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Ophthalmology A

Bilateral Inflammatory Pseudotumor in a Child

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Abstract **Case Report**

Orbital inflammatory pseudotumor is defined as a benign, non-specific, expansive intraorbital process of non-infectious inflammation, for which no local or systemic cause can be identified. In the pediatric population, this entity is rare and presents a distinct diagnostic challenge, as it must be primarily differentiated from rhabdomyosarcoma. We report the case of an 11-year-old girl diagnosed with bilateral orbital pseudotumor.

Keywords: inflammation, children, orbit, benign, exophthalmos, Orbital biopsy.

INTRODUCTION

Inflammatory pseudotumors of the orbit refer to any benign, non-specific, non-infectious, inflammatory intraorbital expansive process with no identified local or systemic cause. +1+ Inflammatory pseudotumors of the orbit are a distinct pathological entity in children: they are rare and primarily pose a differential diagnosis challenge with rhabdomyosarcoma. We report the case of an 11-year-old girl with bilateral orbital pseudotumor. +2+

CASE REPORT

A patient aged 11, followed for juvenile idiopathic arthritis since the age of 9 and treated with NSAIDs, presented with bilateral exophthalmos accompanied by mild pain upon eye movement, without a decrease in visual acuity.

Ophthalmological examination revealed bilateral, symmetrical, painless, axial, non-pulsatile exophthalmos. Oculomotor motility was preserved but slightly painful. Photomotor reflexes were present and symmetrical. Intraocular pressure was 11 mmHg. Examination of the anterior segment was strictly normal. Examination of the posterior segment showed papillary hyperemia with slightly blurred margins and a good foveal reflex. General examination revealed arthralgia of both wrists, knees, and the cervical spine.

Laboratory workup showed an ESR of 90 mm in the first hour, CRP at 50. Thyroid function tests returned normal. Antinuclear antibodies, ANCA, rheumatoid factor, and anti-DNA antibodies were normal.

An orbital CT scan was performed initially, showing a spontaneously hyperdense, bilateral, fusiform, intraconal process encasing the globe and the optic nerve, enhancing after contrast injection, with stage III bilateral exophthalmos.

An orbital MRI was performed and was consistent with bilateral tissue infiltration of the intraconal fat, with enlargement of the intraconal space. The lesion appeared isointense on T1 and hypointense on T2, with no diffusion restriction, and showed intense enhancement after contrast injection. This infiltration exerted a mass effect on the extraocular muscles while preserving a fatty cleavage plane, and was responsible for grade III exophthalmos. The extraconal fat was spared. The MRI concluded with findings suggestive of an inflammatory orbital pseudotumor.

An orbital biopsy was performed and returned showing a reactive lymphocytic infiltrate.

The diagnosis of an inflammatory pseudotumor was established. The child was started on corticosteroid bolus therapy after a pre-bolus workup. The clinical course was marked by improvement and a decrease in the exophthalmos.

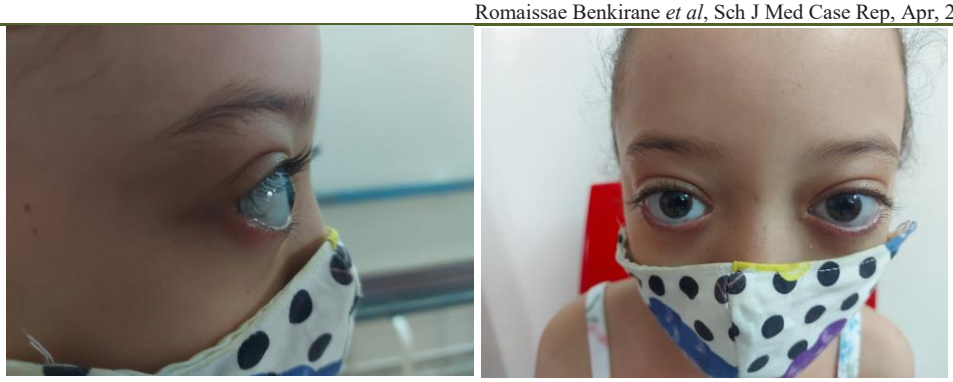

Figure 1: Frontal and profile view of the patient showing bilateral exophthalmos

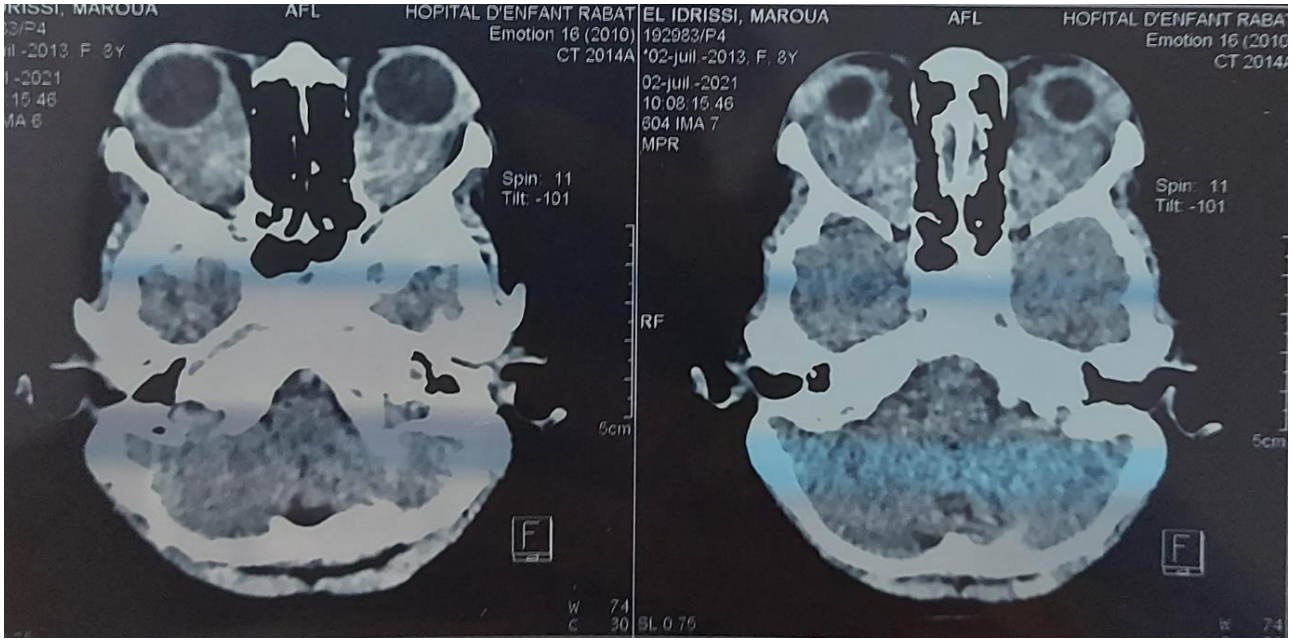

Figure 2: CT scan sections showing a spontaneously hyperdense bilateral intraconal process encasing the globe and the optic nerve

DISCUSSION

Orbital pseudotumors were initially described by Birch-Hirschfeld in 1905. Although they are recognized as idiopathic, there are triggering factors such as microtrauma, microorganisms, or local chronic irritation. +3;4+ Inflammatory pseudotumors represent 8 to 11% of orbital tumors; they occur in all patients regardless of sex, age, or ethnicity, +5;6+ but they are very rare in children. Pediatric forms of inflammatory pseudotumors account for 11.5% of cases. +7+ Clinically, children present with orbital signs, including exophthalmos, periorbital pain, ptosis, limitation of ocular motility, diplopia, eyelid edema, and local inflammatory signs. Ptosis is more common in children. General signs such as headaches, anorexia, altered general condition, and abdominal signs are also noted; these are absent in adults but frequent in children, occurring in more than 50% of cases. +7+ Inflammatory pseudotumors in children are bilateral in 45% of cases, as in our patient, and unilateral in 90 to 95% of cases. +8;9+ Orbital MRI is considered superior to orbital CT scan in these cases, especially with orbital fat suppression. Pseudo-inflammatory tumors present as inflammation of various intraorbital structures: the globe, lacrimal gland, extraocular muscles, orbital fat, and the optic nerve. +10+ The diagnosis of inflammatory pseudotumors is a diagnosis of exclusion, after ruling out differential diagnoses, which are mainly rhabdomyosarcoma, orbital cellulitis, dysthyroid orbitopathy, leukemia, mucocele, dermoid cyst, lymphoma, Wegener's granulomatosis, sarcoidosis, etc. Hence the need to perform laboratory tests, which in the case of inflammatory pseudotumor come back negative, except for hypereosinophilia, elevated erythrocyte sedimentation rate and CRP, and sometimes elevated antinuclear antibodies.+11+ Orbital biopsy with pathological examination is performed to confirm the diagnosis; it reveals a non-specific inflammatory infiltrate composed of macrophages, mature lymphocytes, eosinophils, and occasionally increased density of the fibrovascular stroma.+12+ The diagnosis of an inflammatory pseudotumor in children is based on a combination of clinical, biological, radiological, and histological evidence, allowing the elimination of differential diagnoses. Treatment is essentially based on

systemic oral corticosteroid therapy: 1-1.5 mg/kg/day, resulting in rapid regression of pain and exophthalmos within 48 hours of treatment, as in the case of our patient. Treatment is effective in 75% of cases. +13+ For chronic forms with fibrosis resistant to corticosteroid therapy, or in the presence of a contraindication to treatment, or in cases of recurrence, low-dose radiotherapy may be proposed. +3;14;15+ The use of immunosuppressants combined with corticosteroids, especially azathioprine, is indicated when long-term corticosteroid therapy is necessary, in order to avoid its side effects. As in the case of our patient, treatment may be maintained for more than 6 months. +16+

CONCLUSION

Inflammatory pseudotumors are rare in children and are occasionally bilateral. It is a benign condition for which clinical and paraclinical evaluation must be performed urgently to rule out rhabdomyosarcoma, which requires urgent management, and to initiate corticosteroid therapy, which constitutes the mainstay of treatment.

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