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A hamartoma-like mass on the palate? A possible discussion regarding the components of a pigmented naevus and hyperplastic salivary gland

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Abstract
An extremely rare mass lesion arising on the hard palate is presented. The patient was a 49-year-old Japanese male. He had a painless, semi-globular, smooth-surfaced and partially pigmented mass located on the left hard palate with no evidence of growth for approximately 20 years. The resected specimen included hyperplastic salivary gland tissue, nerve fibres and vessels surrounded by adipose tissue. All constituent tissues showed excessive growth for this location. Also spiralling nests of naevus cells representing inactive intramucosal naevus were included.We consider the whole lesion to be a hamartoma.

Key words: Hamartoma; Palate; Salivary gland; Hyperplasia

Introduction

Pigmented naevus is relatively rare in the oral region and is recognized most frequently on the hard palate (Buchner *et al.*, 1990). Hyperplasia of salivary glands is a rare lesion and was initally described by Giansanti *et al.* (1971) as a clinically asymptomatic and tumour-like mass of the palate and histologically hypertrophic and hyperplastic mucous acini. Buchner *et al.* (1991) studied a series of 40 cases and stated that more than 80 per cent were located on the hard and soft palate.

Here we present an extremely rare mass lesion on the hard palate including components of a pigmented naevus and hyperplastic salivary gland, which has never been described previously. Its pathogenesis is discussed and the literature is reviewed.

Case report

A 49-year-old Japanese male was referred to the Department of Oral and Maxillofacial Surgery, Kurume University School of Medicine in November, 1994. The patient had a painless mass located on the left hard palate, extending to palatal gingiva corresponding to left upper molars and showing no evidence of growth for about 20

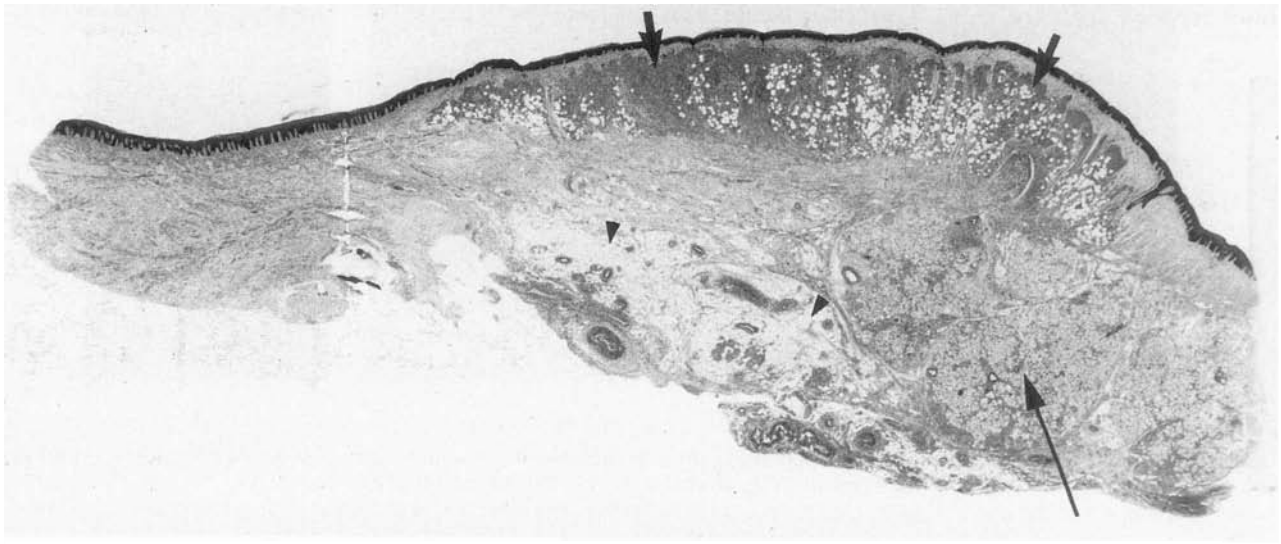

Fig. 1a
Mass showing naevus area (short arrow), hyperplastic salivary gland (long arrow) and adipose tissue (arrow head). (H & E; × 5)

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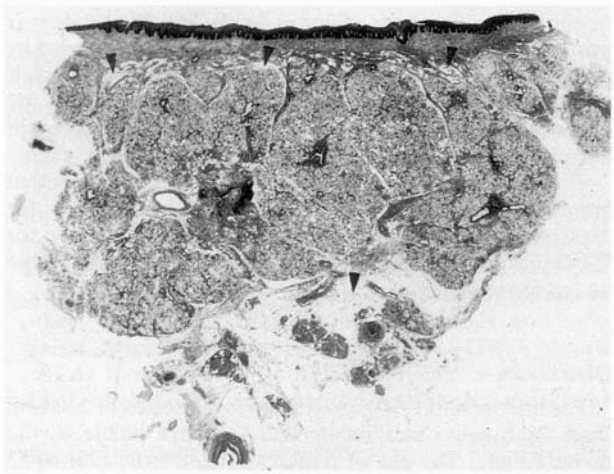

FIG. 1b

Portion of salivary gland tissue and surrounding adipose tissue (arrow head). (H & E; × 3)

years. The mass, semi-globular, smooth-surfaced, partially pigmented, elastic-hard and without ulcer formation, measured 30 × 25 × 15 mm and showed neither radio-opaque material nor bone resorption by radiography. Hydrodipsia was not noted.

The patient drank only a half bottle of beer a day and had been advised of a fatty liver several years before, but there was no history of a biopsy. Laboratory data revealed

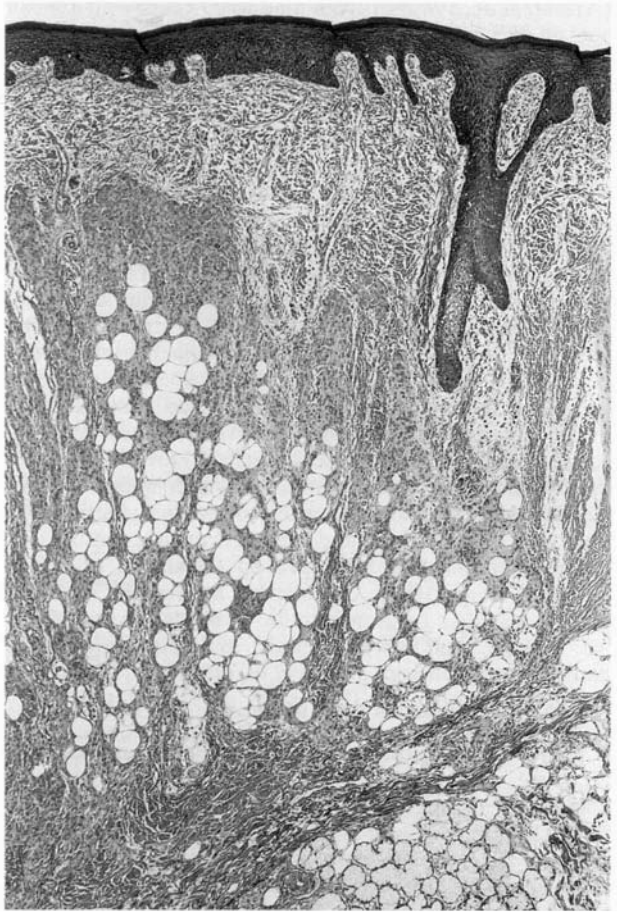

FIG. 2a

Spiralling nests of naevus cells including large fat cells (above) and salivary gland tissue (below) beneath the overlying epithelium showing rete ridge elongation. (H & E; × 56).

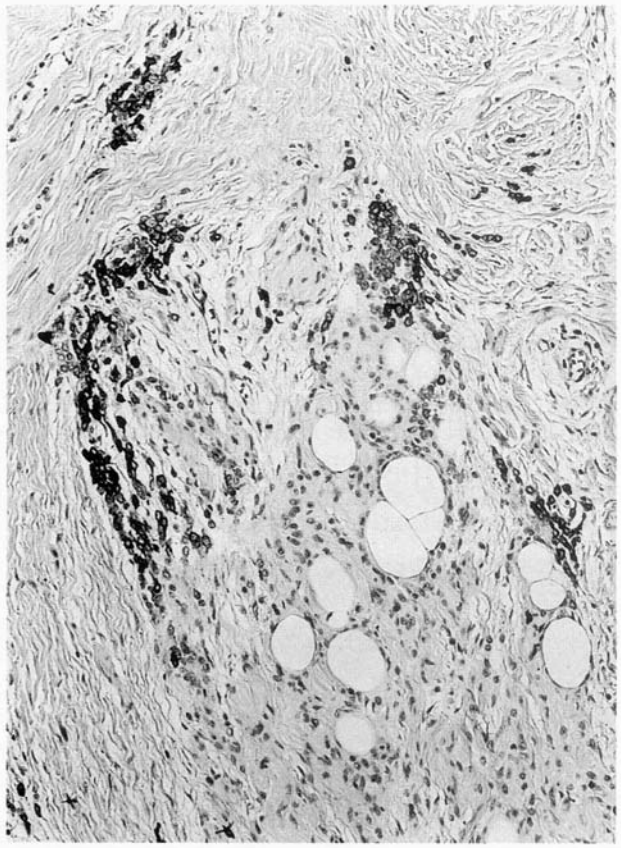

FIG. 2b

Naevus cells and melanocytes showing a positive reaction for Masson-Fontana silver stain. (Masson-Fontana silver stain; × 113).

no abnormal symptoms: glutamic oxaloacetic transaminase (GOT, Karmen method) 24 KU, glutamic pyruvic transaminase (GPT) 23 KU, gamma-glutamyl transaminase (γ-GPT, modified Orlowski method) 18 IU/l, lactate

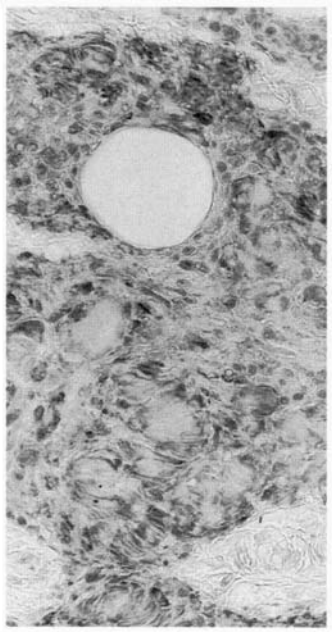

FIG. 2c

Naevus cells showing immunoreactivity for S-100 protein. Organoid structures are recognized in middle to lower portion. (ABC method; × 223).

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dehydrogenase (LDH, Wroblewski Unit) 290, total protein 7.4g/100 ml, total bilirubin 0.41 mg/100 ml, direct bilirubin 0.15 mg/100 ml. No history of denture use, long-term drug therapy, nor trauma to the site were mentioned.

Surgical removal of the mass, clinically diagnosed as a fibroma, was performed under local anaesthesia after histopathological diagnosis of intramucosal naevus was obtained by a prior biopsy.

There has been no evidence of recurrence for 16 months.

Pathological findings

The biopsy specimen revealed spiralling nests of round naevus cells in the dense fibrous tissue beneath the overlying epithelium. The resected specimen included non-neoplastic but hyperplastic salivary gland tissue measuring 13 × 12 × 10 mm on the section in addition to similar naevus nests (Figures 1a, b, 2a). In the deeper portion, nerve fibres and vessels were noted surrounded by adipose tissue. Adipose tissue, nerve fibres and vessels showed excessive growth for this location and some of the nerve fibres were sited beside the acinar structures of mucous salivary glands (Figures 3a, b).

Histochemically and immunohistochemically, pigment granules in melanocytes showed a positive reaction for Masson-Fontana silver stain and naevus cells positive for S-100 protein (Figures 2b, c). These findings confirmed the melanocytic component.

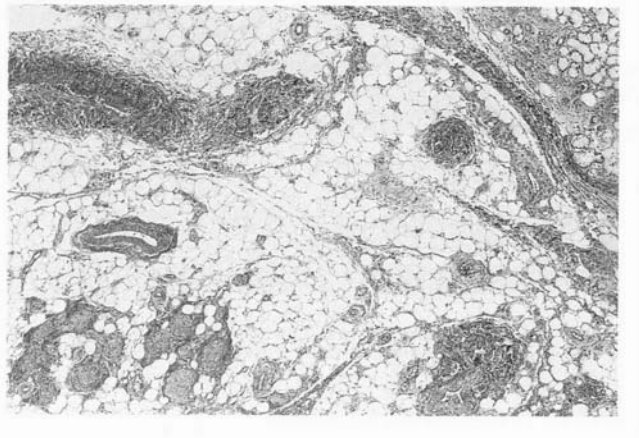

FIG. 3a

Nerve fibres and vessels involved in adipose tissue showing irregular cell sizes. (H & E; × 40).

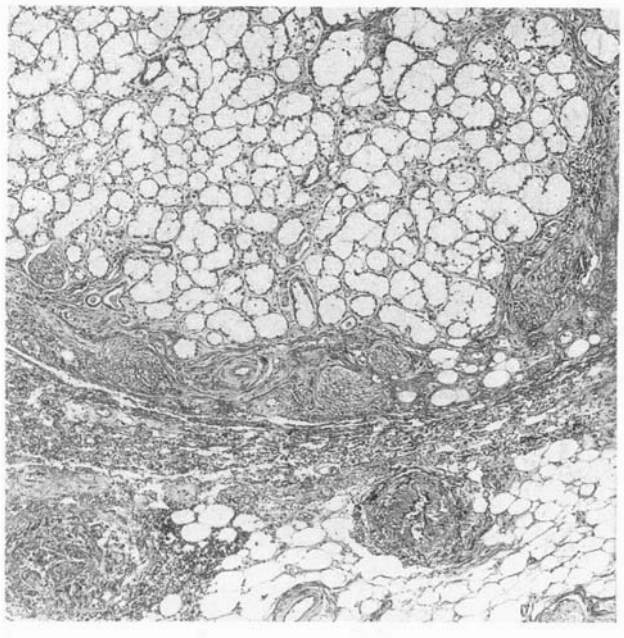

FIG. 3b

Salivary gland tissue surrounded by fibrous and adipose tissues. Note some nerve fibres located closely beside the mucous acini. (H & E; × 56).

Discussion

Buchner *et al.* (1990) analysed a large series of 130 oral pigmented naevi and recognized the hard palate as the favoured site. The age of patients ranged from four to 73 years with a mean age of 32 years. This lesion is assumed to be of neural crest origin (Mehregan and Hashimoto, 1991) and several investigators regard it as a hamartomatous lesion because of the relative overproduction of one element of the tissue without the tendency towards progressive growth in adult life.

Hyperplasia of the salivary gland has been designated by various terms, such as adenocytoma (Hendrick, 1964), simple adenoma (Tyldesley, 1967), adenomatoid hyperplasia of mucous salivary glands (Arafat *et al.*, 1981) and adenomatoid hyperplasia of minor salivary glands (Buchner *et al.*, 1991).

According to published reports, the age of the patients ranged from 24 to 55 years with a mean age of 39 years (Arafati *et al.*, 1981) or from nine to 79 years with a mean age of 44.5 years (Buchner *et al.*, 1991). Expected sites were the hard and soft palate and their junction, where eight out of nine cases of Arafat *et al.* (1981) appeared. Buchner *et al.* (1991) recognized 17 on the hard palate, 10 on the soft palate and six on the junction in their series of 40 cases.

Histological typing of salivary gland tumours (Seifert *et al.*, 1991) describes sialadenosis as a tumour-like lesion, which resembles in its asymptomatic, non-inflammatory and non-neoplastic character hyperplasia of the salivary gland, but is different in affecting serous acini, usually of bilateral parotid glands. This lesion is a disorder of the salivary gland innervation with peripheral autonomic neuropathy and has relation to endocrine disorders (diabetes mellitus, ovarian and thyroid insufficiency), malnutrition, chronic alcoholism, liver cirrhosis and dysfunction of the autonomic nerve system. However, none of these conditions were reported to affect minor salivary glands. Similarly, the relationship between hyperplasia of the minor salivary gland and routine drug use, denture use, and trauma has not been proved (Arafat *et al.*, 1981; Buchner *et al.*, 1991). Considering hyperplasia of minor salivary gland to be an idiopathic proliferation at present, we suggest that several cases in their series were hamartomas, although Arafat *et al.* (1981) stated they did not know whether not the lesions arose during development of the organ.

Hamartoma is a tumour-like malformation in which one or more normal tissue components of an organ are abnormally arranged, and is rare in the oral region. The recent literature describes a few cases arising in the tongue (Takato *al.*, 1985; Takimoto *et al.*, 1989; Miyamoto *et al.*, 1991; Owen *et al.*, 1993; Hanna *et al.*, 1994) and the gingiva (Kitano *et al*, 1991; Semba *et al.*, 1993; Mesa *et al.*, 1994), but seldom seen on the palate. They have various elemental tissues such as squamous (Mesa *et al.*, 1994) and odontogenic epithelium (Kitano *et al.*, 1991), mucous or serous salivary gland (Takato *et al.*, 1985; Takimoto *et*

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al., 1989; Miyamoto *et al.*, 1991; Owen *et al.*, 1993; Hanna *et al.*, 1994), nerve fibre (Owen *et al.*, 1993; Semba *et al.*, 1993; Mesa *et al.*, 1994), smooth muscle (Takao *et al.*, 1985; Takimoto *et al.*, 1989; Owen *et al.*, 1993; Semba *et al.*, 1993; Hanna *et al.*, 1994), and adipose tissue (Takimoto *et al.*, 1989; Owen *et al.*, 1993; Hanna *et al.*, 1994). Salivary gland, smooth muscle and adipose tissue are common and the former includes the small lobular (Takato *et al.*, 1985; Takimoto *et al.*, 1989; Owen *et al.*, 1993; Hanna *et al.*, 1994) and large massive glands (Miyamoto *et al.*, 1991). Some consist of a haphazard mixture of elemental tissues and others show polar organization of mucous and serous glands (Miyamoto *et al.*, 1991). Tsuda *et al.* (1987) reported a hamartoma in the parotid gland, which consisted of well-differentiated organoid structures with normal mucous and serous acini, ducts, fat and lymphoid tissues. They stated that the possibility of hyperplasia as well as neoplasia could not be excluded, although it was pathologically diagnosed as a hamartoma.

As to the pathogenesis of the present case, we need to consider the following four possibilities:

(1) Traumatic or reactive pathogenesis: Trauma to the site and denture use were not mentioned. Reactive proliferation caused by repeated minor injury to a preceding naevus could not be excluded. However, there was no evidence that the naevus preceded the remainder of the lesion.

(2) A vague history of a fatty liver and current alcohol ingestion of half a bottle of beer daily did not prove alcoholism.

(3) A merely incidental combination: as both pigmented naevus and hyperplasia of minor salivary gland have the same predilection for the palate and middle age as noted above, this pathogenesis could not be excluded. The clinical evidence favoured the formation of a single lesion.

(4) Hamartoma-like pathogenesis: we favour this possibility, firstly because both pigmented naevus and hyperplasia of minor salivary gland are possible as hamartomas and secondly because a co-incidental formation seems too unlikely as has been noted above. We cannot tell, like Arafat *et al.* (1981), whether or not this lesion arose during development of the organ. However seven out of 11 cases that Kitano *et al.* (1991) listed as odontogenic epithelial hamartomas were present in patients older than 40 years probably because of their asymptomatic courses and they may have actually arisen at a far younger age.

Adipose tissues, which were abundant for the location and surrounded the salivary gland, and the nerve fibre bundles closely sited to mucous acini may support this hypothesis.

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