CASE STUDIES

# An unusual presentation of Wegener's granulomatosis during pregnancy

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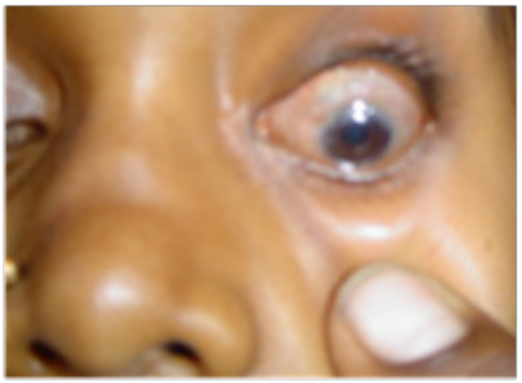
**Abstract**

The occurrence of Wegener's granulomatosis (WG), a multisystem autoimmune disease of unknown etiology, during pregnancy and postpartum is rare. We present here a rare case of a pregnant woman with corneal ulcer that occurred secondary to WG during the postpartum period.

## Case report

A 20-yr-old female primigravida presented to the outpatient department with severe arthritis involving small joints of hands and feet, and a vision threatening (6/60) marginal corneal ulcer in the left eye during postpartum (Fig 1). Although, the patient had a history of recurrent epistaxis in her 5th month of gestation and the occurrence of saddle nose (Fig 1) in the 8th month, she ignored these symptoms. She had a normal full-term delivery and she did not receive any treatment during pregnancy. No history of hemoptysis, reduced urine output, fever, weight loss, weakness on one side of the body, or ear pain was reported. General assessment revealed that the vital signs and the functioning of cardiovascular and neurological systems were normal. The results of laboratory investigations are as follows: Hb-12.2gm/dl,Tc-14800/dl with normal differentials, ESR-120mm/1st hr, urine protein- 75mg/day, negative RF and ANA antibodies, and positive c-ANCA. Chest X-ray examination and urine routine did not report any abnormal findings. She was diagnosed to have vision threatening marginal corneal ulcer due to WG. The patient was treated with intravenous pulse methyl prednisolone (1gm for 3 days) and cyclophosamide (500mg every 15 days for 6 cycles) followed by a tapering dose of methyl prednisolone. Improvement in corneal ulcer was seen following medication. The patient is presently on methotrexate 15 mg/week (as steroid sparing medication) and 5 mg of methyl prednisolone and now the symptoms are minimal.

**Fig 1: Occurrence of saddling of nose and marginal corneal ulcer**



## Discussion

WG primarily affects the upper and lower airways, and the kidneys. Pregnancy is uncommon in women with WG due to the occurrence of the disease in later age group, potential severity of the disease, and the use of cyclophosphamide as a therapeutic agent. The available literature on WG during pregnancy and postpartum consists mainly of case reports.[1] In the present case study, WG was diagnosed only when the corneal ulcer was noted in the postpartum period. Although the symptoms of upper respiratory tract involvement such as saddling of nose and epistaxis

were noted by the patient during the second trimester, she did not consult any physician as the symptoms were minor and did not affect the patient's quality of life. Disease flare-ups are reported more commonly during pregnancy in WG patients. The disease may also occur for the first time during gestation. Active WG at the onset of pregnancy has been correlated with poor pregnancy outcome.[2, 3, 4] But in the present case, the delivery was normal. Similar to the current study, seven cases of newly diagnosed WG during pregnancy have been documented. Additionally, 15 cases with a previous diagnosis of WG, who conceived during remission, have been reported. In patients experiencing disease relapse during pregnancy, treatment is determined on the basis of site of involvement, the severity of the relapse, and the stage of pregnancy.[5] However, very few cases on postpartum flare have been reported. In the current study, the WG diagnosis was done during postpartum, even though the symptoms were dated back to 2 and 3 trimesters. The study highlights the risk of flaring of WG during gestation and the need for meticulous monitoring of clinical symptoms in such patients.

**Competing interests**
The authors declare that they have no competing interests.

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