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A CASE REPORT WITH REVIEW OF LITERATURE ON PYODERMA FACIALE IN PREGNANCY – A THERAPEUTIC DILEMMA

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Abstract

Pyoderma faciale is a rare facial dermatosis. It is characterized by a fulminating course of facial inflammation consisting of numerous pustules, cystic swellings and coalescing sinuses in young women. It has to be differentiated from acne and rosacea. The aetiopathogenesis of pyoderma faciale is not yet identified. It has been associated with pregnancy in a few cases. We report a case of a primigravida who presented with sudden onset of pustules and cystic swellings over the face with no prior similar history which was diagnosed as pyoderma faciale. In view of her pregnancy, systemic retinoids which is the treatment of choice was contraindicated and so was treated with tapering doses of oral steroids in combination with topical therapy. There was complete resolution of symptoms with treatment and a good obstetric outcome.

Key words: pyoderma faciale, pregnancy

Introduction

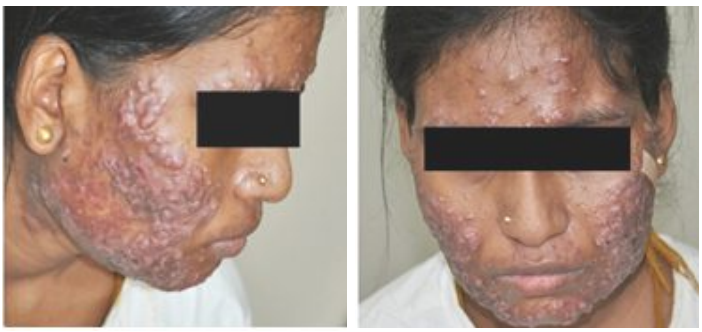
Pyoderma faciale was formerly described by O'Leary and Kierland in 1940.[1] It is a rare facial dermatosis characterized by the sudden onset of severe facial inflammation consisting of numerous pustules, cystic swellings and coalescing sinuses. Plewig et al considered it as an extreme form of rosacea and termed it as rosacea fulminans.[2] But it is not yet clear whether this condition is a variant of rosacea or acne vulgaris or a separate entity. It is not a pyoderma; nor a variant of acne conglobata.[3,4] We report a case of pyoderma faciale in pregnancy due to its rarity and for its therapeutic dilemma.

Case Report

Twenty four years old primigravida, presented at eight weeks of gestation with abrupt onset of pustules, nodules and cystic swellings over the face of three weeks duration. There was no significant past medical history and no history of similar lesions in the past. The lesions were tender and disturbed her sleep. Two weeks prior to presentation to our hospital, the patient was diagnosed to have acne vulgaris and treated elsewhere. As there was no improvement she was referred to us. There is also history of intake of vitamin B complex supplements after the onset of lesions, which aggravated the lesions. She also had hyperemesis gravidarum for which she was admitted to give supportive care.

General and systemic examination was normal. Dermatological examination revealed multiple tender nodules, abscesses, pustules and cystic swellings over the face sparing the central part and temporal region. There were no lesions elsewhere in the body (Figure 1,2).

All investigations were normal. Biopsy was consistent with pyoderma faciale, which showed follicular plugging, dense perivascular and periadnexal infiltrate. Dermis showed granulomatous reaction pattern with infiltrate including neutrophils, eosinophils, lymphocytes, epithelioid histiocytes, plasma cells and multinucleate giant cells. (Figures 5, 6 and 7)



Figs 1,2: Figure 1,2: multiple tender nodules, abscesses, pustules and cystic swellings over the face sparing the central part and temporal region

As there was minimal crusting of the lesions she was given a course of cloxacillin for 1 week. Then she was initiated on topical steroid along with low dose of systemic steroid (Tab Prednisolone 20 mg) and topical clindamycin. Oral prednisolone was maintained at the dose of 20 mg for 4 weeks then it was tapered by 2.5 mg every week. As all of her lesions subsided, oral prednisolone was tapered and stopped over 12 weeks (Fig. 8,9). There was no recurrence of lesions during the follow up for the next 5 months. She had a full term normal vaginal delivery.

Discussion

Pyoderma faciale is an uncommon disorder of unknown etiology that mainly affects post adolescent women, with abrupt onset and disfiguring sequelae if left untreated. There is sudden onset of severe facial inflammation consisting of numerous pustules, cystic swellings and coalescing sinuses. Edema and at times an

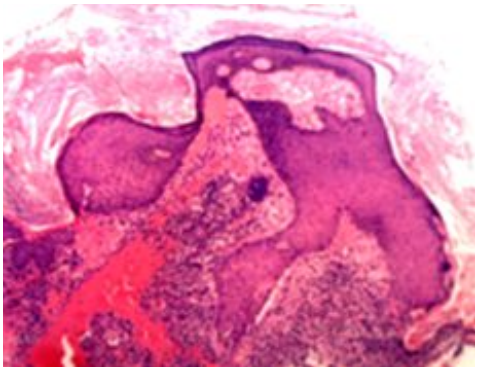


Figure 5: Pseudoepitheliomatous hyperplasia with dense dermal inflammation (H/E;20X)

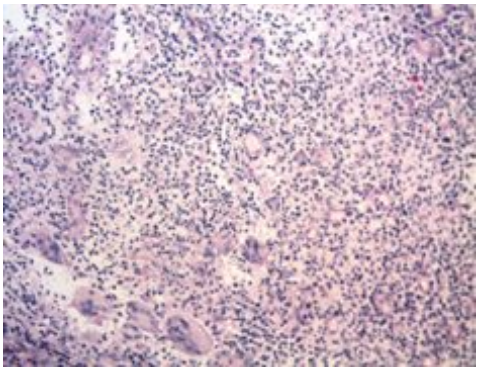


Figure 6: Dense dermal infiltrates of lymphocytes, neutrophils, plasma cells & few multinucleate giant cells (H/E;20X)

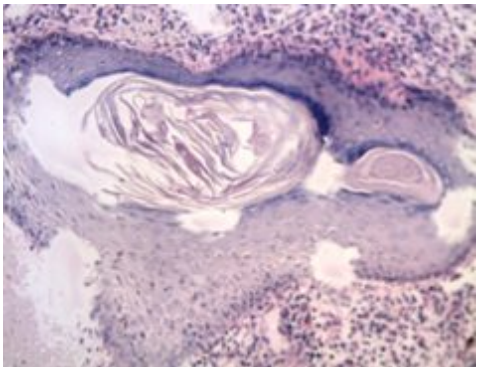
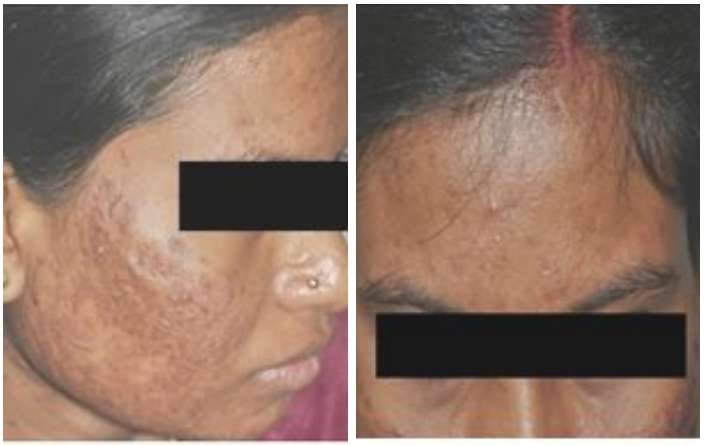


Figure 7: Periadnexal inflammation (H/E;20X)



Figs 8,9: Post treatment

intense reddish or cyanotic erythema accompanies this pustular process. [2,4]

The aetiopathogenesis of pyoderma faciale is unidentified. It is associated with multiple conditions (table 1).[5-12]

No.	Condition
1	Pregnancy[5]
2	Erythema nodosum[6]
3	Crohn's disease[7]
4	Ulcerative colitis[8]
5	interferon 2B and ribavirin therapy for hepatitis C[9]
6	High-dose vitamin B supplements[10]
7	Emotional distress[11]
8	Thyroid disorders[12]

Table 1: Associations of pyoderma faciale

Hormonal imbalance has been proposed in view of its almost exclusive occurrence in women; furthermore, the eruption has been associated with pregnancy in a few cases.[4,5,12] Usually no recurrence of pyoderma faciale is seen except few case reports.[3,12] This condition has to be differentiated from acne vulgaris and rosacea. As compared to acne vulgaris it is abrupt in onset with devastating sequelae of scarring if not treated early. But it does not last more than a year. It is confined to the face and does not arise from comedones. Except for two case reports in males, it occurs exclusively in females.[13, 14] It should also be differentiated from rosacea. There is no consistent history of flushing in cases of pyoderma faciale. Furthermore the features such as pre-existing erythema or telangiectasia of the convex portions of the face are absent. The lesions in pyoderma faciale are characteristically large abscesses and nodules. It is not associated with sun exposure, Helicobacter pylori and Demodex. In pyoderma faciale induration and rhinophyma never develop as a sequelae.[3,4,15]

The diagnosis of pyoderma faciale is often exclusively made based on clinical findings, but can be aided by biopsy.

The differential diagnoses of pyoderma faciale include gram negative folliculitis,acne conglobata, acne fulminans, fungal and mycobacterial infections, iododerma, bromoderma, and neutrophilic dermatosis like sweet's syndrome only affecting the face.[16]

It has been recommended that treatment should begin with potent topical corticosteroids for no more than two weeks, oral prednisolone at 1 mg/kg daily for 1–2 weeks followed by a gradual tapering along with oral isotretinoin at 0.2–0.5 mg/kg daily for three to four months until complete healing occurs. Oral tetracycline antibiotics and dapsone have been found to be effective as in many case reports.[17, 18] But during pregnancy retinoids and tetracylines are contraindicated. Pyoderma faciale is the only indication for topical or systemic corticosteroids in the treatment of rosacea.[18]

To our knowledge, this is the twentieth reported case of pyoderma faciale associated with pregnancy in literature, thus contributing further evidence that pregnancy can aggravate this condition and this is the first case report of the same from India. Previous reported cases of pyoderma faciale (rosacea fulminans) associated with pregnancy and their comparisons with our case are given in the table 2.[4,5,12,19-25]

Most of the cases of pyoderma faciale presented in first trimester. Out of the eighteen cases of pyoderma faciale in pregnancy, where treatment details were available eight cases were treated with oral steroids and topical antibiotics. All of the patients had an improvement with treatment except two cases which includes one case who had persistence of lesions throughout the pregnancy and other case had recurrence of symptoms on tapering of steroids.[21, 23] Out of the eleven cases where obstetric outcome details were available, two had intrauterine death, one

Table 2: Case reports of pyoderma faciale (rosacea fulminans) in pregnancy

Serial no.	Authors	No. of cases	Development of pyoderma faciale(PF)	Treatment given	Outcome of treatment	Outcome of pregnancy
1	Massa MC and Su WP [12]	5	Third trimester / postpartum	Not specified	Improved	Not specified
2	Marks VJ and Briggaman RA [19]	1	Second trimester	Topical antibiotics, intralesional triamcinolone acetonide and prednisolone	Improved	Full term healthy baby
3	Plewig G, Jansen T, and Kligman AM [4]	4	Two cases in first trimester; one case in third trimester,one case developed in postpartum	Topical antibiotics including clindamycin and erythromycin	Improved	Not specified
4	Haugstvedt A and Bjerke JR [20]	1	Not specified	Not specified	Not specified	Not specified
5	Lewis et al[21]	1	First trimester	Prednisolone and erythromycin	Poor response Lesions and recurrence of lesions on tapering prednisolone	Intrauterine death
6	Fehrabas et al[22]	1	First trimester	Oral steroids, surgical drainage, topical antibiotics	Moderate response	Full term delivery
7	Cisse et al[23]	1	First trimester	Topical and oral macrolides, topical metronidazole, amoxicillin, oxacillin and fusidic acid	Skin disease persisted throughout pregnancy	Not specified
8	Jarrett R, Gonsalves R and Anstey AV [5]	3	All three patients first trimester	Prednisolone and azithromycin	First patient had moderate improvement with persistence of symptoms till 2 months postpartum. Second and third patients had complete resolution of lesions.	First case intrauterine death, Second case termination of pregnancy, and third case had a normal vaginal Delivery
9	Fuentelsaz et al[24]	1	First trimester	Oral azithromycin, topical antibiotics and steroids	Resolution by sixth month of pregnancy	Full term normal delivery
10	Fernanda et al[25]	1	Second trimester	Oral erythromycin and prednisolone	Improved	Full term caesarean delivery
11	Current case	1	First trimester	Topical and systemic steroids, Topical and systemic antibiotics,benzoyl peroxide face wash, incision and drainage	Complete resolution of the lesions after 12 weeks of therapy	Full term normal delivery

had medical termination of pregnancy while others had full term delivery. Cisse et al reported a case of pyoderma faciale during pregnancy which was initially thought to be due to administration of follicle stimulating hormone (FSH) and luteinizing hormone releasing inhibitor, but later this was thought to be unlikely since lesions persisted throughout pregnancy even after withdrawal of the drugs.[23] Fernanda et al reported a case of pyoderma faciale in second trimester associated with relentless ocular involvement which ended up with ocular perforation. She was treated with oral erythromycin,

predinisolone and successful corneal transplant. She had a full term normal delivery.[25]

It is imperative to recognize this entity and its importance in relation to pregnancy. It should be diagnosed and treated without delay as the sequelae of scarring can have a negative psychosocial impact on the patient. Our patient received immediate attention and was started on specific therapy, with complete resolution of lesions without much scarring. So it highlights the significance of early diagnosis of this condition and initiation of steroids in the early phase of the disease to bring remission.

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