Short communication

Unsuccessful treatment with OK-432 picibanil for orbital lymphangioma

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ABSTRACT

Introduction: Lymphangioma is a malformation of the lymphatic system. The classic approach is surgery. We report a case of orbital lymphangioma in a girl who was given OK-432 to avoid surgery and its complications.

Discussion: OK-432 is a lyophilized mixture of group A Streptococcus pyogenes which produces a fibrosis limited to the lesion with a high cure rate. The main advantages are the easy intralesional application, with no scars and or damage of closed areas. Its main disadvantage is a significant local inflammatory reaction.

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Resultado sin éxito en el tratamiento de un linfangioma orbitario con OK-432. Picibanil

RESUMEN

Introducción: El linfangioma es una malformación linfática cuyo tratamiento habitual ha sido la cirugía. Aportamos un caso de linfangioma orbitario en una niña a la que se trató con OK-432 intralesional para evitar la cirugía y sus complicaciones.

Discusión: OK-432, es una mezcla liofilizada de Streptococcus pyogenes del grupo A, que produce una fibrosis circunscrita de la lesión con una alta tasa de curación. Sus principales ventajas son su fácil aplicación intralesional y la falta de cicatrices y lesiones a las estructuras adyacentes de la lesión. Aunque su gran inconveniente es la gran reacción inflamatoria local que provoca.

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Introduction

A 13-year-old girl visited the emergency service due to upper palpebral ptosis in the left eye (Fig. 1). When the eyelid was lifted it revealed a soft, painless, mobile and richly vascularized mass which was suspected to be lymphangioma (Fig. 2). Nuclear magnetic resonance (NMR) was performed which confirmed a left temporal superior orbitary lymphangioma.


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Fig. 1 – Patients at day one, mechanical ptosis can be seen.

Fig. 2 – Lymphangioma with inverted eyelid.

Fig. 3 – Nuclear magnetic resonance of the lesion at T2.

Fig. 4 – Acute inflammatory reaction 48 h after the treatment.

Fig. 5 – Appearance of the lesion after treatment with increased lymphangioma content.

is in the internal third of the upper eyelid. This case is upper external.1

Diagnostic is clinical and is based on imaging techniques, with NMR supplying the highest amounts of information. The progression of the lymphangioma size is a consequence of bleeding. The treatment of choice is surgical but it is not free of relapses and complications.1,2

Treatment of lymphangioma comprises a range of therapeutic options, notably substances with sclerotizing effect on the vascular walls of the lymphangioma, such as intrallesional corticoids and tetracyclines with their well-known side effects.3

In order to prevent publications derived from surgery and the above mentioned sclerotizing effects we opted for an intrallesional injection of OK-432 (Picibanil) as the first therapeutic option.

(Fig. 3). The patient did not exhibit relevant ophthalmological or general history. With the exception of the mechanical ptosis, the ophthalmological exploration gave normal results.

Lymphangiomas are benign vascular malformations that tend to bleed spontaneously but circumscribed to the lesion. They do not exhibit volume changes with the Valsalva maneuver. The most frequent location of palpebral lymphangiomas
exhibiting allergy to β-lactam products due to the risk of anaphylactic reaction. It has been utilized in digestive and pleural tumors due to its sclerotizing action. Recently, this substance started to be applied for treating lymphangiomas due to its action mechanism which involves an increase in inflammatory cells, the natural killer cells, CD3 lymphocytes, interferon-α and interleukin-6 causing an increase in the permeability of the lymphangioma endothelium and enhanced in its lymphatic drainage, leading to an emptying of cystic spaces and their subsequent collapse and sclerotization which causes lesion size reductions and therefore their disappearance.²,⁴

The therapeutic dosage is of 0.02 mg of OK-432 to be injected in the lesion. If one does not achieve the expected results a second dose of the same amount of OK-432 is indicated 10–15 days after the first dose of OK-432 to enhance the sclerotizing effects of the substance due to the increase of the intralesional dose.³

In this case, surgery was dismissed as the first choice due to the post-surgery complications that arise in the majority of cases.³,⁴ The patient recovered her initial appearance after the treatment and the NMR did not reveal reduction in the size of the lesion.

Surgical treatment of lymphangiomas is not free of complications and relapses. The utilization of OK-432 exhibits the advantage of easy application, the absence of scars and of lesions in actuation structures. However, its drawback is a high local inflammatory reaction which, even though it diminishes with time, makes its application controversial in intra-orbital lesions. Despite the achievements obtained in other localizations, orbital locations present difficulties. It could be that in this patient the early bleeding prevented the sclerotizing action of the product. The poor result in our case could add to the findings of other authors in the sense that this treatment is not appropriate for orbital locations.⁵

References


Surgical maneuver

0.02 ml of the lymphangioma contents were extracted under general anesthesia for anatomopathological study. Subsequently, 0.02 mg of the OK-432 sclerotizing substance were injected into the lymphangioma. No secondary or local effects arose during the application of OK-432. Even though the procedure is simple, the patient was admitted due to possible local complications, prescribing prophylactic analgesia and antibiotics.

Forty-eight hours after the application of OK-432. The patient exhibited a considerable increase in the size of the lymphangioma which prevented her from opening the eye due to the local inflammatory reaction (Fig. 4). She exhibited brownish lesions typical of lymphangioma bleeding and a palpebral blood clot that produced the ptosis in the left eye (Fig. 5).

Despite the condition, intraocular pressure, visual acuity, funduscopy and pupil reflexes were normal and the patient did not exhibit fever. Systemic corticoids were added to the initial treatment to counteract the local inflammatory reaction.

A computerized axial tomography was taken in the emergency ward to confirm that the increase in size of the lymphangioma was due to bleeding (Fig. 6). It was decided to observe the evolution of the OK-432 in the lesion.

Two weeks after the application of OK-432. The size of the lymphangioma began to diminish. Two months later, a 2 mm ptosis still persists.

Due to local complications, a new dose of OK-432 was not prescribed even though the literature recommends it when the first intralesional injection does not yield the expected results.

Discussion

OK-432 is low virulence liohpilized substance from Group A Estreptococci pyogenes. It is contraindicated in patients

Fig. 6 – Computerized axial tomography showing a significant increase of the lymphangioma.