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## Short communication

# Choroidal metastasis of a minor salivary gland adenoid cystic carcinoma: A case report<sup>☆</sup>

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### ABSTRACT

**Case report:** A 61-year-old man with a lower lip minor salivary gland adenoid cystic carcinoma, suffered from a unilateral progressive visual acuity loss due to choroidal metastasis. **Discussion:** Adenoid cystic carcinoma is a rare primary tumor with significant metastatic potential. Our patient presented with a unilateral choroidal metastasis. According to the current literature, 8 cases of choroidal metastasis of salivary gland adenoid cystic carcinoma have been reported. This is the second case reported of choroidal metastasis with origin in a minor salivary gland, and the first one with origin in the minor salivary glands of the lower lip.

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## Metástasis coroideas de un carcinoma adenoide quístico de glándula salival menor: caso clínico

### RESUMEN

**Caso clínico:** Varón de 61 años con un carcinoma adenoide quístico de glándulas salivales menores de labio inferior presentó deterioro unilateral y progresivo de la agudeza visual debido a una metástasis coroidea.

**Discusión:** El carcinoma adenoide quístico es un tumor infrecuente con un importante potencial metastásico. Nuestro paciente presentó una metástasis coroidea unilateral. Actualmente hay descritos 8 casos de metástasis coroidea de un carcinoma adenoide quístico de glándulas salivales. Este es el segundo caso descrito de metástasis coroidea con origen en un tumor de glándulas salivales menores y el primero con origen en las glándulas salivales menores del labio inferior.

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#### Palabras clave:

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## Introduction

The ocular globe is rarely compromised by the dissemination of malign tumors due to the lack of its own lymphatic system. Metastases involving ocular structures are usually secondary to hematogenous dissemination. The uveal tract is the ocular structure having the highest blood irrigation, as nearly 90% is constituted by the choroids, which is the target of the majority of ocular metastases.<sup>1</sup>

After mucoepidermoid carcinoma, adenoid cystic carcinoma (ACC) is the second most frequent primary malign salivary gland (SG) neoplasia. It can originate in the major SG (parotid, submandibular and sublingual) or in the minor SG, distributed along the higher aerodigestive tract.<sup>2</sup> Approximately 50% of minor SG carcinoma are ACC.<sup>3</sup> In general, they are small and slow growing. However, they could exhibit subclinical invasion with silent course during many years and distant metastasis in approximately 50% of cases.<sup>2</sup> ACC have poor prognosis in the long-term with lower survival rate (23–40% at 15–20 years) and high late local recurrence rates (16–67%).<sup>2</sup> To date only 8 cases of ACC choroidal metastasis have been described, of which only one originated in the minor SG of the hard palate, with the present case being the first described with choroidal metastasis originating in a minor SG lip ACC.

## Clinical case report

Male, 61, with complete surgical resection of a lower lip minor SG in 2003 (Fig. 1A). Pathological anatomy confirmed the diagnostic of ACC with predominantly tubular pattern and perineural invasion (Fig. 1B). The patient was administered adjuvant radical radiotherapy (68 Gy) over the tumor and the left side of the neck. In 2014, the patient presented with bilateral pulmonary and ganglionic mediastinic metastases (Fig. 1C). Cytostatic treatment was initiated but the patient exhibited toxicity problems and clinic progression (hepatic, umbilical and bone metastases) (Fig. 1D–F). In 2004, during a third line of cytostatic treatment, the patient was admitted to the Oncology Dept. due to hemoptysis and dyspnea secondary to thoracic progression. While admitted, the patient exhibited progressive visual acuity deterioration in the left eye (OI) with one-month evolution.

Ophthalmological examination showed best-corrected visual acuity of 0.4 (decimal scale) in the right eye and movement of hands in the LE. Ocular fundus examination showed inferior exudative retina detachment associated to choroidal detachment and temporal subretinal hemorrhage in the LE (Fig. 2A–C). Optical coherence tomography showed subretinal macular fluid (Fig. 2D). Ocular echography showed a fixed choroidal mass in the inferior temporal wall (Fig. 3A). Orbital magnetic resonance (RM) with gadolinium showed moderate homogeneous enhancement of a mass in the posteroinferior area of the LE, suggesting choroidal metastasis (Fig. 3B). Cerebral MRI showed multiple nodular metastases in the cerebellum and brain (Fig. 3C and D). After multidisciplinary assessment, the patient was treated with palliative radiotherapy on the lesion in the left pulmonary hilum (overall doses

of 30Gy in 2 weeks). Five days later palliative holocraneal radiotherapy was administered (overall dose of 37.5 Gy in 15 fractions) that were not completed due to demise of the patient 1 week later.

## Discussion

ACC is a rare type of tumor that primarily involves the head and neck, particularly the SG.<sup>2,4</sup> It accounts for 0.6% of malign head and neck tumors, and approximately 10% of SG tumors. ACC presents local recurrences and about 50% develop distant metastasis even after the primary tumor has been controlled.<sup>2,4</sup> The most frequent metastasis occurs in the lungs although the literature describes metastases in bones, central nervous system, liver, thyroid and spleen.<sup>4</sup>

The present case exhibited belated distant pulmonary, mediastinal, hepatic, umbilical and bone metastases 10 years after primary tumor diagnostic, and one year later developed metastasis in the central nervous system and the choroids. Even though choroidal metastases are rare, the literature describes 8 cases of SG ACC with choroidal metastases. Of these, 6 described the origin of choroidal metastases in the submandibular gland<sup>4</sup> and one in the parotid gland.<sup>5</sup> Gutmann et al. described the first and only case of choroidal metastases originating in the minor SD (hard palate mucosa) in 1986.<sup>6</sup> The present case also originated in the minor SG although not in the hard palate mucosa; accordingly, this is the first published case of choroidal metastasis originating in a minor lip mucosa SG.

On the other hand, metastases comprise the most frequent intraocular malign tumors in adults.<sup>7</sup> In 1997, Shields et al. published data of 520 eyes of 420 patients with uveal metastasis. The origin thereof was mainly primary tumors in mammary glands (40%), lungs (36%) and one cutaneous melanoma (8%).<sup>8</sup> None of the choroidal metastasis cases originated in a minor SG.

As in the present case, in the 8 cases published to date patients debuted with progressive unilateral or bilateral visual acuity impairment, with the most frequent clinic being associated to choroidal metastases.<sup>1</sup> The second most frequent choroidal metastasis clinic are visual field defects and the appearance of myodesopsia. Overall, 90% could exhibit secondary exudative retinal detachment.<sup>1</sup>

When choroidal metastasis is suspected or confirmed, cranial imaging tests could be useful before considering treatment because some data indicate that approximately 22% of choroidal metastases have a concurrent central nervous system metastasis diagnostic.<sup>1</sup> Accordingly, cerebral MR could be a decisive test even in asymptomatic patients as in the present case because cerebral and intraocular malign neoplasia frequently coexist or follow each other.

Therapeutic alternatives for choroidal metastases include observation, cleavage, enucleation, radio and chemotherapy.<sup>1,5</sup> At present, radiotherapy is the treatment of choice for choroidal metastasis<sup>1,9</sup> as it facilitates local control and avoids enucleation and visual loss. In small tumors far from the macula and the optic nerve, plate brachytherapy can be used.<sup>5</sup> Despite its palliative nature, radiotherapy can

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