTRACT

Oncocytoma is a benign neoplasm derived from epithelial cells. It localizes in many different organs, such as the kidney, pancreas, thyroid, and respiratory tract. It can also occur within the ocular area. The following paper presents a literature review of lacrimal gland oncocytoma. This neoplasm in this location is extremely rare, with several cases described to date. This tumor occurs mainly in adults, although some cases described in children also exist. Often, the course is asymptomatic, but it can also lead to pain, swelling, or visual disturbances. Slow growth is characteristic. Differential diagnosis and exclusion of other neoplastic lesions are essential. Histopathological examination can confirm the diagnosis, while imaging tests can determine the extent of the tumor. Management relies on surgical excision of the lesion. Follow-up of patients is necessary, although the recurrence rate is rare.

Keywords: Oncocytoma, lacrimal gland, nodular lesion, tumor, orbital oncocytoma

1. INTRODUCTION

Oncocytoma is a benign tumor derived from epithelial cells. It is rare in the head and neck area (Limb et al., 2013). Oncocytoma can occur in many organs, such as the kidneys, pancreas, adrenal glands, thyroid gland, larynx, or pharynx. However, oncocytoma of the lacrimal gland is highly uncommon. In addition, oncocytoma in the orbital region can involve the lacrimal sac, caruncle, eyelid margin, or conjunctiva. Among oncocytic tumors of the eye, about 6% of them appear in the lacrimal gland. These tumors have a slow growth characteristic. They often have an asymptomatic course. They occur mainly among older adults. The prognosis for lacrimal gland oncocytomas is generally good, with rare recurrences after surgical excision. After treatment, the patient requires regular follow-up (Fenelon et al., 2017). The etiological factors are not entirely understood.

Objective

The purpose of this paper is a literature review of oncocytoma, which is extremely rare in the lacrimal glands.

2. METHODOLOGY

We used databases such as PubMed and Google Scholar to establish this literature review. We utilized keywords such as oncocytoma of the lacrimal gland, oncocytoma etiology, oncocytoma treatment, and orbital oncocytoma. Under these keywords, we found 74 papers published in PubMed and Google Scholar. After excluding papers on oncocytoma of other organs, malignant lesions or available only abstracts, we included 13 articles published from January 2006 to August 2024. We based the paper on case reports of oncocytoma involving the lacrimal gland, review papers, and one retrospective clinicopathological case series.

We included 9 different case reports in this review paper. We mainly included case reports of lacrimal gland oncocytoma. Due to several described cases of tumors in the lacrimal gland, we also analyzed one case report of lacrimal sac oncocytoma. We excluded studies on other ocular adnexa oncocytoma such as caruncle, conjunctiva, and eyelid margin. We also excluded case descriptions of oncocytoma of kidneys, thyroid, pancreas, parathyroid, respiratory tract and other organs besides the organ of vision. One of the papers cited is a case report of malignant oncocytic neoplasm.

3. RESULTS AND DISCUSSION

Etiology

The etiology of lacrimal gland oncocytoma is unknown. Some studies suggest that mitochondria are involved in the pathogenesis of oncocytoma. In their research, Mikkelsen et al., (2017) identified a gain of one copy of chromosome 8 and a loss of one copy of chromosome 22 in a patient with lacrimal gland oncocytoma. However, the significance of the above finding remains unrecognized. Moreover, oncocytomas of other organs also show chromosomal aberrations. In addition, the above study showed the presence of point mutations in mitochondrial DNA (mtDNA). Similar mtDNA abnormalities occur in different tumor locations (Mikkelsen et al., 2017). Research papers emphasize the existence of truncating mtDNA mutations in subunits of respiratory complex I, which impairs the cellular respiration process, leading to the maintenance of the tumor in a benign state with minor proliferation (Bartoletti-Stella et al., 2011).

Epidemiology

Lesions of the lacrimal glands are more common in the female gender. The incidence of oncocytoma of the lacrimal gland is more common in the older population. Researchers report that oncocytoma in the adnexa of the eye occurs in an average of 0.3 people per million per year (Jittapiromsak et al., 2017). Based on 13 case reports of oncocytoma of the lacrimal gland, Mikkelsen et al., (2017) determined that the median age of onset of the lesion was 57 years. The literature describes only a dozen cases of such a tumor (Mikkelsen et al., 2017). Benign oncocytic neoplasms can also occur in children. Fenelon et al., (2017) described a case of lacrimal gland oncocytoma in a 4-year-old girl.

Clinical manifestation

Oncocytomas are usually asymptomatic (Jittapiromsak et al., 2017). Some patients report experiencing swelling of the orbital region and pain (Calle et al., 2006). Mikkelsen et al., (2017) analyzed 12 previously reported cases of lacrimal gland oncocytoma. Patients reported symptoms such as protoptosis, diplopia, edema, orbital mass, lid swelling, and asymmetry. In the above analysis, tumor sizes ranged from 10mm to 50mm. A 20-year-old patient diagnosed with lacrimal gland oncocytoma showed reduced visual acuity in the eye on the side of the tumor due to impaired refraction through the tumor mass. In contrast, visual field and color vision were standard (Mikkelsen et al., 2017).

Oncocytoma of the lacrimal gland is characteristic of slow growth. In a case report, Calle et al., (2006) presented a patient with lacrimal gland oncocytoma who reported swelling of the orbital region and pain for 7 months, which did not resolve after non-steroidal anti-inflammatory drugs (NSAIDs). Another case report involved a patient who observed a lesion of the upper eyelid area with discomfort for 6 months (Economou et al., 2007). In addition, Kim et al., (2010) described the case of a 64-year-old patient who had observed recurrent swelling of the orbital region for 7 years, accompanied by ptosis.

Diagnosis

Histopathological examination allows confirmation of the presence of oncocytoma. There are two types of oncocytoma cells, the first being polygonal and the second columnar type. These cells differ in the number of mitochondria. On light microscopy, one can observe the presence of large epithelial cells with eosinophilic cytoplasm and small nuclei without atypia (Economou et al., 2007). Using phosphotungstic acid-hematoxylin (PTAH) staining on histopathology allows observation of dark blue stained, mitochondria-rich cytoplasm (Calle et al., 2006).

In the histopathological image, normal cells of the lacrimal gland are separated from tumor cells by narrow stripes of collagen fibers (Economou et al., 2007). The cells have a granular appearance on electron microscopy due to the large number of irregular mitochondria (Calle et al., 2006). We presented the comparison of light and electron microscopy descriptions in (Table 1). Immunohistochemical staining described immunoreactivity for pan-cytokeratins (CK) (Fenelon et al., 2017).

Table 1 Comparison of light and electron microscopy in oncocytoma of lacrimal glands.

Light microscopy	Electron microscopy
Large epithelial cells present, cell nuclei without atypia. Eosinophilic cytoplasm with granularity.	Granular cells, large number of mitochondria varying in shape and size with irregular cristae.

The tumor mass is visible on a computed tomography (CT) scan (Kim et al., 2010). Imaging diagnostics include magnetic resonance imaging (MRI). This examination can determine the tumor's size and the gland's anatomical arrangement and helps assess margins (Almutairi et al., 2022). The MRI image of an oncocytoma is a well-demarcated, solid tumor. On T2- dependent images, the lesion is heterogeneously hyperintense. There are possible hollow areas within the lesion, which are the result of neovascularization. In turn, bone remodeling suggests slow growth of the lesion. The authors emphasize that in the case of oncocytic carcinoma, the lesion shows more aggressive growth and may lead to invasion of the extraocular muscles.

Jittapiromsak et al., (2017) described dynamic contrast-enhanced MRI (DCE MRI), which can complement the diagnosis of oncocytoma. The study involves the administration of contrast, and the images are from before, during, and after the administration of the substance. The above test allows the assessment of vascular permeability and blood flow (Jittapiromsak et al., 2017). Differential diagnosis includes other neoplastic lesions of the lacrimal glands, such as pleomorphic adenoma, adenoid cystic carcinoma, or mucoepidermoid carcinoma (Mulay et al., 2018).

Treatment

The primary treatment for lacrimal gland oncocytoma is surgery. After surgery, it is necessary to monitor the patient for possible recurrence. The surgical method, when the tumor localizes extraocularly, is a lateral orbitotomy through the zygomatic bone flap. The procedure undergoes under general anesthesia (Fenelon et al., 2017; Kim et al., 2010). In contrast, Economou et al., (2007) described the removal of the tumor via anterior orbitotomy. In this case, the course of surgery was uncomplicated (Economou et al., 2007). Sweeney and Allen, (2021) described a richly vascularized lesion where bleeding occurred during surgery.

Recurrences after surgical treatment are infrequent. Calle et al., (2006) observed no tumor recurrence in a patient after surgery at follow-up for 21 months. In the case described by Economou et al., (2007) the patient had a follow-up for 2 years, with no recurrence during that time. The literature describes a case of an 82-year-old patient with oncocytoma of the lacrimal sac with recurrence after surgical treatment after several years. The lesion was still benign (Almutairi et al., 2022).

Oncocytic carcinoma

Oncocytic carcinoma is a rare malignant neoplasm composed of oncocytic cells. These cells present polymorphic, granular, eosinophilic cytoplasm. They show invasive growth. This neoplasm can be a primary lesion, but transformation from a benign tumor is also possible (Vahdani and Rose, 2025). However, there is no evidence of malignant transformation for tumors located in the lacrimal gland (Calle et al., 2006). The literature describes a case of a 61-year-old female patient with a four-year history of ptosis. The patient underwent surgery with an excisional biopsy. Histopathological examination diagnosed malignant oncocytoma of the lacrimal gland. In addition to surgery, the patient underwent radiation therapy. No recurrence was observed 10 months after treatment (Timoney et al., 2011).

4. CONCLUSIONS

Benign oncocytic neoplasm of the lacrimal gland is extremely rare. Only about a dozen case descriptions of such a tumor are available. In addition to the lacrimal gland, other localizations within the ocular region are possible, such as lacrimal sac, eyelid, conjunctiva, and caruncle. Oncocytoma is more characteristic in the elderly population. Usually, the course is asymptomatic or sparsely symptomatic. The most common symptoms include pain, swelling of the eye area, and protoptosis.

Proper diagnosis is essential, as well as providing histopathological examination and additional imaging tests such as CT or MRI. It is also necessary for the doctor to perform a proper differential diagnosis and exclude other cancers of the area, including malignant lesions such as adenoid cystic carcinoma. Surgical treatment of lacrimal gland oncocytoma is the treatment of choice. After treatment, the patients require follow-up for possible recurrences; however, these are rare.

Authors' Contribution

Jagoda Saniuk: Conceptualization, writing- rough preparation, investigation, methodology, project administration, writing - review and editing
Aleksandra Anioła: Supervision, data curation, writing - review and editing, investigation
Klaudia Romejko: Methodology, data curation, resources, formal analysis
Sandra Sienkiewicz: Formal analysis, visualistaion, writing- rough preparation
Justyna Rajczyk: Investigation, methodology, data curation
All authors have read and agreed to the published version of the manuscript.

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Ethical approval

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Informed consent

Not applicable.

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Conflict of interest

The authors declare that there is no conflict of interests.

Data and materials availability

All data sets collected during this study are available upon reasonable request from the corresponding author.

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