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Unilateral periocular Granulomatous Rosacea

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Letter to the Editor:

Granulomatous Rosacea (GR), first described in 19701, is thought to be a peculiar variant of rosacea thanks to the presence of granulomas at histopathological examination, although some authors sustain that it is a distinct histological variant found among most of the clinical spectrum of rosacea2 . GR is mostly reported in middle aged women and may present centro-facial localization3. Its histology is similar to that of Periorificial Granulomatous Dermatitis (PGD), although the latter is more common in the pediatric population4.

We present a peculiar case of a unilateral periorbital GR. A 22-year-old male in good health conditions had a dermatological consultation, complaining of a periorbital non-itching rash on the left side of his face that had been present for a few weeks. He denied any medical history or having used any topical or oral medication prior to the eruption's onset. At clinical examination (Figure 1) papules and pustules on an erythematous background were found on the left periorbital area, whereas the contralateral side was unaffected. Mucous membranes were uninvolved and there was no palpable lymphadenopathy. Dermoscopy examination (Figure 2) showed widespread linear vessels arranged in a polygonal network and follicular opening containing whitish-grey plugs.

Histology revealed moderate acanthosis with dilation of the hair follicles infundibula with some keratin plugs. Telangiectatic vessels were visible in the dermis, together with prominent perivascular lymphoplasmacytic infiltrate and perifollicular epithelioid granulomas (Figure 3). Stains for acid-fast bacilli (i.e. Ziehl-Neelsen) were negative. No abnormalities emerged at the serological routine exams.

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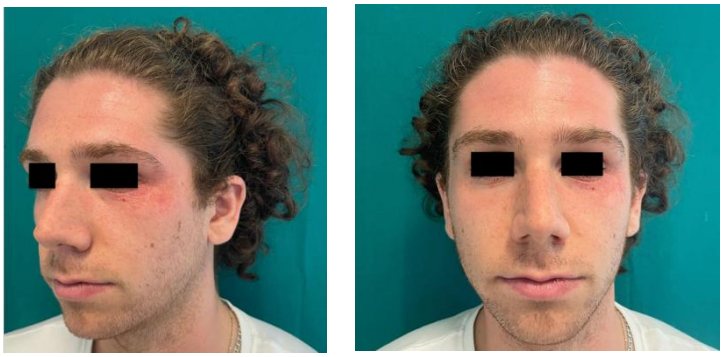

Figure 1.A and 1.B: papules and pustules on an erythematous base involving the left periorbital area.

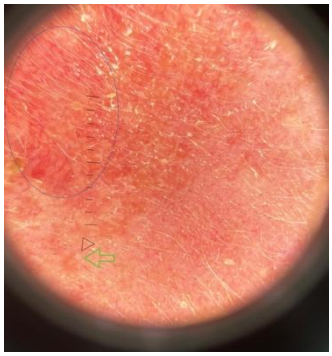

Figure 2: Dermoscopy image showing linear vessels arranged in a polygonal network (blue circle) and follicular openings containing whitish-grey plugs (green arrow).

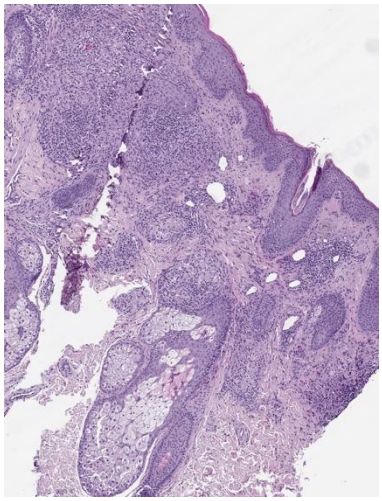

Figure 3. Dilated blood vessels in the dermis (telangiectasia) and perifollicular epithelioid granulomas.

Clinical, dermoscopy and histology findings were consistent with GR. The patient was treated with topical metronidazole 5% ointment every night for 3 months.

Three months later, the left-sided periorbital eruption had improved with marked reduction of the erythematous component and persistence of few papules and pustules, but the periorbital area on the right side of his face was now showing a similar erythematous papulo-pustular eruption. The patient was placed on additional oral therapy consisting of oral doxycycline (40 mg/die for three months).

GR is thought to be distinct from other forms of rosacea: it can presents minimal centro-facial erythema, often has periocular lesions and can show asymmetrical distribution3,5,6. Diagnosis is confirmed in the presence of histopathological findings of granulomatous infiltrates7, after ruling out other entities as Periorificial Dermatitis (PD) and sarcoidosis. PD, which can involve the perinasal, perioral, and/or periorbital area, is characterized by erythematous papules and pustules on the face. PD has been erroneously referred to as perioral dermatitis even when lesions appear in locations other than the perioral area. PD is commonly found in young-adult women8, but

there is a granulomatous variant typically observed in pre-pubertal children of darker skin color, called childhood granulomatous periorificial dermatitis (CGPD; also called GPD). This granulomatous variant can be also observed in adults4 and is clinically and histologically similar to GR. Therefore, they can be confused for one another and their distinction and management is controversial.8,9. From a clinical standpoint, both GPD and GR present yellow-brown papules and pustules that are often scaly and erythematous and involve the periorificial areas; adult patients with GPD are more often complain of burning and itching. GR tends to have a chronic course whereas GPD resolves more quickly8.

Our patient presented also telangiectasias, which is a peculiarity of GR. Moreover, dermoscopy showed linear vessels arranged in a polygonal network: a typical finding of rosacea10,11,12.
In addition, he did not report any itching, pain or burning on the affected area, which are commonly reported in GPD. We had to consider histology to make the final diagnosis: it showed telangiectasias in the dermis together with prominent lymphoplasmacytic infiltrate and perifollicular epithelioid granulomas; GPD usually lacks telangiectasias.

As said before, the distinction between GPD and GR has been controversial over the years. Some authors view GPD and GR as variants of the same disease in different age and ethnic groups, whereas others suspect that cases initially diagnosed as GPD (e.g. they lacked telangiectasias at histology), but later reclassified as GR, were actually early forms of GR9. Adding to this controversy, one study reviewed biopsies from a large group of patients with rosacea and concluded that GR is not a clinical subtype of rosacea per se, but a distinct histological variant, which can be found among most of its clinical spectrum2. Moreover, the 2017 update by the National Rosacea Society Expert Committee of Standard classification and pathophysiology of rosacea13 does not mention any longer GR as a clinical variant (the

previous version did), and this seems to corroborate this hypothesis.
In conclusion, differentiating between GR and other diseases such as sarcoidosis, PD and especially GPD can be challenging, therefore we must consider clinical and histopathological correlates together.

Dermoscopy can be helpful, given its rising role in aiding diagnoses also in non-tumoral pathologies. Doubts persist on whether GR represents a clinical variant of rosacea per se.

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