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Non-Gestational Impetigo Herpetiformis

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Impetigo herpetiformis (IH) is a rarely seen disorder with pustular eruptions occurring particularly in pregnant women during the last trimester of pregnancy. The disease is characterized by extensive pustular eruptions arranged in groups or solitary with symmetrical localizations particularly on flexural regions (1, 2). The etiology is unknown. Although some authors have suggested that IH is actually pustular psoriasis occurring during pregnancy, it has also been reported in male patients and in woman who underwent thyroidectomy and in woman with hypocalcemia. In the literature, there are some reports about the triggering role of hypoparathyroidism and hypocalcemia in IH (3-6).

The aim of this study was to present a nonpregnant female IH case with polyglandular syndrome.

A 41-year old female patient was admitted to the Outpatient Dermatology Clinic due to complaints of skin eruptions on her trunk. The patient had pustular eruptions on her trunk for one week and alopecia for 10 days. In addition, she had contractions in upper and lower extremities for 20 years and amenorrhea since 1986. On physical examination, the general appearance of the patient was not good. She was conscious and cooperative. Chvostek's and Trausseu's signs were positive. Her temperature at time of hospital admission was 38.5 ˚C.

Dermatological examination revealed desquamations and pustules on abdominal, back, gluteal, genital regions and upper parts of thigh. There were uninvolved areas between lesions. Widespread ulcerations with purulant material were seen on mucosal surface of mouth, hard palate and tongue. Her lips were fissured and she had diffuse alopecia (Figure 1).

Laboratory examination results were summarized as follows: Hb: 10 mg/dL, sedimentation: 35 mm/hour, glucose: 95 mg/dL, Ca: 4.1 mg/dL, P: 6.7 mg/dL, total protein: 4.7 mg/dL, albumin: 2.6 mg/dL, parathyroid hormone: 4.1 pg/mL.

Bacterial and fungal cultures of the pustular lesions were negative. ECG showed findings were consistent with to hypocalcemia.

Biopsy specimen showed subcorneal pustules filled with neutrophils; that finding was consistent with the diagnosis of IH (Figure 2). The patient was referred to an endocrinologist and was reported to have hypoparathyroidism.

For the treatment, prednisolone 60 mg/day and calcium 2 g/day were started. The eruptions improved and the fever subsided within a few days. At the end of 15 days of therapy, the skin was completely cleared. Then, the patient was transported to the endocrinology department for the treatment of her hyproparathyroidism.

IH was first described in 1872 by Hebra and to date, about 350 cases have been reported in the literature. The name IH was chosen by Hebra, but this skin disease is neither an infectious disease, nor in any way associated with the herpes virus (1, 2). The etiology of this disorder has not been clearly identified yet. However, IH is generally thought to represent a variety of pustular psoriasis due to its close resemblance in terms of clinical appearance (5, 7, 8). Even though this very rare skin disorder is usually seen in pregnant females, particularly during the last trimester of pregnancy, there have been some case reports of IH in nonpregnant women and in men, also. Furthermore, surgical resection of the thyroid

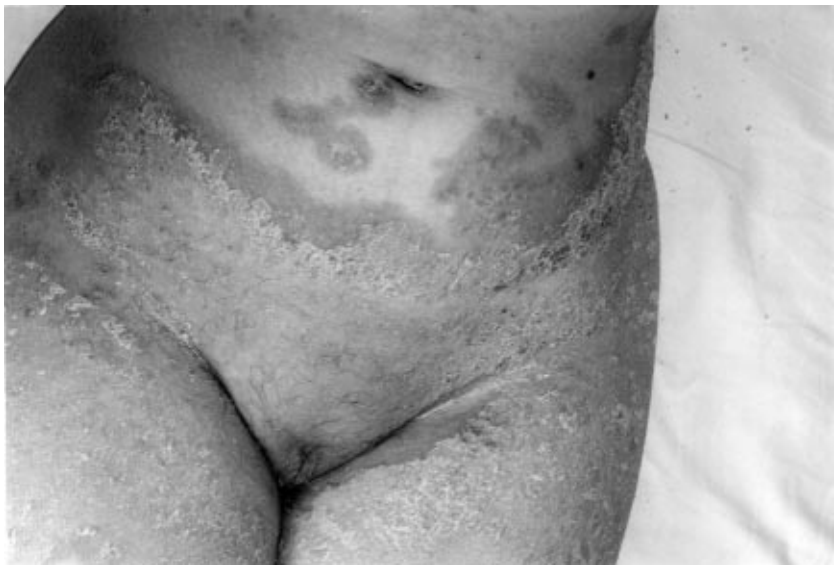

Figure 1. Clinical appearance of the case.

Figure 2. Biopsy specimen of skin lesion. Nate neutrophils invading epidermis to form a subcorneal microabcess

and parathyroid glands and hypocalcemic conditions have been thought to be cause of IH in nonpregnant women and men. Our patient was not pregnant and had hypoparathyroidism. Nevertheless, the role of hypocalcemia in IH has not been accurately determined yet (1, 2, 4, 9).

Patients with IH have been shown to respond dramatically to calcium therapy. On the other hand, some investigators have reported recurrences of IH in subsequent hypocalcemic episodes. Hypocalcemia is a cause or result of IH. This condition is not known. Although there are reports stating that hypocalcamia might precipitate IH, it has been claimed that hypocalcemia seen in IH might be a secondary phenomenon due to hypoalbunemia or malabsorption conditions seen in IH (2-4). According to Holm and Goldsmith, hypocalcemia and hypoalbunemia seen in IH are secondary to the extensive cutaneous inflammation that is the result of an extravasation of albumin and albumin-bound calcium to the intestinal space (6).

The skin eruptions in IH consist of hundreds of pinhead pustules on an erythematous base that may

disseminate all over the body through centrifugal spreading from the preferential localizations (abdomen, submammary and inguinal regions). Mucous membranes may be very rarely affected. The tongue, buccal mucosa and the oesophagus may be involved, with circinate or erosive lesions following short-lived pustules (1, 10). Our case had desquamation and pustules on the abdominal, back, gluteal and genital regions and upper parts of the thigh. Also, she had widespread ulcerations with purulent material on the mucosal surface of the mouth, hard palate and tongue. Histopathologically, there are intraepidermal pustules containing neutrophils (11). Histologic findings of our case were consistent with IH. The main recommended therapy for IH includes: fluid and electrolyte replenishment, and systemic administration of steroids (2). We gave systemic prednisolone and calcium replenishment to the patient who responded well to this therapy.

The case presented in this study was diagnosed as hypoparathyroidism. We think that the onset of symptoms of IH in our case might be related to hypocalcemia.

In conclusion since the patient was not pregnant and had hypoparathyroidism and widespread mucosal lesions, we think that this case is very interesting and we wanted to present it.

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