Short communication

Bilateral conjunctival oedema as a symptom of adrenal adenocarcinoma☆,☆☆

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ABSTRACT

Clinical case: We present a case of a 67-year-old female suffering from bilateral conjunctival oedema that did not improve with antibiotic and antiinflammatory treatment. After a complete systemic examination she was diagnosed with an adrenal adenocarcinoma and treated by surgery and chemotherapy. The conjunctival chemosis subsequently improved. Unfortunately the patient died of respiratory failure due to metastasis.

Conclusion: We emphasise the need of an exhaustive and complete examination by the ophthalmologist in cases of non-responders to topical antibiotic and antiinflammatory treatment in patients with bilateral conjunctival oedema, in order to make a correct diagnosis of systemic potentially lethal diseases.

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Quemosis conjuntival bilateral como síntoma de adenocarcinoma suprarrenal

RESUMEN

Caso clínico: Se presenta el caso de una paciente de 67 años que acude por edema conjuntival bilateral resistente a tratamiento tópico con antibióticos antiinflamatorios. Tras un completo examen sistémico llegamos al diagnóstico de adenocarcinoma suprarrenal que fue tratado mediante cirugía y quimioterapia, tras los cuales la quemosis mejoró. Desafortunadamente, la paciente falleció por insuficiencia respiratoria debido a metástasis.

Conclusión: Se destaca la necesidad de realizar un examen exhaustivo y completo por el oftalmólogo en casos de pacientes con edema conjuntival bilateral no respondedores a tratamiento tópico con antibióticos y antiinflamatorios, para descartar enfermedades sistémicas potencialmente graves.

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Introduction

In its localized and diffuse form, conjunctival chemosis can be expressed as a sign in different ophthalmological or systemic diseases as well as in inflammatory, allergic or other conditions. Cortical adrenal tumors have a prevalence of at least 3% in populations over 50.1 The Cushing’s syndrome screening is appropriate for diabetic patients exhibiting poor glycemic control as over 2% of this population exhibits said entity.2

Clinical case

Female, 67, who visited the emergency service due to bilateral conjunctival oedema with 3 weeks of evolution after being treated with artificial tears and topical antibiotic-corticosteroid ointment during said period without improvement. Relevant personal history included arterial hypertension in pharmacological treatment, poorly controlled diabetes mellitus and recently diagnosed oral candidiasis. The ophthalmological exploration revealed a bilateral conjunctival chemosis with intense tearing (Fig. 1), with the rest of the exploration being normal. The patient referred shedding 5 kg of weight in the past 2 weeks, a general feeling of swelling, mainly in the face, together with generalized fatigue and pruritus.

After a systemic exploration, she is diagnosed with suspected Cushing’s syndrome. Hormonal analysis revealed ACTH 3 pg/mL (normal 10–55), baseline plasmatic cortisol of 35.3 mcg/dL and urinary free cortisol (CLU) of 632 mcg/24 h (normal <180). Thorax X-ray and computerized axial tomography (CAT) were normal, but the abdominal CAT revealed an adrenal mass of 8.2 cm × 5.4 cm on the left side, suggesting adrenal carcinoma, without locoregional adenopathy (Fig. 2).

Considering the clinical evolution of the patient, it was decided to begin treatment with ketoconazole 400 mg/day prior to surgery in order to inhibit the secretion of suprarenal cortisol, subsequently performing the adrenal tumoration excision together with surrounding fat, exhibiting a complete resolution of the chemosis (Fig. 3).

The histology of the adrenal tumoration was reported as “carcinoma of the suprarrenal cortex with extensive necrosis along with proliferative activity, infiltrating the surrounding capsule and invading the pericapsular lymphatic vessels”. 3 months later, after the first indication of chemotherapy with cisplatinum-etoposide, a thorax-abdomen–pelvic CAT revealed multiple bilateral pulmonary metastasis in both hemi-thorax (Fig. 4) and a local relapse at the left suprarenal level. Accordingly, the patient was administered 3 chemotherapy cycles with cyclophosphamide, adriamycin and vincristine without improvement and progression of the respiratory and cardiac insufficiency. Then, treatment was established with weekly taxol for 2 months (antineoplastic chemotherapy), during which the patient died due to progressive respiratory insufficiency.

Discussion

Bilateral conjunctival chemosis is a frequent clinical finding in daily clinical practice. It is characterized by the presence of an excess liquid in the conjunctiva. Typically, it is a self-limited or reversible, non-specific clinical entity. However, in the presence of a persistent conjunctival oedema clinical condition, in addition to a complete ophthalmological exploration, a hemogram and biochemistry analysis and neuroimaging must
be made in order to confirm any obstruction of venous or lymphatic drainage caused by, for example, a superior vena cava syndrome or a carotid-cavernous fistula. But in our case, this was not done. From a diagnostic point of view, a conjunctival biopsy can be carried out because, in our case, some systemic diseases are present which could have an important visual repercussion and even threaten the patient’s survival and which could be diagnosed by a conjunctival biopsy. The exact etiopathogeny of this edematous condition is not entirely clear. The various causal mechanisms proposed include vaso- genic or cytotoxic stimuli or altered hydro-electrolytic balance due to the increase in circulating cortisol.

Frequently, the diagnosis of Cushing’s syndrome cannot be based on exploratory clinical findings because not all symptoms are expressed in all patients. The patients exhibiting an excess of steroid hormones of adrenal origin exhibit the predominant traits of a sudden onset of Cushing’s syndrome with or without virilization signs.

Prior to tumor surgery, the treatment of choice, there is a requirement to carry out a complete endocrine study as well as an extended cranium–thorax–abdominal imaging, in addition to discarding the presence of a pheochromocytoma.

Due to the variable levels of hypercortisolemia and the rapid reproductive development of adrenal carcinomas, the clinic of Cushing’s syndrome is frequently subdued in our patient.

However, local ophthalmological reactions such as conjunctivitis or conjunctival chemosis are exceptional possible expression of Cushing’s syndrome. Reviewing the published literature to date, this is the only case of ophthalmological suspicion of the Cushing’s syndrome caused by adrenal carcinoma.

To conclude, we advocate for an exhaustive and complete study of patients with bilateral conjunctival oedema who did not respond to conventional topical treatment (antibiotic and anti-inflammatory) in order to carry out an early diagnostic of potentially severe systemic diseases.

Conflict of interests

None of the authors have declared any conflict of interests.

References