

Should Idiopathic Facial Aseptic Granuloma Be Considered Granulomatous Rosacea? Report of Three Pediatric Cases

Iria Neri, M.D.,* Beatrice Raone, M.D.,* Arianna Dondi, M.D.,*,† Cosimo Misciali, M.D.,* and Annalisa Patrizi, M.D.*

*Dermatology Clinic, Department of Internal Medicine, Aging and Nephrological Diseases, †Pediatric Unit, Department of Gynecologic, Obstetric and Pediatric Sciences, University of Bologna, Bologna, Italy

Abstract: Idiopathic facial aseptic granuloma (IFAG), or *pyodermite froide du visage*, is a skin disease reported only in children and characterized by painless red nodules usually located on the cheeks. Its etiology is still unclear, but some authors considered the possibility that IFAG might be included in the spectrum of granulomatous rosacea (GR). The histopathological features of IFAG and GR are quite similar, showing perifolliculitis, granulomas, folliculitis, and lymphocytes and plasmacells around epithelioid histiocytes. In the present article, we discuss three cases in which an association between a facial nodule, compatible with both IFAG and GR, and recurrent chalazia make us support the hypothesis that IFAG should be considered as GR.

Idiopathic facial aseptic granuloma (IFAG) is a dermatological entity that occurs specifically in children. In the French literature, it has also been named *pyodermite froide du visage*, because it resembles a cold abscess of the face (1). It is characterized by chronic, painless, red or purplish nodules with a soft or elastic consistency. IFAG are usually located in a particular triangle-shaped cheek area, delimited by the external limit of the orbit, the labial angle, and the ear lobe, as shown in Fig. 1 on the left (1,2).

No particular predisposing factors, family history, or associated clinical features have been identified for this dermatosis (2). Ultrasound examination shows a well-demarcated, hypoechoic, dermal lesion (2). IFAG should be carefully differentiated from localized infectious

pyodermas, nodulocystic acne, pilomatrixoma, cutaneous leishmaniasis, botryomycosis, and pyogenic granuloma (2). A well-defined therapeutic approach has not been established; some lesions heal spontaneously, but in some cases, antibiotic therapy is administered, and rarely surgical excision is required (2).

IFAG's etiology is still unclear, but an interesting hypothesis was suggested a few years ago by Boralevi et al, who proposed that this kind of lesion might be part of the spectrum of childhood rosacea (2). Some authors have considered this possibility after observing a simultaneous presentation of IFAG and eyelid chalazia (3,4), which are well-known ocular manifestations of rosacea (5), whereas others regard IFAG to be a granulomatous form of rosacea (6).

Address correspondence to Annalisa Patrizi, M.D., Clinica Dermatologica, Via Massarenti 1, 40138 Bologna, Italy or e-mail: annalisa.patrizi@unibo.it.



Figure 1. The figure shows a 20-mm raised red nodule of the cheek and a chalazion of the inferior eyelid (left) and improvement after therapy (right). The dashed triangle on the left represents the area where most idiopathic facial aseptic granuloma lesions appear.

To evaluate the hypothesis that IFAG should be considered as granulomatous rosacea, we describe three cases examined in our Pediatric Dermatology Outpatient Unit at the University of Bologna.

CASE 1

A 2-year-old girl was referred to our department for an asymptomatic nodule of the cheek; the nodule had appeared 5 months before, and it had not improved after treatment with topical steroids and antibiotics. The parents also reported recurrent chalazia. Upon physical examination, a 7- to 8-mm red nodule of the left cheek and a chalazion of the left inferior eyelid were present. A cheek ultrasound scan revealed a 9- by 3-mm superficial hypoechoic benign lesion. The facial nodule was compatible with an IFAG, but because of the association with chalazia, a diagnosis of granulomatous rosacea was made. Clearing was obtained using systemic clarithromycin at a dose of 15 mg/kg twice a day for 2 weeks and topical metronidazole, which was applied for 3 months. At 1-year follow-up, no relapses had occurred.

CASE 2

A 2.5-year-old girl was examined in our clinic for a nodule on the left cheek; it had appeared 4 months before and had not improved after administration of systemic azithromycin and topical clindamycin. A chalazion of the left inferior eyelid had also appeared 2 weeks before the nodule. Dermatologic examination of the left cheek revealed a 15-mm solitary red nodule with an elastic consistency. An ultrasound scan showed a 20- by 15-mm hyperechoic lesion with a hypoechoic centre, peripheral vascular signals, and perilesional edema. The cheek nodule was compatible with IFAG or granulomatous rosacea; the presence of chalazia led us to the second diagnosis. The child was treated with a 2-month cycle of clarithromycin (15 mg/kg twice a day) that led to clinical healing and no relapses in the following year.

CASE 3

We examined a 20-month-old boy for an 8-month history of an asymptomatic facial nodule and recurrent

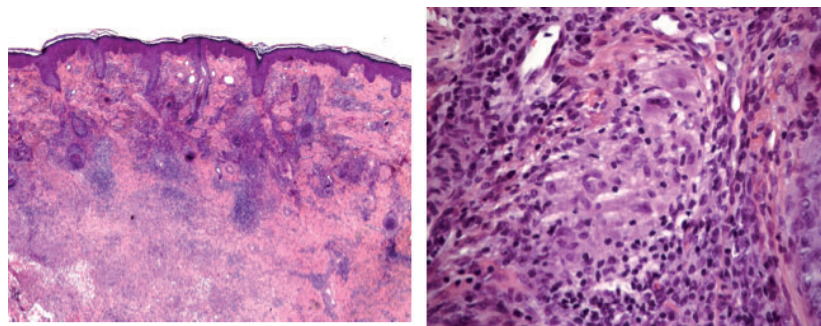


Figure 2. Histologic examination: on the left, perifolliculitis, granulomatous folliculitis (hematoxylin and eosin (H&E) $\times 4$); on the right, lymphocytes and plasma cells around collections of epithelioid histiocytes (H&E $\times 40$).

chalazia occurring since the age of 11 months. Dermatologic examination revealed a 20-mm raised red nodule of the right cheek and a chalazion of the right inferior eyelid (Fig. 1, left). Ultrasound examination showed a hypoechoic central lesion with hyperechoic peripheral signals. Punch 3-mm biopsy showed perifolliculitis, granulomas, folliculitis, and lymphocytes and plasma cells around collections of epithelioid histiocytes (Fig. 2), supporting a diagnosis of IFAG and granulomatous rosacea. Tissue cultures were also performed, but no microorganisms could be identified. Because of the presence of the chalazion, we decided on granulomatous rosacea and suggested a 3-week therapy with systemic erythromycin (50 mg/kg three times per day). Despite partial improvement, we performed an incision and drainage of the nodule, and antibiotic therapy was continued for five more weeks, leading to an almost complete regression of the lesion (Fig. 1, right).

DISCUSSION

Granulomatous rosacea is a rare variant of childhood rosacea, appearing as hard cutaneous papules and nodules in relatively normal-appearing skin and not requiring other rosacea signs, such as flushing and telangiectasia, for diagnosis (7). The pathogenesis of childhood rosacea is unknown, it may result from the influence of environmental, climatic, or infectious factors in genetically predisposed children (6). Ophthalmologic signs often appear in patients with rosacea. In a recent 20-patient pediatric case series, ocular signs were present in most of them (70%) and frequently preceded the cutaneous signs; meibomian gland inflammation, manifesting as chalazion, was the most common (4). The treatment of choice for adult granulomatous rosacea is oral isotretinoin, but safety and efficacy in children have not been tested for this form, although other valid treatment options exist, such as tetracycline, and the use of a certain drug varies from practice to practice. Topical metronidazole, azelaic acid, or nicotinamide can be used in children, but oral tetracycline in children aged 8 and older and oral erythromycin or metronidazole in younger children can be necessary (6).

As Al Dhaybi et al suggested, IFAG might be considered to be granulomatous rosacea (1,6). The histopathologic appearance of granulomatous rosacea is

reported to be a suppurative folliculitis that becomes granulomatous perifolliculitis (7). Rosacea histologic features consist of well-circumscribed collections of epithelioid histiocytes, mostly in perifollicular locations, granulomas surrounded by lymphocytes, and few plasma cells. Sometimes, rupture of infundibular epithelium, collections of neutrophils, and necrosis within granulomas are also present. Telangiectases in the papillary dermis are strongly evident (8). The description of the histologic aspects of rosacea and IFAG highlights the similarities between the two diseases.

In conclusion, the clinical (painless red or purplish cutaneous nodules appearing on relatively normal surrounding skin) and histologic (folliculitis and perifolliculitis with granulomas surrounded by lymphocytes and plasma cells) features that are reported in this small case series seem to further support the strong relationship that has already been noticed between the two previously discussed diseases. Moreover, the association between ocular findings and the response to medications which are effective in treating rosacea adds weight to should reinforce the hypothesis that IFAG should be considered to be a form of granulomatous rosacea.

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