Unexpected Complications: A Case of Rosacea Fulminans in Pregnancy

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PRACTICE **POINTS**

- Rosacea fulminans (RF) is a rare facial dermatosis that can present in pregnant patients.
- Treatment of RF in a pregnant patient requires special considerations because typical therapies are contraindicated in pregnancy.

Rosacea fulminans (RF) is a rare facial dermatosis that typically affects women with a fulminating course that presents with superficial and deep-seated papules, pustules, and nodules, as well as an intense reddish or cyanotic erythema localized to the face. Although the etiology of RF remains unknown, immunologic, hormonal, and vascular factors have been implicated. We describe a case of a 32-year-old pregnant woman presenting with RF. Presentation in a pregnant patient is not commonly reported and requires special consideration to manage.

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Rosacea fulminans (RF) is a rare facial dermatosis characterized by its fulminating course.[1] It presents with superficial and deep-seated papules, pustules, and nodules combined with an intense reddish or cyanotic erythema localized to the face. Furthermore, there is an absence of comedones and involvement of the chest or back.[2] Rosacea fulminans primarily affects women and often is, but not always, proceeded by seborrhea, chronic acne vulgaris, or rosacea. Although the etiology of RF remains unknown, immunologic, hormonal, and vascular factors have been implicated.[3] We report a case of RF in a pregnant patient with a history of mild acne as a teenager that was long ago resolved.

Case Report

A 32-year-old pregnant woman (10 weeks' gestation) presented with a rapidly progressing inflammatory disorder of the face of 1 month's duration. The lesions developed 3 weeks after beginning progesterone therapy (200 mg vaginal suppository) for infertility due to polycystic ovary syndrome. Despite discontinuing progesterone for the last month, the patient's lesions had dramatically worsened (Figure 1). Empiric cephalosporin treatment prescribed by her primary care physician yielded no improvement. Physical examination at the current presentation revealed erythematous nodules and pustules all over the face, coalescing into large thick plaques on the patient's right cheek and chin. Submental nodes were palpable and tender. Based on the initial clinical findings, acne conglobata secondary to progesterone therapy was considered. The patient was given intralesional triamcinolone (2.5 mg/cc) injections to all larger nodules and several blue light treatments.

The injected areas had improved 5 days after the initial visit; however, the chin and right paranasal cheek developed even more nodules and papules coalescing into large plaques. After consulting the patient's obstetrician, prednisone (20 mg once daily) was initiated. Three weeks later, the patient's nodular lesions had improved, but there was a showering of more than

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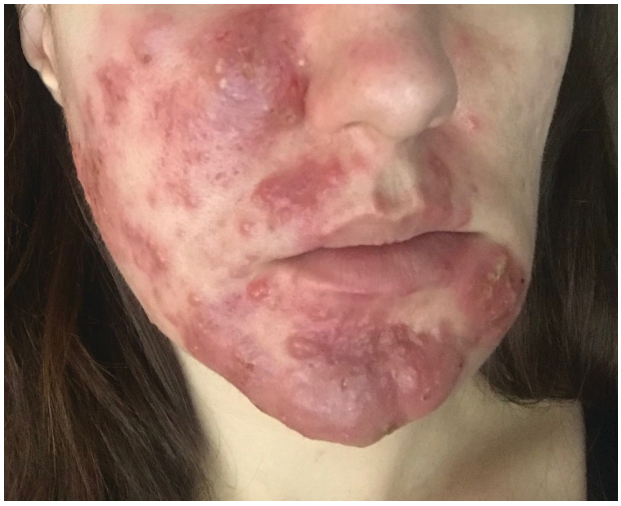

FIGURE 1. Rosacea fulminans in a pregnant woman at presentation (10 weeks' gestation).

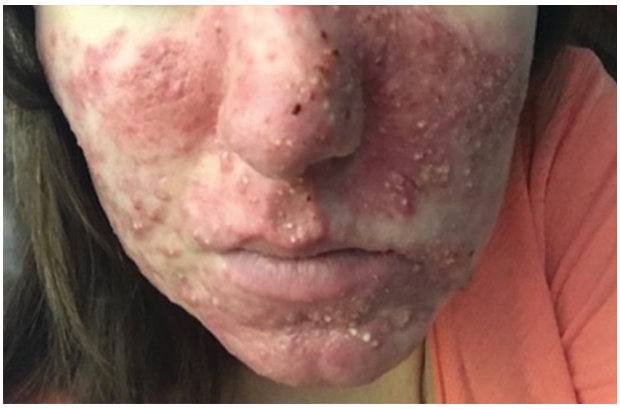

FIGURE 2. Three weeks after starting prednisone, there was a showering of more than 100 pustules and increased general erythema of the entire face due to rosacea fulminans.

100 pustules and increased general erythema of the entire face (Figure 2). Crotamiton cream 10% (every day before noon), ivermectin cream 1% (every night at bedtime), and sodium sulfacetamide cleanser 10% once daily were added to the treatment plan.

At 16 weeks' gestation, there was slight improvement; however, there was still erythema on the entire face with scattered pustules and multiple papules and nodules. Many small ice-pick scars were seen on the cheeks and forehead. No comedones were observed. A punch biopsy of an intact papule showed a prominent inflammatory infiltrate with granulomatous reaction and numerous neutrophils predominantly affecting hair follicles. Based on the clinical presentation and histopathology, a diagnosis of RF was made. Azithromycin (250 mg once daily) and metronidazole cream 0.75% twice daily were added. Two weeks later there were fewer nodules but many papules, edema, and intense erythema. The prednisone dosage was increased to 40 mg once daily. Two weeks later, the patient showed improvement with fewer lesions, less edema, and less erythema. The patient was instructed to finish the azithromycin course and discontinue use. At 28 weeks' gestation, a prednisone taper was started with the intention to reduce the daily dose by delivery.

The patient delivered a healthy girl (birth weight, 1.985 kg) prematurely at 34 weeks' gestation. At 2 months postpartum, the patient's existing lesions continued to spontaneously improve; however, she still had numerous nodules and papules and continued to develop new lesions and form additional scars. Isotretinoin was instituted at 3 months postpartum upon cessation of nursing. Three months later (40 mg/d isotretinoin), the patient was nearly clear. At 8 months postpartum, isotretinoin was discontinued after a course of 150 mg/kg.

Comment

Rosacea fulminans initially was called pyoderma faciale but was later regarded as a severe form of rosacea and was renamed *rosacea fulminans*.[2] According to a PubMed search of articles indexed for MEDLINE using the terms *pregnancy* and *rosacea fulminans* or *pyoderma faciale*, we identified 12 publications reporting 20 cases of RF associated with pregnancy (Table). Although there is no substantial evidence regarding the exact mechanism, these cases indicate that pregnancy can be an exacerbating or causative factor in the pathogenesis of RF.

In addition to pregnancy, RF has been associated with inflammatory bowel disease, thyroid and liver disease, erythema nodosum, and severe emotional trauma. However, no organism has been consistently isolated, and no evidence of family history has been reported.[1] Histopathologic findings are dependent on the stage of disease. Massive infiltrates of neutrophils may be observed in early stages. In older lesions, infiltrates take the form of epithelioid cell granulomas.[2]

Treatment of RF during pregnancy is challenging. Early and aggressive treatment with retinoids, tetracycline antibiotics, antiandrogenic contraceptives, and dapsone is recommended in patients who are not pregnant; these therapies are all contraindicated in pregnancy. Topical steroids can be safely used; however, systemic steroids usually are required to control RF. The use of systemic steroids can only be justified if the risks for intrauterine growth retardation, maternal diabetes mellitus, and hypertension outweigh the benefits of treating this severe disfiguring skin condition.[10] A study by Bakar et al[13] indicated that azithromycin is an effective and safe alternative in the treatment of RF. It has a superior pharmacokinetic profile compared to other macrolides and does not pose increased risks for congenital malformation or miscarriage. Because of the concomitant use of both azithromycin and prednisone, it is not possible to determine which had the larger role in the patient's improvement.

Reported Cases of Rosacea Fulminans Associated With Pregnancy

Reference (year)	Patient age, y	Gestation at presentation (pregnancy number)	Therapy	Outcomes/comments
Current case	32	10 wk (1)	Intralesional triamcinolone, crotamiton cream 10%, ivermectin cream 1%, sodium sulfacetamide cleanser 10%, prednisone, azithromycin, metronidazole cream 0.75%	Improvement with prednisone and azithromycin; complete clearance postpregnancy with isotretinoin treatment
Garayar Cantero et al[4] (2020)	28	13 wk (2)	Permethrin cream 5% monotherapy	Complete clearance; no relapse at 13-mo follow-up
Demir et al[5] (2018)	22	6 wk (1)	Oral amoxicillin–clavulanic acid, wet compresses, and fusidic acid cream	Resolution 1 mo after cessation of treatment
Markou et al[6] (2017)	37	37 wk (4)	Azithromycin, prednisone	Notable improvement after delivery; associated with ocular manifestations
Haenen et al[7] (2015)	38	14 wk	Oral erythromycin	None reported
Fuentelsaz et al[3] (2011)	33	11 wk (1)	Oral azithromycin and topical metronidazole (12 wk); topical metronidazole and topical clindamycin after discontinuation of azithromycin; topical combination of fusidic acid and 0.1% betamethasone used for 1 nodule	Clearance by 6 mo of pregnancy
de Morais e Silva et al[8] (2011)	26	21 wk (2)	Oral prednisone and oral erythromycin stearate	Improved clinical features after 10 d of treatment; severe blepharitis with advanced keratitis and ocular perforation bilaterally
Jarrett et al[1] (2010)	35	NA (2)	Oral prednisolone, oral erythromycin	Slight improvement; termination of pregnancy at 12 wk; prednisolone and isotretinoin used for treatment posttermination
	31	8 (3)	Oral erythromycin	Improvement but not clearance; isotretinoin and prednisolone used for treatment postdelivery
Cisse et al[9] (2008)	32	First 3 wk (1)	Unknown	No improvement throughout pregnancy despite different treatments; pregnancy via in vitro fertilization; isotretinoin used for treatment postdelivery
Ferahbas et al[2] (2006)	31	First trimester (2)	Oral methylprednisolone, wet compresses, fusidic acid cream, and metronidazole cream 0.75%	Resolution and no relapse at 1 y posttreatment
Lewis et al[10] (2004)	28	4 wk (1)	Oral erythromycin, oral prednisolone	Improvement; clinical features similar to low-grade rosacea at 2 mo postpartum; resulted in stillbirth
Plewig et al[11] (1992)	23	First trimester	Topical antibiotics (including erythromycin and clindamycin)	None reported
	33	Last trimester		
	25	First trimester		
Massa and Su[12] (1982)	Total of 6 cases associated with pregnancy; unknown details			

Isotretinoin therapy in our patient led to substantial improvement of RF. Time will tell if the response will be durable. Also unknown is the risk for recurrence with subsequent pregnancies, which has not been reported in the literature. Although it is difficult to confidently say that pregnancy was the inciting factor in this patient's RF, this case certainly provides more evidence for a link between pregnancy and RF.

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