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Regression of the Sweet's syndrome after Bacillus Calmette-Guérin therapy: A bladder cancer case report

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ABSTRACT

The Sweet's syndrome is a rare dermatosis which can be related to underlying malignancies. In this case we presented a case, who applied to our clinic with severe dermatosis and hematuria. We diagnosed high grade non-muscle invasive bladder tumor, which was treated with transurethral resection of bladder-tumor and six cycle of Bacillus Calmette-Guérin therapy. After the treatment the dermatosis was regressed almost completely. However, recurrent bladder tumor was detected at the first control cystoscopy.

Introduction

The Sweet's syndrome was first identified by Dr. Robert Douglas Sweet. It can be also referred as febrile neutrophilic dermatosis.[1] Sweet's syndrome is a rare dermatosis characterized by rapid onset of tender erythematous plaques and nodules. It can present in three forms; classical (idiopathic), drug- and malignancy-related.

According to literature, most accepted theory is the cytokine induced hypersensitivity. As far as we know, interleukin-1 (IL-1) is the main cytokine that activates neutrophils, therefore it is highly possible that it can flare up the syndrome. Furthermore, in addition to IL-1; IL-2 and interferon gamma (IFN-γ); T_1 helper cells also responsible for the underlying pathology of the classical form of the syndrome. Moreover, Paydas[2] suggested that the underlying pathology of the malignancy-related form of the Sweet's syndrome may be the abundant production of the inflammatory cytokines, such as interleukins (IL-1-3-6-8), granulocyte colony stimulating factor (G-CSF) and granulocyte macrophage colony stimulating factor (GM-CSF).

In this paper, we aimed to present a patient, who was diagnosed with high grade non-muscle invasive bladder tumor with painful tender erythematous skin lesions and hematuria. The patient was treated with transurethral resection of bladder-tumor (TUR-BT) and six cycle of Bacillus Calmette-Guérin therapy (BCG).

Case

Sixty-five years old woman applied to the urology clinic with macroscopic hematuria. Physical examination revealed spontaneous eruption of symptomatic erythematous papulonodular lesions on the dorsal part of the upper extremities and the ventral parts of the both feet (Fig. 1).

Although the urinalysis proved the hematuria, the other lab tests were normal. We performed a urinary ultrasonography (USG) to evaluate the hematuria. USG showed a solid 18 × 27mm mass, which was located left wall of the bladder. We planned to perform TUR-BT to the patient. The pathology of the TUR proved high grade papillary urothelial carcinoma without an invasion to muscularis propria (T1HG). We performed Re-TUR to prove the pathology 4th week after the procedure. We planned six cycles of intravesical BCG therapy due to high grade nature of the tumor.

Afterward patient consulted to dermatology clinic regarding her skin lesions. Dermatologist decided to perform a skin biopsy. The biopsy results were consistent with neutrophilic dermatosis (Fig. 2). Therefore, we decided that these lesions and the malignancy altogether might the components of the Sweet's Syndrome. Dermatologist suggested to follow-up the skin lesions. After the BCG treatment were terminated, we performed an extensive physical examination and a cystoscopy. During the examination, we found that skin lesions regressed almost completely (Fig. 3), however cystoscopy revealed a recurrent tumor in the bladder.

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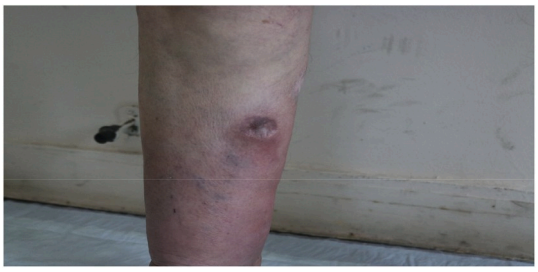

Fig. 1. Erythematous papulonodular skin lesions before BCG therapy.

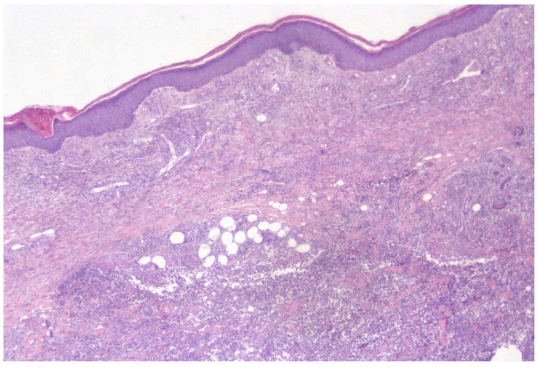

Fig. 2. Hyperkeratosis can be seen on the surface. Low achantosis on the epidermis and active supurative inflamation invasion to subcutaneous fatty tissue on the dermis, (Haematoxylin-eozin x40).

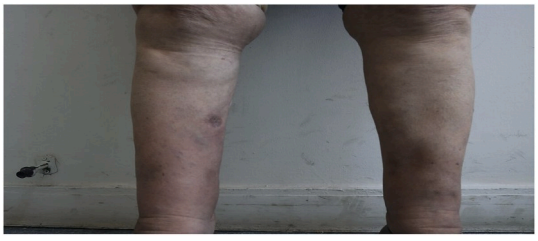

Fig. 3. The skin lesions regressed almost completely after BCG therapy.

Discussion

In the literature, Cohen et al.[3] published the first paper about malignancy-related Sweet's syndrome. Nearly 21% of patients with Sweet's syndrome have malignancy. Although the most common malignancies are hematological disorders (85%) such as acute myelogenous leukemia, Hodgkin disease and polycythemia vera, Sweet's syndrome also related with solid tumors such as adenocarcinomas of the genitourinary tract, gastrointestinal tract and breast.[4] In this form of the syndrome, the skin lesions can occur before, after or concomitant to the diagnosis of the malignancy. Therefore, especially in oncology patients, the Sweet's syndrome can be a strong sign of recurrence.

In our case, Sweet's syndrome occurred as a first sign of the tumor, alongside with hematuria. It was presented with high-grade bladder cancer as a paraneoplastic syndrome. In the literature it usually means a poor prognosis. After TUR-BT operation and six cycle of intravesical BCG therapy, the skin lesions regressed dramatically. However, we could not decide whether the surgery or the BCG therapy treated the skin lesions. In the literature, Cohen et al.[3] suggested that after the treatment of the malignancy, the related dermatosis should be regressed. However, we found a recurrent tumor in the bladder at the first control. Therefore, we decided that the BCG therapy might be the real reason for regressing the dermatosis. Although its exact mechanism of action is still unclear, complementary effect on the immune response have been shown in some studies.[5] Thus, the BCG therapy might somehow modulate the global immune response and clear the skin lesions.

Declaration of competing interest

All authors declare that he/she has no conflict of interest.

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent was obtained from all individual participants included in the study.

The work was performed at Urology Department of Sisli Hamidiye Etfal Training and Research Hospital Istanbul, Turkey.

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