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Case presentation

Lichen planus-like drug reaction associated with recombinant human growth hormone therapy in a child patient with Turner syndrome

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Abstract

Turner syndrome (TS) is a genetic disease with an incidence rate of between 1:2000 and 1:5000 live female births. The treatment of TS differs according to age and Recombinant Human Growth Hormone (RHGH) therapy is usually given for the treatment of short stature in girls with TS in childhood. We describe the first case of a TS patient who presented with a clinical picture compatible with oral and palmoplantar lichen planus-like reaction during RHGH therapy; spontaneous remission occurred after therapy suspension.

Keywords: Lichen planus, Recombinant growth hormone; Turner syndrome; RHGH

Introduction

Turner syndrome (TS) is a genetic disease with an incidence rate of between 1:2000 and 1:5000 live female births [1]. The phenotypic abnormalities are related to monosomy of the X-chromosome or to many other X-chromosome abnormalities such as rings, deletions, isochromosomes, and mosaicisms [2].

The degree to which girls are affected is determined by the specific chromosomal abnormality: they may have only a few of the features associated with the syndrome, but short stature and infertility are nearly always present [3]. Oral findings, such as malocclusions, premature eruption, crowding of teeth, and alterations in tooth shape, size, and enamel (hypoplasia) have been observed [4].

The treatment of TS differs according to age and Recombinant Human Growth Hormone (RHGH) therapy is usually given for the treatment of short stature in girls with TS in childhood [5]. We describe the first case of a TS patient who presented with a clinical picture compatible with oral and palmoplantar lichenoid reaction during RHGH therapy. Spontaneous remission was observed after the suspension of therapy.

Case synopsis

A nine-year-old girl, previously diagnosed as having monosomy of the X-chromosome, reported “feeding difficulties” owing to burning in the mouth. The burning sensation had appeared one month previously making it difficult for her to eat and she had lost weight. The patient was under medical care with a cardiologist and endocrinologist and had been receiving HRGH for two months (Figure 1).



Figure 1. Clinical features of the patient with Turner syndrome showing low-set pinna and webbed neck.

During the physical examination numerous scaly papules were observed on the palms and soles. The lesions of the hands were seen on the fingers (Figure 2a); lesions on the soles were seen on the internal plantar arch (Figure 2b). The patient reported itching in both areas. Numerous interlacing white keratotic lines were noted involving labial and buccal mucosa (Figure 2c and 2d). Biopsy was carried out and histological findings were compatible with lichen planus-like drug reaction (Figure 3a and 3b).



Figure 2. Pruriginous scaly papules on the fingers and on soles on the internal plantar arch (a and b). Multiple interlacing white keratotic lines were noted involving labial and buccal mucosa (c and d)

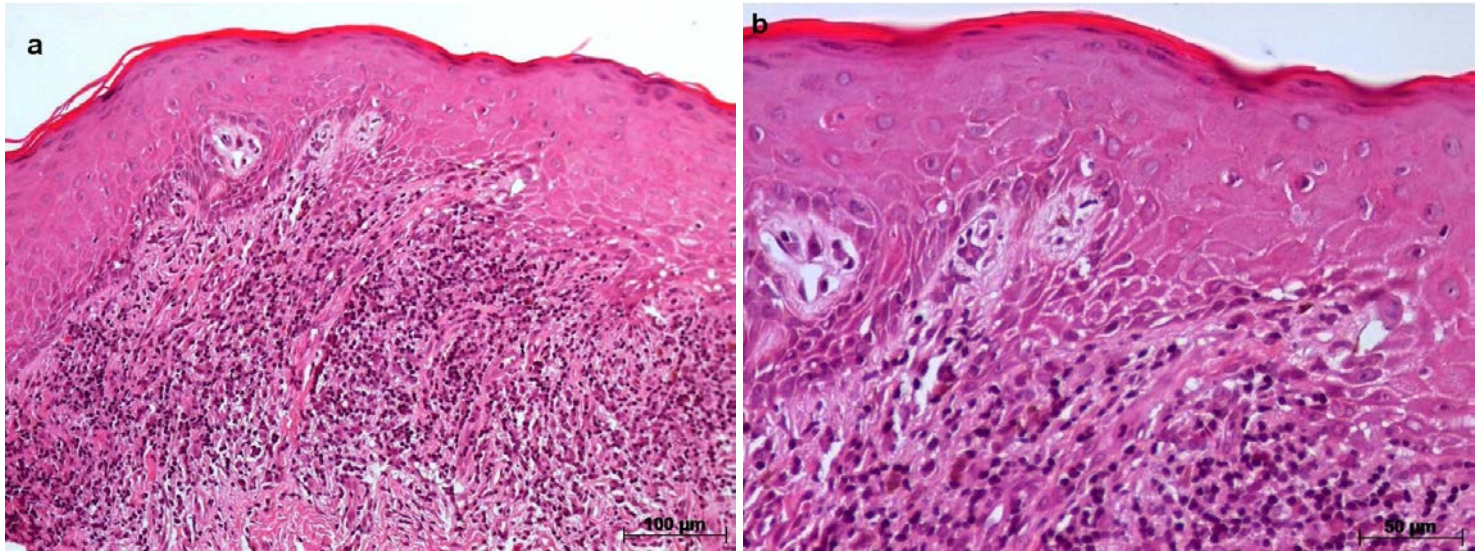


Figure 3. Oral mucosa with band-like inflammatory cell infiltrate, mainly of lymphocytes at the connective tissue junction (a and b)

Treatment was initiated with topical corticosteroid. Clobetasol propionate 0,05% cream was applied on palmoplantar lesions and dexamethasone elixir 0,5 mg mouthwash was prescribed for oral lesions three times a day. Significant improvement in the symptoms was observed after one month of treatment and the palmoplantar lesions completely disappeared whereas the oral lesions remained present. The patient was monitored and the oral lesions remained stable until the patient discontinued the use of HRGH and the oral lesions completely disappeared (Figure 4, a and b). The patient continues to be followed up.

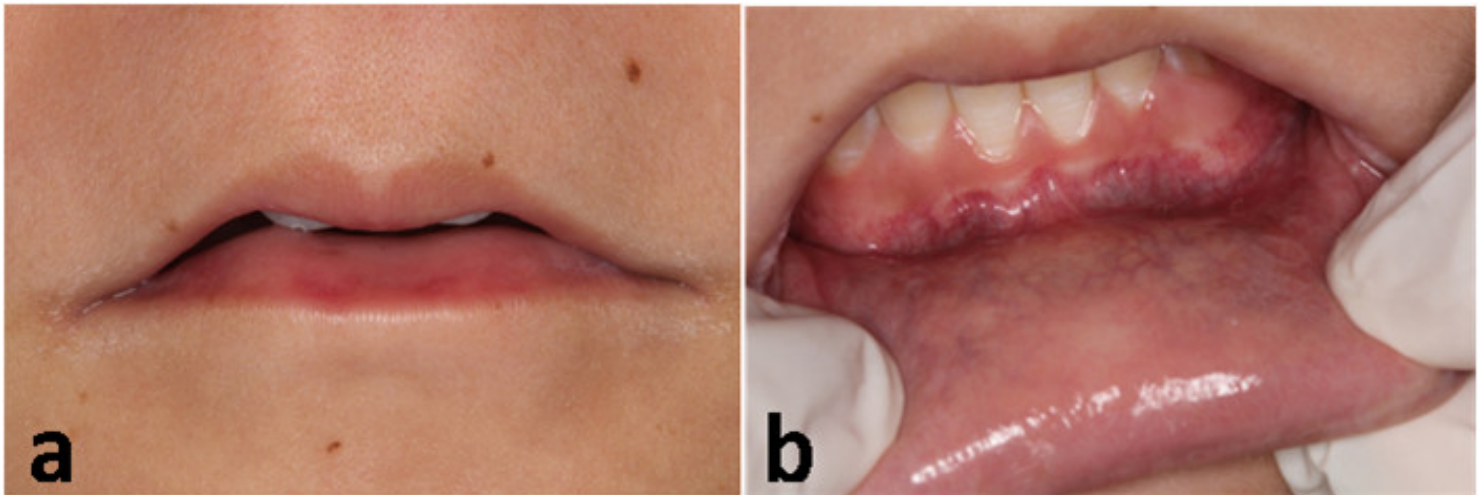


Figure 4. Remission of the lesions in oral mucosa after suspension of hormone therapy (a and b).

Discussion

A high prevalence of autoimmunity and associated disease has been reported in TS patients, especially hypothyroidism, celiac disease, and type I diabetes. Although less common, psoriasis and vitiligo are also reported in the literature [1, 6]. The occurrence of LP in Turner syndrome patients is rare and there are just a few cases reported in the literature in adult patients [6, 7]. However, this is the first reported case of LP-like reaction in a child.

The most interesting feature about this case is that the patient had just started to receive HRGH when she first presented with the clinical signs of the disease, all of which disappeared when the therapy was discontinued. To our knowledge, just one similar case has been reported in the literature; a nine-year-old boy with dwarfism who presented with skin-colored discrete pinpoint papules in the skin and a violaceous, polygonal plaque on the glans penis during the course of HRGH treatment. His condition was diagnosed as lichen planus and it remained resistant to treatment during HRGH therapy [8].

Lichen planus-like drug reaction or lichenoid drug reactions (LDR) occur when oral and/or cutaneous lesions arise in a close temporal association with the taking of certain medications and can occur anytime during the drug administration. Oral LDR is uncommon [9]. However, its prevalence is increasing, probably because of the introduction of new categories of medications [10].

There are no clear or distinct clinical and histologic features that reliably distinguish LDR and lichen planus. The most accurate way of diagnosing LDR is based on the resolution of the reaction after withdrawal of the suspected inciting drug. If

reintroduction of the drug produces recurrence, the association is confirmed. Nevertheless, this may not be acceptable in clinical practice. Furthermore, a recurrence may take months to regress after drug withdrawal [9-11]. Hence, the differential diagnosis of LDR remains problematic and there are no evidence-based studies regarding this condition [9].

Lichenoid drug reactions, such as lichen planus, are T-cell-mediated inflammatory diseases and evidence supports a role of immune dysregulation in the LDR process mainly involving CD8+ cells [12]. As previous investigations demonstrate that growth hormone exerts physiological effects on the immune system (increasing thymocyte migratory responses and intrathymic traffic of developing T cells and peripheral T cell migration) [13, 14] it is possible that HRGH can contribute to autoimmune diseases progression mediated by T cell such as LDR.

In conclusion, this is the first case report of a lichen planus-like reaction in a TS child receiving HRGH therapy. Although there is no causal relationship established, it is known that TS patients have a high prevalence of autoimmunity and associated disease and in this case the growth hormone therapy seems to have had a decisive role in the course of the disease. We emphasize the need for multidisciplinary attention to TS patients during HRGH therapy.

References

1. Lowenstein EJ, Kim KH, Glick SA. Turner's syndrome in dermatology. *Journal of the American Academy of Dermatology* 2004 50:767-76 [PMID: 15097963]
2. Larizza D, Calcaterra V, Martinetti M. Autoimmune stigmata in Turner syndrome: when lacks an X chromosome. *Journal of autoimmunity* 2009 33:25-30 [PMID: 19349146]
3. Reh CS, Geffner ME. Somatotropin in the treatment of growth hormone deficiency and Turner syndrome in pediatric patients: a review. *Clinical pharmacology : advances and applications* 2010 2:111-22 [PMID: 22291494]
4. Lopez ME, Bazan C, Lorca IA, Chervonagura A. Oral and clinical characteristics of a group of patients with Turner syndrome. *Oral surgery, oral medicine, oral pathology, oral radiology, and endodontics* 2002 94:196-204 [PMID: 12221388]
5. Spiliotis BE. Recombinant human growth hormone in the treatment of Turner syndrome. *Therapeutics and clinical risk management* 2008 4:1177-83 [PMID:19337425]
6. Kurgansky D, Burnett JW. Widespread lichen planus in association with Turner's syndrome and multiple endocrinopathies. *Cutis* 1994 54:108-10 [PMID:7956333]
7. Cornebleet TW, JR; Musgrave, DP. Turner's syndrome associated whit lichen planus. *AMA Arch Derm Syphilol* 1950 62:564-8 [PMID: 14770493]
8. Oono T, Arata J. Childhood lichen planus in a patient receiving growth hormone for dwarfism. *Dermatology* 1996 192:87-8 [PMID: 8832965]
9. Al-Hashimi I, Schifter M, Lockhart PB, Wray D, Brennan M, Migliorati CA, et al. Oral lichen planus and oral lichenoid lesions: diagnostic and therapeutic considerations. *Oral surgery, oral medicine, oral pathology, oral radiology, and endodontics* 2007 103 Suppl:S25 e1-12 [PMID: 17261375]
10. De Rossi SS, Ciarrocca K. Oral lichen planus and lichenoid mucositis. *Dental clinics of North America* 2014 58:299-313 [PMID: 24655524]
11. Nigen S, Knowles SR, Shear NH. Drug eruptions: approaching the diagnosis of drug-induced skin diseases. *Journal of drugs in dermatology : JDD* 2003 2:278-99[PMID:12848112]
12. Roopashree MR, Gondhalekar RV, Shashikanth MC, George J, Thippeswamy SH, Shukla A. Pathogenesis of oral lichen planus-a review. *Journal of Oral pathology & Medicine* 2010 39:729-34 [PMID:20923445]
13. Smaniotto S, Martins-Neto AA, Dardenne M, Savino W. Growth hormone is a modulator of lymphocyte migration. *Neuroimmunomodulation* 2011 18:309-13 [PMID:21952682]
14. Dardenne M, Smaniotto S, de Mello-Coelho V, Villa-Verde DM, Savino W. Growth hormone modulates migration of developing T cells. *Annals of the New York Academy of Sciences* 2009 1153:1-5 [PMID: 19236321]