Scholars Journal of Medical Case Reports

Sch J Med Case Rep 2015; 3(2):102-103 ©Scholars Academic and Scientific Publishers (SAS Publishers) (An International Publisher for Academic and Scientific Resources) www.saspublishers.com ISSN 2347-6559 (Online) ISSN 2347-9507 (Print)

DOI: 10.36347/sjmcr.2015.v03i02.010

Palmoplantar Pustulosis Following Olfactory Cleft Inflammation

Nobuko Makino¹, Shinji Makino²

¹Department of Public Health, Jichi Medical University, Shimotsuke, Tochigi, Japan ²Department of Ophthalmology, Jichi Medical University, Shimotsuke, Tochigi, Japan

*Corresponding Author: Name: Shinji Makino Email: makichan@jichi.ac.jp

Abstract: We present a case of palmoplantar pustulosis (PPP) secondary to olfactory cleft inflammation in a 65-year-old man. He had a 10-year history of PPP. Computed tomography showed an abnormal intensity that corresponded to chronic inflammatory sinusitis in the right maxillary and ethomoidal sinuses, and obstruction of the olfactory cleft. After administration of intranasal steroid drops, the olfactory cleft inflammation improved and healthy skin was restored. Discontinuing this treatment resulted in worsening of the skin lesions 3 times over 3 years. Although the chronic inflammatory sinusitis persisted, the olfactory cleft inflammation improved. Therefore, we conclude that PPP might be caused by olfactory cleft inflammation.

Keywords: Palmoplantar pustulosis, Olfactory cleft inflammation, Focal infection

INTRODUCTION

Palmoplantar pustulosis (PPP) is a chronic, relapsing, pustular dermatosis eruption that is localized to the palms and soles [1-3]. PPP is characterized by repeated remissions and exacerbations, and there is no treatment. PPP is widely considered to manifest secondary to a primary focal infection such as tonsillitis, sinusitis, and dental infection [4-6]. To our knowledge, PPP associated with olfactory cleft inflammation is extremely rare [7]. Here, we present a case of PPP following olfactory cleft inflammation.

CASE REPORT

A 65-year-old man was referred to Jichi Medical University Hospital for an olfactory complaint. He had prior surgery for left chronic inflammatory sinusitis at 17 years of age. He also suffered from a 10year history of recurrent and painful eruptions of pustules on his palms and soles that resolved with redbrown discoloration. Based on our dermatological findings, he was diagnosed with PPP. His palms (Fig. 1A, 1B) and soles (Fig. 1C, 1D) showed thin erythematous plaques with desquamation, mixed with numerous brown macules and scattered pustules, consistent with a diagnosis of PPP.

Otolaryngological endoscopic examination was unremarkable in both nasal cavities (Fig. 2A and 2B). Tonsillitis was not detected.

Coronal computed tomography (CT) showed total obstruction of the olfactory cleft by mucosal

thickening and marked mucosal thickening in the right maxillary sinus (Fig. 3).

Administration of intranasal steroid drops resulted in improved olfactory function and his skin was restored to normal (Fig. 4A–C). However, the treatment was discontinued 3 times over 3 years and each time the skin lesions worsened.



Fig. 1: Palms (A, B) and soles (C, D) showing thin erythematous plaques with desquamation, mixed with numerous brown macules and scattered pustules.



Fig. 2: Endoscopic photographs in the right (A) and the left (B) nasal cavities.



Fig. 3: Coronal CT demonstrating obstruction of the olfactory cleft





Fig. 4: Palms (A) and soles (B, C) showing normal skin appearance after administration of intranasal steroid drops.

DISCUSSION

The olfactory cleft is a paired orifice located in the medial and upper regions of the nasal cavity [8]. As the first stage of the olfactory process involves the passage of odorant molecules to the neuroepithelium near the upper part of the olfactory cleft, the patency of the cleft is critical for normal olfactory function [8]. Adequate space between the septum and the middle turbinate that allows normal airflow through the olfactory cleft is also important for normal olfaction [8]. In this present case, we considered an olfactory disturbance caused by chronic inflammatory sinusitis and olfactory cleft inflammation.

To our knowledge, PPP associated with olfactory cleft inflammation is extremely rare [7]. We previously reported a case of PPP following olfactory cleft inflammation [7]. In that report, the patient had olfactory cleft inflammation without chronic inflammatory sinusitis. That patient's skin, as in the present case, was restored to normal after administration of intranasal steroid drops. Furthermore, as in this case, discontinuing the intranasal steroid drop treatment resulted in worsened skin lesions (5 times over 2 years). In the current case, because both the PPP and the olfactory cleft inflammation improved, we, therefore, propose that PPP might be caused by olfactory cleft inflammation. This is the second known case report describing this rare and unusual association.

CONCLUSION

Our findings were based on a single case, additional cases and further immunological evaluations are necessary to definitively characterize the association between PPP and olfactory cleft inflammation.

REFERENCES

- 1. Mrowietz U, van de Kerkhof PC; Management of palmoplantar pustulosis: do we need to change? Br J Dermatol., 2011; 164 (5):942-946.
- Yamamoto T; Extra-palmoplantar lesions associated with palmoplantar pustulosis. J Eur Acad Dermatol Venereol., 2009; 23(11): 1227-1232.
- Wetter DA; Palmoplantar pustulosis. CMAJ, 2013; 185 (11): 982.
- 4. Yokoyama M, Hashigucci K, Yamasaki Y; Effect of tonsillectomy in patients with pustulosis palmaris et plantaris. Acta Otolaryngol., 2004; 124(9): 1109-1110.
- Takahara M; Clinical outcome of tonsillectomy for palmoplantar pustulosis and etiological relationship between palmoplantar pustulosis and tonsils. Adv Otorhinolaryngol., 2011; 72: 86-88.
- Harabuchi Y; Clinical manifestations and pathogenesis of tonsillar focal diseases: IgA nephropathy and palmoplantar pustulosis. Adv Otorhinolaryngol., 2011; 72: 1-5.
- Makino N, Ohta Y, Ishikawa T, Ichimura K; Intranasal steroid drops effective in palmoplantar pustulosis probably caused by olfactory cleft inflammation. Jpn J Rhinol., 2005; 44 (2): 123-126.
- Biscabe B, Faulcon P, Amanou L, Bonfils P; Olfactory cleft disease: An analysis of 13 cases. Otolaryngol Head Neck Surg., 2004; 130 (2): 202-208.