

A child with idiopathic facial aseptic granuloma

Müzeyyen Gönül¹, Şensu Tufan¹, Deniz Ateş Özdemir², Fatma Aksoy-Khurami²

Departments of ¹Dermatology and ²Pathology, University of Health Sciences Diskapi Yıldırım Beyazıt Training and Research Hospital, Ankara, Turkey. E-mail: sensutufan@hotmail.com

Received: 24th September 2017, Revised: 26th November 2017, Accepted: 24th December 2017

Idiopathic facial aseptic granuloma (IFAG) is a rare skin disorder characterized by one or few asymptomatic red or purplish papules or nodules that occur specifically on cheeks or eyelids of children.^{1,2} The etiology of IFAG is unclear.² The disease heals spontaneously within a few months, up to 1 year, without a scar. ¹ We present a peculiar case of IFAG with multiple nodules associated with an eyelid chalazion in a 3-year-old boy.

A 3-year-old boy presented to our outpatient clinic with papules and nodules located on his right cheek. The lesions occurred on his right cheek 9 months before he was presented and did not respond to many topical and systemic antibiotics, topical steroids and intramuscular triamcinolone acetonid therapies. His parents defined that a hordeolum recurred two times on the right eyelid. There was no history of bleeding, purulent discharge or ulceration and trauma or insect bite before it. Dermatological examination showed a few about 1 cm sized, painless, separate, erythematous-violaceous nodules located on the right cheek and a chalazion of the right upper eyelid (Fig. 1). There were no accompanying blackheads, telangiectasia or flushing. Ultrasonographic examination of the nodules on the right cheek revealed a well-limited, hypoechoic lesion without calcium deposits. Histopathological examination showed dermal inflammation with histiocytes, scattered giant cells, lymphocytes, neutrophils extending into deep dermis (Fig. 2). Leishmania amastigotes by Giemsa staining and demodex mites could not be seen in histopathological examination. No specific microorganism was detected with Ehrlich-Ziehl-Neelsen, Periodic Acid Schiff and Gram staining. IFAG was diagnosed on the basis of medical history, physical examination, ultrasound and pathological findings. When parents were informed about the illness, they preferred untreated follow-up. After six months,

the lesions gradually and spontaneously healed without scars (Fig. 3).

IFAG characterized by generally asymptomatic facial papules or nodules originally was described as “pyodermitis frigida” because of clinical similarity to pyoderma or cystic lesions.^{1,3} It is usually located within a triangle limited by the external corner of the orbit, the labial angle, and the ear lobe.² Blackheads and telangiectasia are not associated with this disease.³ Satta et al.³ reported that mean onset age of the disorder was 12 months. The disorder was seen slightly higher in girls.³ In most cases of IFAG it is mostly solitary and multiple lesions are rarely seen.³ Our patient was a 3 year-old boy and he had 4 nodules.

The etiology of IFAG is unclear.² However, there have been several hypothesis and one of them is a possible granulomatous response to an uncertain embryologic remnant. ⁴ Another hypothesis is that IFAG might be a granulomatous form of rosacea because some cutaneous findings are similar to granulomatous rosacea (GR) and association with facial telangiectasias, recurrent chalazions and conjunctivitis.² Prey et al.⁵ reported that the children with IFAG had a greater risk of developing rosacea, especially ocular rosacea, and prevalence of rosacea was 42.1% in their IFAG population. But some authors think that IFAG is a different disease from GR because it has tendency of spontaneous regression and more diffuse and deeper infiltration than GR.³ He has followed for his recurrent chalazion.

The differential diagnoses of IFAG include much dermatological entity such as nodular infantile acne, pediatric rosacea, pyoderma, dermoid cyst, leishmania, xanthogranulomas, tuberculids, papulonodular skin lesions of Crohn’s disease, vascular malformations and insect bite.^{2,3} Skin ultrasound may be useful for diagnosis and it shows a well-limited, hypoechoic lesion



Fig. 1. A few, about 1cm sized, painless, separate, erythematous-violaceous nodules located on the right cheek and a chalazion of the right upper eyelid.

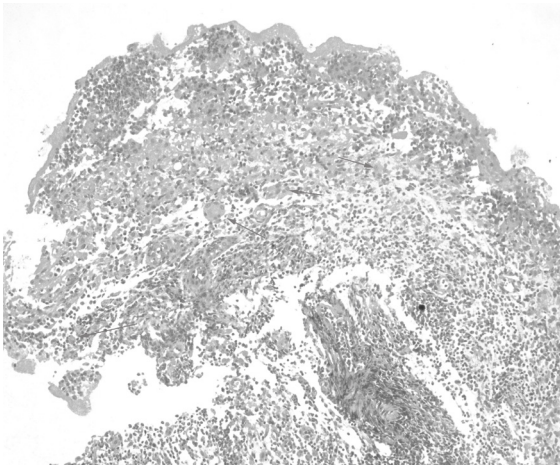


Fig. 2. Polymorphic inflammatory infiltrate which is composed of histiocytes, lymphocytes, neutrophils and eosinophils (giant cells were marked with arrows) (H&E X 100)

without calcium deposits.¹ Cultures obtained from purulent material produce negative results for bacteria, fungi except superinfection.⁴ Histopathological examination of IFAGs shows diffuse infiltrate of histiocytes, giant cells, lymphocytes and neutrophils in the deep dermis.³ Clinical, histopathological and



Fig. 3. After six months, the lesions spontaneously healed without scars

ultrasounographic findings of our case were compatible with IFAG.

The diagnosis of IFAG can be missed as in our case because of its rarity. The dermatologist should keep this disease in mind in a child with a nodular lesion on the face.

Key words: *aseptic granuloma, children, face, nodules.*

REFERENCES

1. Zitelli KB, Sheil AT, Fleck R, Schwentker A, Lucky AW. Idiopathic facial aseptic -granuloma: Review of an evolving clinical entity. *Pediatr Dermatol* 2015; 32: e136-e139.
2. Neri I, Raone B, Dondi A, Misciali C, Patrizi A. Should idiopathic facial aseptic granuloma be considered granulomatous rosacea? Report of three pediatric cases. *Pediatr Dermatol* 2013; 30: 109-111.
3. Satta R, Montesu MA, Biondi G, Lissia A. Idiopathic facial aseptic granuloma: Case report and literature review. *Int J Dermatol* 2016; 55: 1381-1387.
4. Boralevi F, Léauté-Labrèze C, Lepreux S, et al; Groupe de Recherche Clinique en Dermatologie Pédiatrique. Idiopathic facial aseptic granuloma: A multicentre prospective study of 30 cases. *Br J Dermatol* 2007; 156: 705-708.

5. Prey S, Ezzedine K, Mazereeuw-Hautier J, et al.; Groupe de Recherche Clinique en Dermatologie Pédiatrique. IFAG and childhood rosacea: A possible link? *Pediatr Dermatol* 2013; 30: 429-432.