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ADVANCES IN PEDIATRIC RESEARCH

Idiopathic facial aseptic granuloma in a two-year old boy: A case report

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Abstract

Idiopathic facial aseptic granuloma is a chronic, benign, and painless facial nodule occurring exclusively in childhood. A case of idiopathic facial aseptic granuloma located on the right cheek of a 2.3-year old boy is described. Repeated assessment of the skin, using a skin pressurization method, revealed no Demodex spp. parasites. Good progress was made after a long treatment with 1% metronidazole cream.

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Introduction

Idiopathic facial aseptic granuloma (IFAG) is a chronic, benign, and painless facial nodule occurring exclusively in childhood. The condition was first described by French dermatologists in 1999 [1], who gave it the name *pyodermite froide*. Roul et al [2] coined the modern name: idiopathic facial aseptic granuloma.

IFAG is not well known and is rarely reported, only ever in children. Clinically, the disease is characterized by one or more nodules, having a long evolution, red, purplish or otherwise hyper-pigmented, painless or accompanied by tenderness, no pruritus, normal consistency or fluctuant. The location is quite particular: in a triangle-shaped area of the cheek, delimited by the external limit of the orbit, the labial angle, and the ear lobe [3], or more simply on the convex area of the face [4]. Neither comedones nor mycrocysts are noted in IFAG, supporting the differential diagnosis with acne infantum.

Sometimes parents recall a previous trauma or insect bite at the site of the nodule [3]. The etiology of IFAG's remains unidentified, although a few years ago Boralevi *et al*. hypothesized that it is a part of large spectrum of childhood rosacea [2, 5]. Since then a number of papers have been published describing clinical presentations supporting this hypothesis: simultaneous presentations of IFAG and ocular signs of rosacea (especially eyelid chalasia); good response to the same treatment modalities as in rosacea [6]; and histologic similarities between IFAG and rosacea (e.g. folliculitis and perifolliculitis with granulomas surrounded by lymphocytes and plasma cells) [3, 7].

Case report

A 2.3-year old boy was referred to the Dermatology Department for a fluctuant nodule of the right cheek that had appeared three months earlier (Fig. 1) with abscess of the cheek, acne infantum, a traumatic vascular lesion, and tumor of unknown origin. The parents reported that several diagnoses had been made and a number of therapies administered with no improvement. The child had endured many weeks of therapy with systemic erythromycin, azythromycin, topical steroids and antibiotics.

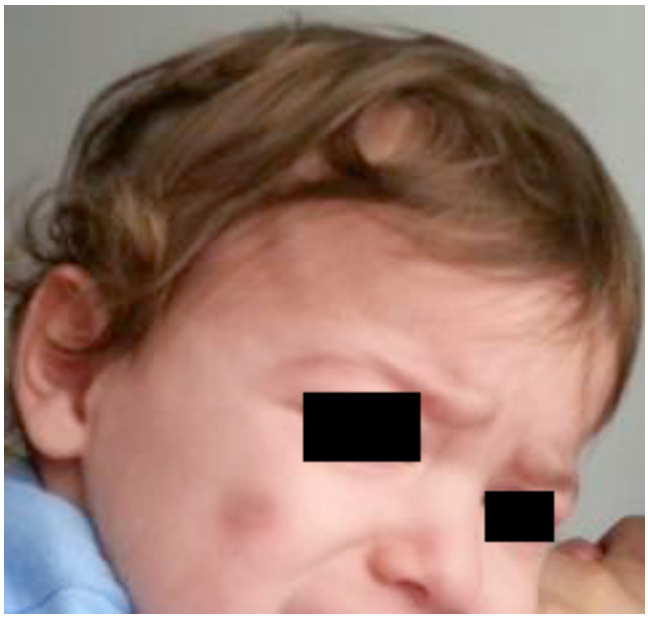

Figure 1. Nodule on the right cheek

Upon physical examination a fluctuant red-brown nodule, 1 mm in diameter, was observed on the right cheek. No ophthalmologic problems were noted, no cervical lymphadenopathy, and no other medical concerns. A small local incision was made, which discharged blood and pus, but cultures for fungus and bacteria were negative. Use of a skin pressurization method [8] revealed no Demodex spp. parasites.

An ultrasound scan was carried out and confirmed a superficial hypoechoic benign nodule of the cheek. The family were unwilling for their son to undergo a punch-biopsy, and a diagnosis of IFAG was admitted.

A two-month treatment regimen involving twice-daily application of 1% metronidazole cream was started. Excellent results were seen with no recurrence after one year.

Discussion

Idiopathic facial aseptic granuloma (IFAG) is a disorder usually occurring during early childhood. Its pathogenesis remains poorly understood.

Clinically simple or multiple nodules should be differentiated from other nodules on the face [9,10]; for example: primary cutaneous lymphoma [11, 12], or facial sporotrichosis [13].

Zhao et al. described a new method to investigate the presence of Demodex spp. parasites by using a skin pressurization method that negates the need to take a biopsy. The method is simple, easy to perform, gives rapid results, and is widely applied in mainland China. It is based on the examination of sebum taken from the nasolabial fold and nasal ala (about 1 cm^2); after squeezing the skin with the thumb, a drop of liquid paraffin is applied to the slide and the sample is examined under a microscope to look for Demodex mites (at any stage of development) [8]. Using this method, we collected sebum from skin around the nodule and from the surrounding skin. The results were repeatedly negative.

IFAG lesions have a long evolution, with no response to antibiotics (whether systemic or topic); topical metronidazole, azelaic acid, nicotinamide associated with oral tetracycline (in children older than 8), or oral erythromycine or metronidazolole in young children (with gastric adverse reactions) [5].

Parents are often unwilling to allow incision and drainage of the nodule due to esthetic concerns, or to administer oral isotretinoin (especially in young children).

This particular case was noteworthy for: (i) a clinical diagnosis of IFAG in a 2.3-year old boy, with no support of histology; (ii) no signs of ocular rosacea, no flushing, and discrete telangiectasia on both cheeks; (iii) its delayed diagnosis, chronic evolution, long term follow-up, and sustained therapy; (iv) no response to antibiotics, despite continued topical and systemic therapy; (v) excellent results obtained by twice-daily applications of 1% metronidazole cream for one month, followed by night administration (occlusion) for one month.

This case report highlights the existence of this kind of lesion, which is not well known and rarely reported. The long evolution of the disease can have a great impact on the patient and their family. It sometimes causes ocular problems, which raises many questions regarding the pathogenic mechanism and treatment.

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