CASE REPORT OLGU SUNUMU

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Surrenal Adenoma in a Patient with Lichen Myxedematosus

Liken Miksödematozuslu Bir Hastada Sürrenal Adenom

Himmet ÖZDEMİR[a], Ceyda TETİK AYDOĞDU[a]

[a]Muğla Sıtkı Koçman University Faculty of Medicine, Department of Dermatology, Muğla, Türkiye

ABSTRACT Lichen myxedematosus (LM) is an idiopathic cutaneous mucinosis due to dermal mucin accumulation. It has localized, diffuse and atypical forms. Since it may affect other organs in the body other than the skin, various screenings are required. Adrenal incidentaloma is defined as an adrenal mass detected on an imaging scan for an indication other than suspected adrenal disease. Although LM has been previously associated with hematologic malignancies, there is no evidence in the literature that it is associated with surrenal adenoma. In this case report, we present the 1st case of a surrenal adenoma in a patient with localized lichen myxedematosus.

Keywords: Mucinoses; lichen myxedematosus; adenoma; adrenal incidentaloma

ÖZET Liken miksödematozus (LM) dermal müsin birikimine bağlı idiyopatik bir kutanöz müsinozdur. Lokalize, yaygın ve atipik formları mevcuttur. Vücutta cilt dışında başka organları da etkileyebileceğinden dolayı çeşitli taramalar yapılması gerekmektedir. Adrenal insidentaloma, şüpheli adrenal hastalık dışında bir endikasyon için yapılan görüntüleme taramasında tespit edilen adrenal kitle olarak tanımlanır. LM daha öncesinde hematolojik malignitelerle ilişkilendirilmiş olmasına rağmen, literatürde sürrenal adenom ile ilişkili olduğuna dair bir kanıt yoktur. Bu olgu sunumunda, lokalize liken miksödematozuslu bir hastada görülen ilk sürrenal adenom olgusunu paylaşıyoruz.

Anahtar Kelimeler: Müsinozis; liken miksödematozus; adenom; adrenal insidentalom

Lichen myxedematosus (LM) is an idiopathic cutaneous mucinosis characterized by lichenoid papules due to dermal accumulation of mucin without thyroid dysfunction.[1] LM may be associated with monoclonal gammopathy and related hematological malignancies. Cases of LM associated with lung adenocarcinoma, thymic carcinoma, seminoma, hepatocellular carcinoma, gastric adenocarcinoma, pancreatic carcinoma, esophageal carcinoma have been reported in the literature.[2,3] This case report, we share a 1st case of surrenal adenoma in a patient with localized lichen myxedematosus.

CASE REPORT

A 62-year-old man patient presented to our clinic with itchy, skin-colored papules on his upper extremities, shoulders and back (Figure 1). These papular lesions had appeared 5 months ago.

His medical history included irritable bowel disease for 32 years, hypertension for 26 years, hepatosteatosis grade 1, and insulin resistance. Laboratory studies and thyroid function tests were normal at the time of diagnosis.

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Correspondence: Himmet ÖZDEMİR
Muğla Sıtkı Koçman University Faculty of Medicine, Department of Dermatology, Muğla, Türkiye
E-mail: himmetozdemir@hotmail.com

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Histopathologic examination revealed orthokeratosis in the epidermis, basal vacuolar degeneration, mild fibroblast increase in the reticular dermis and focal mucin deposition by Alcian blue (Figure 2 A, B).

Hematology consultation was requested to investigate paraproteinemia. Protein electrophoresis was normal. Bone marrow biopsy was not required for the diagnosis. Given the presence of clinical lesions and histopathological evidence and the lack of monoclonal gammopathy and thyroid disease, a diagnosis of localized LM was made.

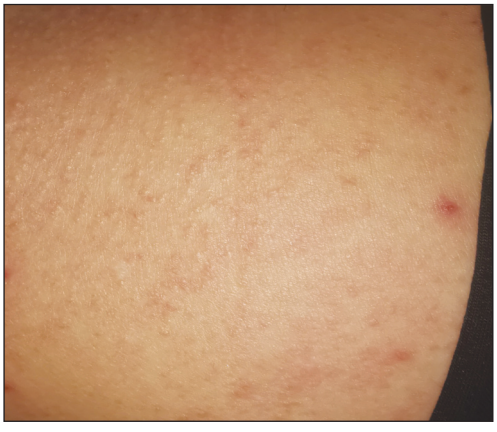

FIGURE 1: Itchy, skin-colored papules on his upper extremities, shoulders and back. These papular lesions had appeared 5 months ago.

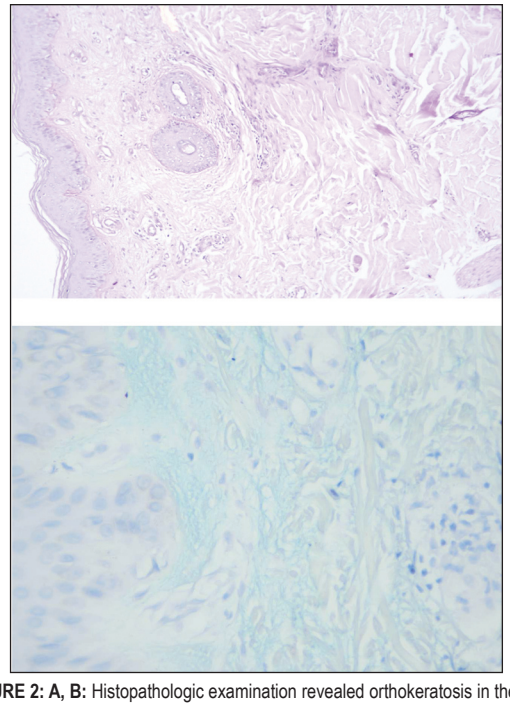

FIGURE 2: A, B: Histopathologic examination revealed orthokeratosis in the epidermis, basal vacuolar degeneration, mild fibroblast increase in the reticular dermis and focal mucin deposition by Alcian blue.

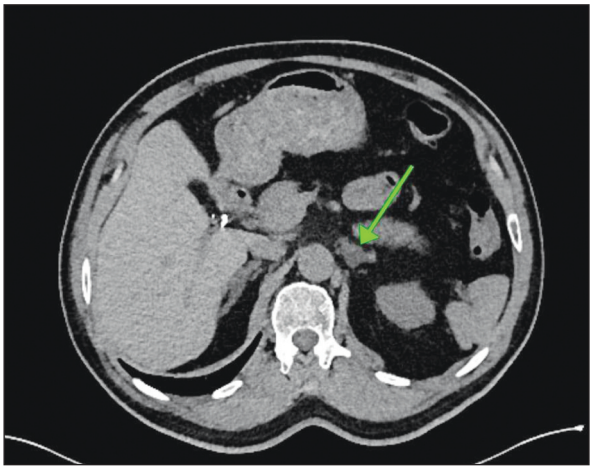

FIGURE 3: In the imaging studies performed after the diagnosis, a nodular lesion with a size of approximately 14 mm and 14 Hounsfield units (HU) was detected in the left surrenal gland and evaluated as adenoma.

In the imaging studies performed after the diagnosis, a nodular lesion with a size of approximately 14 mm and 14 Hounsfield units (HU) was detected in the left surrenal gland and evaluated as adenoma (Figure 3). The patient was diagnosed with mild autonomic cortisol secretion (MACS) as a result of laboratory tests performed by the endocrinology department. No cortisol-induced cushingoid skin findings were detected. The patient's adenoma is being followed up and no growth has been detected so far and is considered benign.

The patient was treated with 0.1% betamethasone ointment. After 3 months of follow-up, significant regression was observed in the lesions and itching complaint. The patient has been under follow-up for 2 years and no new lesions have appeared.

Written informed consent was obtained from the individual(s) for publication of potentially identifiable images or data contained in this article.

DISCUSSION

LM is an idiopathic cutaneous mucinosis characterized papular or nodular eruption, monoclonal gammopathy, mucin accumulation with fibroblast proliferation and lack of thyroid disease. Rongioletti and Rebora divided LM into 3 distinct clinicopatho-

logical subgroups. These are scleromyxedema, localized LM and other atypical or intermediate forms that do not fit into these 2 subgroups.[4]

Adrenal incidentalomas are defined as adrenal enlargements detected on imaging performed for an indication other than suspicion of adrenal disease. Adrenal incidentaloma patients may have MACS. This condition should be suspected if the cortisol level is ≥50 nmol/L after suppression with 1 mg dexamethasone.[5]

Additional imaging is recommended for adrenal tumors that are non-functional, smaller than 4 cm and 11-20 HU. If additional imaging confirms that the adrenal tumor is benign, further follow-up is not recommended. An alternative approach is a non-contrast computed tomography scan after 12 months.[6]

It has been reported that findings such as acanthosis nigricans, moon face, buffalo hump, hirsutism, facial flushing may be seen as skin findings in the course of malignant adrenal tumors.[7-9] Skin findings associated with benign adrenal tumors are limited in the literature.

Treatment of LM is challenging because the literature on treatment is limited to case reports and series. Thalidomide, high dose dexamethasone, methotrexate, retinoids, psoralen plus ultraviolet A, plasmapheresis, IVIG are possible treatment options.[10]

However, successful results have been obtained with topical steroids especially in the treatment of symptomatic localized lichen myxedematosus. Steroid treatment is thought to target paraprotein production with both immunosuppressive and anti-fibroblast effects.[10] We applied 0.1% betamethasone ointment and obtained successful results.

The association of LM and surrenal adenoma has not been previously reported in the literature. Surrenal adenoma may not have been detected in these patients due to screening for hematologic malignancies only. Additionally, adrenal adenomas can be detected incidentally, which may have made it more difficult to establish a connection with dermatological diseases.

In the literature, a case of autoimmune hypothyroidism and adrenal insufficiency with LM has been reported.[11] Additionally, autoimmune thyroid diseases have been more frequently reported in patients with incidentally detected adrenal adenomas.[12] The occurrence of autoimmune diseases together with both diseases suggests that autoimmune processes may play a role in the development mechanisms of these diseases. Therefore, we suggest that autoimmune diseases should be screened in cases with unclear pathogenesis in the future.

It has been previously described in a case of cutaneous mucinosis and Cushing syndrome together. In this case, improvement in the lesions on the feet and hands was observed 1 month after adrenalectomy.[13] Chronic inflammation may be observed due to changes in the immune system in Cushing syndrome.[14,15] In our case, the surrenal adenoma secreted mild levels of cortisone. This suggests that the extra amount of cortisone secreted in the body may cause mucin accumulation through inflammatory pathways.

This case report, we shared a case of a surrenal adenoma found in a patient with localized lichen myxedematosus. In the literature to the best of our knowledge, the togetherness of surrenal adenoma and LM in the same patient has never been documented before. In general, literature data on LM is limited and further studies are needed.

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

All authors contributed equally while this study preparing.

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