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Idiopathic Facial Aseptic Granuloma: Usefulness of Cutaneous Ultrasound☆

Granuloma aséptico facial idiopático. Utilidad de la ecografía cutánea

To the Editor:

Idiopathic facial aseptic granuloma (IFAG) is a benign condition that affects children. It was first described in 1999 and named pyodermite froide du visage, due to its similarity with an abscess with few inflammatory signs.[1] Subsequently, in 2001, Roul et al.[2] gave it its current name. It is characterized by the appearance of 1 or several asymptomatic erythematous-violaceous nodules that arise in the cheeks, with no predisposing factors.[3] We present a case of IFAG, paying particular attention to the ultrasound findings that, associated with compatible clinical manifestations, allow us to reach a correct diagnosis without resorting to unnecessary procedures.

The patient was a healthy, 2-year-old boy who was seen for 2 asymptomatic facial lesions that had arisen 4 months earlier. Physical examination revealed 2 relatively firm, painless erythematous-violaceous nodules in the left cheek and right lower eyelid, measuring respectively 1 cm and 3 cm in diameter (Fig. 1). The nodules did not fluctuate. There were no palpable locoregional lymph nodes.

The parents did not report bleeding, ulceration or drainage of purulent fluid, and there was no history of insect bite or trauma. At another center, cultures performed for bacteria, fungi, and mycobacteria were negative, and treatment with oral erythromycin and topical metronidazole had been precribed.

Skin ultrasound (18 MHz transducer) of the nodule in the left cheek revealed a well-defined, hypoechoic dermal lesion measuring 1.37 × 0.24 cm, with no posterior enhancement. No Doppler signal was observed and the lesion did not contain calcium deposits (Fig. 2).

A diagnosis of IFAG was made based on the medical history, physical examination, and ultrasound findings. The lesions presented a progressive improvement (Fig. 3) and resolved spontaneously after 8 months.

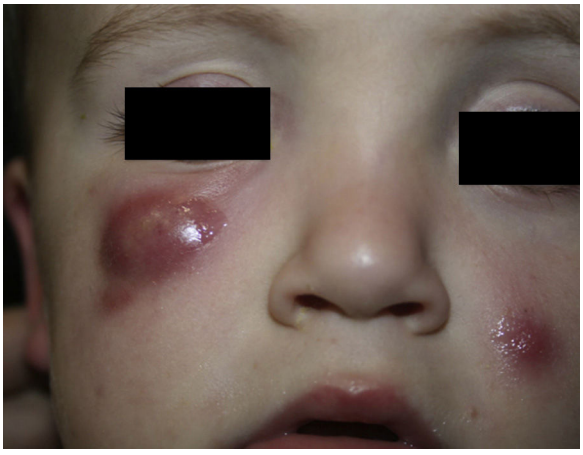

Figure 1 Asymptomatic erythematous-violaceous nodular lesions in the lower right eyelid and in the left cheek.

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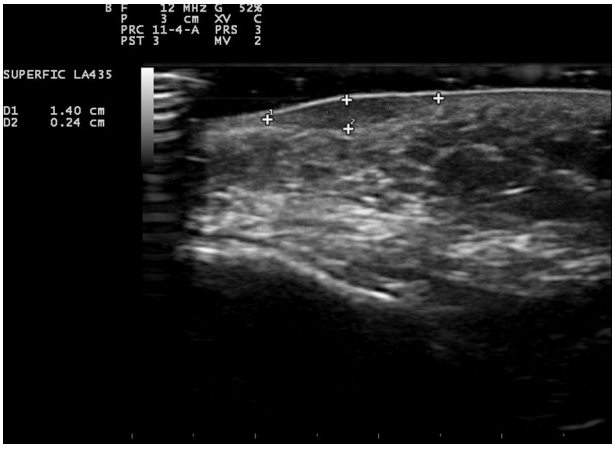

Figure 2 Ultrasound study of the nodule in the left cheek showing a well-defined oval hypoechoic dermal lesion containing no calcium deposits.

IFAG is more common in girls, and the mean age at presentation is 42.3 months.[4] In 90% of cases the lesions are single and arise in the triangle between the labial commissure, the earlobe, and the medial canthus of the eye.[3]

The etiology and pathogenesis of IFAG are unknown. It has been associated with insect bites and trauma, but no relationship with immunodeficiency or serious diseases has been demonstrated. No triggering infection has been detected, as microbiology cultures have been negative and there has been no response to antibiotic treatment. An embryogenic theory has been proposed, in which IFAG is the result of a granulomatous reaction to an embryologic remnant derived from cell migration.[3,4]

Because of the site of IFAGs, their association with recurrent chalazion, and the good response, in some cases, to treatments used in rosacea, some authors have suggested the hypothesis that IFAGs form part of the spectrum of childhood rosacea.[3,5] However, the spontaneous resolution of IFAGs without the need for treatment—something not observed in granulomatous rosacea—would argue against such a hypothesis. Prey et al.[6] reported that children with IFAG have a higher risk of developing childhood rosacea, particularly the ocular form, and they proposed annual ophthalmologic evaluation to facilitate early diagnosis.

Cultures are systematically negative, except in the event of superinfection, which should be suspected if sudden and rapid growth of the lesion is observed.[3,4] Histologically, lesions are characterized by a chronic granulomatous reaction in the superficial and deep dermis, similar to that observed in foreign body or mycobacterial granulomas.[3,4]

Skin ultrasound can be a useful tool to confirm a diagnosis of IFAG. The typical ultrasound pattern is one of a clearly defined, solid, hypoechoic dermal lesion with no calcium deposits.[1] A hyperechoic lesion with a hypoechoic center has only been observed in 2 lesions, 1 of which showed posterior enhancement.[5,7] Doppler study was negative except in 2 cases.[7,8] In our patient, the absence of a Doppler signal may have been related to the fact that ultrasound examination was performed during an advanced phase of the lesion, as Doppler findings in IFAG may depend on the stage of the lesion when the study is performed.

The following conditions should be included in the sonographic differential diagnosis: epidermal cyst, a round anechoic lesion with posterior enhancement and a lateral acoustic shadow; pilomatrixoma, a round dermal lesion with variable central ecogenicity due to the presence of hyperechoic areas corresponding to calcifications, surrounded by a hypoechoic halo; pyogenic granuloma, a poorly defined, oval hypoechoic lesion, with a peripheral nutrient vessel and intense doppler flow internally; infantile hemangioma, a well-defined, homogeneous hypoechoic solid lesion with abundant blood vessels; and abscesses, well-defined hypoechoic or anechoic lesions with intense Doppler flow and anechoic linear fistulous tracts that communicate with the epidermis.[9]

The clinical differential diagnosis is also broad: other benign tumors, such as dermoid cyst, juvenile xanthogranuloma, and Spitz nevus, bacterial, fungal, protozoal, or mycobacterial infections; insect bites; arteriovenous malformations; and paucisymptomatic nodulocystic childhood acne.[3,10]

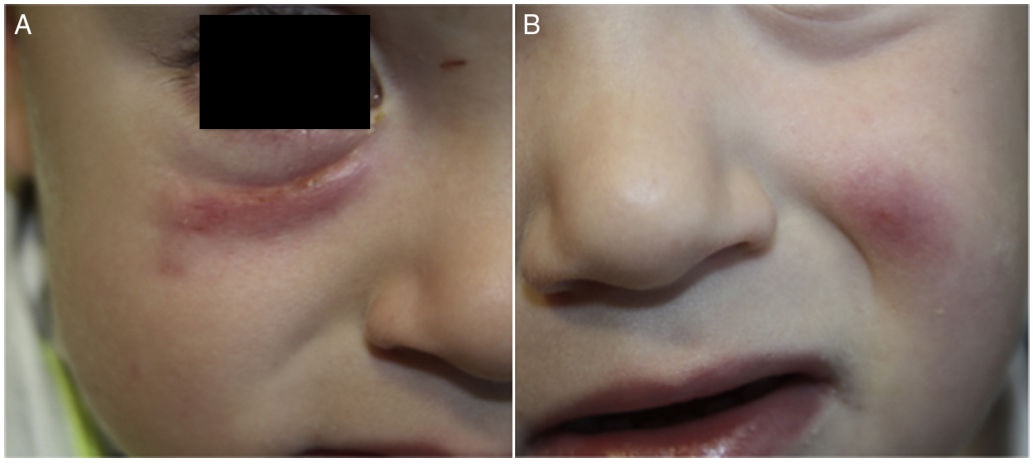

Figure 3 Spontaneous course of the lesions after 4 months. A, Right lower eyelid. B, Left cheek.

The lesions tend to resolve spontaneously after a mean period of 11 months and do not leave a scar.[3] In general, antibiotic therapy is ineffective, although some cases have shown a good response to oral macrolides or topical metronidazole.[7]

We must include IFAG in the differential diagnosis of acquired facial nodules in children. The medical history and the clinical, microbiologic, and ultrasound findings enable us to make an early diagnosis and to avoid unnecessary aggressive procedures.

Conflicts of Interest

The authors declare that they have no conflicts of interest.

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Lymphangioma-like Kaposi sarcoma☆

Sarcoma de Kaposi de tipo linfangiomatoso

To the Editor:

Lymphangioma-like Kaposi sarcoma (LLKS) is a rare histologic variant of Kaposi sarcoma that can present as any of the 4 known clinical variants. LLKS is a vascular neoplasm that develops secondary to infection by human herpesvirus type 8 (HHV-8), which is also known as the Kaposi sarcoma virus. Clinically, it can present with the usual manifestations, namely, patches, plaques, or nodules. However, in some cases, it presents as blisters that may be confused with bullous skin disease.

Case Description

The patient was an 80-year-old man whose history was unremarkable. He presented with raised erythematous, oval plaques measuring 1-3 cm in diameter that had first appeared 5 years previously. The plaques occasionally coalesced and were found on the upper and lower limbs and lower back. The lesions had gradually increased in number and size, although they were neither painful nor pruriginous. The physical examination revealed flaccid blisters (1 cm) containing serum (Fig. 1). Treatment with various topical options had been unsuccessful.

A complete laboratory workup including complete blood count and biochemistry revealed iron-deficiency anemia. Serology testing for HIV was negative.

Histopathology revealed that the epidermis was conserved and highlighted a proliferation of anastomosed vascular spaces that occupied the complete thickness of the dermis, dissected the collagen bundles, and surrounded cutaneous muscles and adnexa. No blood was identified in these structures. Clusters of lymphocytes and plasma cells were common in the stroma. Closer examination revealed that the vascular channels were lined with a layer of flattened endothelial cells and that there was no atypia or mitosis. Immunohistochemistry showed that tumor proliferation was positive for the endothelial markers CD31 and CD34 and for the lymphatic marker D2-40. It also showed clear nuclear staining for latent nuclear antigen 1 of HHV-8 (Fig. 2).

The patient was diagnosed with LLKS and referred to the medical oncology department. Given the poorly aggressive clinical course and the patient's age, it was decided—after

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