Vitiligo aggravated after major surgery for Crohn’s disease: a consequence of severe surgical stress?

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Vitiligo is an autoimmune skin disorder characterized by acquired white patches of skin and overlying hair as a consequence of loss of melanocytes and cutaneous pigment from the involved areas [1]. It appears mainly in patients with autoimmune disorders, although it has also been rarely described in patients with inflammatory bowel disease either before or after the establishment of diagnosis of bowel disease [2,3]. Some recent reports claim that vitiligo can also occur during treatment with anti-tumor necrosis factor (TNF)-α factors (infliximab or adalimumab) in patients with either Crohn’s disease (CD) [4] or ulcerative colitis [5].

We describe hereinafter the case of a patient with severe CD who developed extensive vitiligo, following a major surgical procedure performed for complicated CD. A woman, aged 44, developed CD of the small and large bowel at the age of 17. During the subsequent years, the disease was running with exacerbations of mild-to-moderate severity. The main clinical symptom was abdominal pain largely due to incomplete bowel obstruction and signs of impaired nutritional status. She also described vitiligo, covering a very small area of her hands. There was no family history of vitiligo and no known history of an autoimmune disorder. Maintenance treatment included only mesalamine (2.4 g/d) as azathioprine was not tolerated. In April 2010, she underwent right hemicolectomy with resection of 25 cm of the terminal ileum plus end-to-end anastomosis, because of the development of clinical signs of obstructive ileus. On May 25, 2010, she suddenly developed clinical and laboratory signs of acute peritonitis. The WBC was 14,490, the CRP 38 (nv: <3 mg/L) and the Hct 32%. Abdominal CT confirmed the clinical diagnosis of generalized peritonitis due to perforation and leakage at the site of the previous anastomosis. Careful lysis of the abundance of adhesions was performed along with resection of a small part of the large intestine (5 cm) and ileum (10 cm). A lateral anastomosis plus loop ileostomy proximally to anastomosis was performed. Subsequently, she developed septic shock and was transferred to the emergency unit. Total parenteral nutrition plus antibiotics were applied. She gradually improved and finally, she was discharged from the emergency unit after 20 days. Soon after, she noticed large areas of different sized achromatic patches occupying most of her body’s skin, localized mainly in her face, upper and low extremities and trunk (Fig. 1). Cutaneous biopsy showed a typical picture of vitiligo (i.e., absence of melanocytes, increased number of Langerhans’ cells, epidermal vacuolization, T cell inflammatory infiltrate, and neural alterations). The course of CD following operation was satisfactory with the patient’s main problem being her poor nutritional status. Up to September 2012, the course of vitiligo also remained unaffected. She is on immunomodulating diet (MODULEN IBD) with satisfactory results. So far, no treatment for vitiligo has been applied.

This patient presents some interesting peculiarities which include the sudden aggravation of the pre-existing skin disorder after the stressful major surgical event, and the extremely large area of her body involved by vitiligo in the absence of both family history of vitiligo and underlying autoimmune disorders. We have no obvious explanation for the appearance of this extensive skin lesion. In spite of recent findings implicating oxidative stress and genetic and immune factors in the pathogenesis of vitiligo, it remains largely obscure. It is possible however, that both oxidative stress and psychological stressful events (in our case the stressful event of the major surgical operation) equally contributed to the appearance of this skin lesion. In conclusion, we suggest a possible association between severe surgical stress and the aggravation of pre-existing vitiligo in a patient with CD.

References

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