

Oral findings in a patient with Sebaceous Nevi - A Case Report

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ABSTRACT

The presentation herewith as scripted is to describe a case with Nevus sebaceous with oral manifestations. Nevus Sebaceous or Jadassohn's nevus is an epidermal nevus with predominant sebaceous glands seen histologically. Reports of oral involvement have been few ranging from papillomatous growths of the tongue, gingiva, palate to dental abnormalities such as anodontia and dysodontia. The present case describes a nevus sebaceous present on the right half of the face and neck, showing intraoral papillomatous growth on the lateral part of the tongue on the right side. The patient was healthy and did not report involvement of any other organ systems. Intraoral involvement may be seen in patients with Nevus Sebaceous, hence proper screening is important. In patients presenting with large nevi on the head and neck such as ours, involvement of other systems such as ocular, neurologic and oral lesions may be seen, therefore screening of such patients is of importance. Patients with nevus sebaceous may be predisposed to the occurrence of tumours. Therefore, careful screening of such patients is necessary.

Key Words: Case report, intraoral, sebaceous nevus, sebaceous glands.

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Introduction

An epidermal nevus is a skin hamartoma consisting of normal skin components in an abnormal distribution.¹ Epidermal nevi have been subdivided based on their predominant histologic component.² Various classifications include verrucous epidermal nevus, nevus sebaceous of Jadassohn, nevus comedonicus, apocrine nevus and eccrine nevus with the predominant histologic components being surface epidermis, sebaceous glands, hair follicles, apocrine glands, and eccrine glands, respectively.³ Jadassohn first described sebaceous nevi in 1895.⁴ Sebaceous nevi usually occur sporadically, a feature

that led to a belief that the sebaceous nevus is not a hereditary trait. Although uncommon, reports of familial aggregation of sebaceous nevi are accumulating and run counter to this view.⁴ It has been proposed that the sebaceous nevus may be inherited as a paradominant trait.⁵ Sebaceous nevus is a benign lesion that occurs most frequently on the scalp (59.3%) but has also been found on the face (32.6%), preauricular area (3.8%), neck (3.2%), and locations off the head and neck (1.3%),⁶ characteristically in the midline.⁷ The usual presentation being as a verrucous, granulated, yellow-orange plaque that may be round, crescentic, or linear in shape.⁶

Sebaceous nevus is theorized to result from genomic mosaicism in stem cells that expand in the distribution of the lines of Blaschko.⁸ A syndrome involving linear sebaceous nevi known as Sebaceous nevus syndrome, with a broad spectrum of multisystem disorders has also been described including epilepsy, mental retardation, neurologic and skeletal defects.⁶ This report describes a case of sebaceous nevus involving the left side of the forehead, face, neck and the scalp with intraoral patchy-pigmentation of the tongue.

Case Report

A 18-year-old female patient reported to the department of Periodontics for routine prophylaxis. Her medical and family history were insignificant. Extraorally pigmented wart-like lesions were seen on the left side of the forehead extending to the scalp.



Fig. 1: Extraoral photograph showing the nevi on the face.

Hair was sparse in the area circumscribing the lesion. Pigmented wart-like lesions were also seen involving the left side of the face, extending to the preauricular area and the neck (Figure 1). Aplasia cutis was seen

where the lesions extended to the scalp (Figure 2). The pigmented lesions were present since birth and no changes were seen since then. Intraorally, slightly pigmented papillomatous lesion was seen on the dorsum of the tongue on the right side (Figure 3).The



Fig. 2: Extraoral photograph showing aplasia cutis.

lesion was asymptomatic, and the patient did not want excision of the lesion. Fordyce's granules were seen on the oral mucosa on the left side and the lower lip. Due to esthetic problems the patient wanted excision of the extraoral pigmented lesions. Biopsy of the lesion on the scalp showed the presence of hair follicles and sebaceous glands (Figure 4).Based on the clinical and histopathologic findings a diagnosis of nevus sebaceous was made. Since the patient did not want excision of the intraoral lesion no treatment other than routine prophylaxis was performed. The patient was referred to a dermatologist for excision of cutaneous nevi and is currently undergoing carbon dioxide laser resurfacing and is under follow up.

Discussion



Fig. 3: Intraoral papillomatous lesion on lateral part of the tongue.

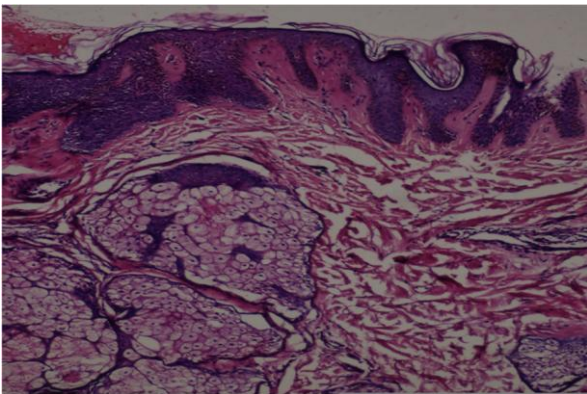


Fig. 4: Histologic section showing sebaceous glands and hair follicles (H&E stain, 40X)

Nevus sebaceous is usually seen at birth, and in childhood may present as a circumscribed, slightly raised, hairless plaque that is often linear in configuration because the sebaceous glands are underdeveloped and, therefore greatly reduced in size and number. At puberty the lesion becomes more verrucous and nodular and assumes its diagnostic histologic appearance.⁷ Multiple, linear plaques that strictly were confined to the midline were seen in the present case giving an impression of sebaceous nevus. However, for a diagnosis of nevus sebaceous the clinical findings have to be substantiated by histopathologic findings. In the differential diagnosis lesions such as sebaceous hyperplasia or sebaceous adenoma can be considered. The combination of apocrine glands, mature sebaceous glands and hair follicles, is characteristic of nevus sebaceous and cannot be found in sebaceous adenoma or sebaceous hyperplasia.

Intraoral anomalies have been described in association with nevus sebaceous. Mostly benign lesions, such as papillomas of the gingival mucosa, buccal mucosa or tongue have been described.^{9,10} One case of gingival papilloma associated with sebaceous nevus syndrome was presented by Reichart et al.⁹ In a review of epidermal nevi, Brown and Gorlin¹¹ collected 24 reports describing intraoral involvement, usually in the form of papillomatous lesions of the lips, the tongue or the palate. Hypodontia, unerupted teeth, abnormal spacing of teeth and teeth of abnormal size were also described. Intraoral involvement has also been described in association with nevus sebaceous syndrome (NSS), where multi-organ systems are involved.¹² In addition, two reports have also described oral mucosal lesions contiguous to a sebaceous nevus of the face.^{12,13} In the present case, papillomatous lesion was found on the tongue. The patient did not show involvement of any other organ systems apart from the nevi, ruling out a diagnosis of NSS.

Various neoplasms have been described to arise within the sebaceous nevi themselves.^{14,15} An examination of 596 sebaceous nevi showed that more than 90% of these tumours are benign, and about five cases of basal cell carcinomas were reported.¹⁴ Occurrence of tumours was found to be rare in children less than 16 years of age and all of them were benign. Other malignant tumours that may arise from sebaceous nevi include squamous cell carcinoma and malignancies of apocrine, eccrine and sebaceous glands.¹⁶

Conclusion:

Intraoral involvement may be seen in patients with Nevus Sebaceous, hence proper screening is important. In patients presenting with large nevi on the head and neck such as ours, involvement of other systems such as ocular, neurologic and oral lesions may be seen, therefore screening of such patients is of importance.

Clinical Significance

Patients with nevus sebaceous may be predisposed to the occurrence of tumors. Therefore, careful screening of such patients is necessary.

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