

An Oral 'Follicular' Choristoma Presenting in the Anterior Floor of the Mouth

V. SOOD

Abstract: A 46-year-old Asian man presented with hair growing from the anterior floor of his mouth. The diagnosis of this lesion was an oral choristoma of a 'follicular' variety. A choristoma is defined as an overgrowth of normal tissue at an abnormal site. Only one other case of this particular lesion has been reported to date. This article proposes mechanisms as to the aetiology of this lesion, and its inclusion in the classification of oral choristomas.

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Clinical Relevance: Oral mucosa is derived from ectoderm and endoderm, therefore it has the potential to form normal structures not usually associated with the oral cavity

A choristoma is defined as an overgrowth of normal tissue at an abnormal site. Choristomas are distinguished from hamartomas in that the latter are overgrowths of normal tissue in their normal location, for example an odontome. Neither are neoplastic.

Chou *et al.*¹ devised a classification of oral choristomas, but did not include an oral follicular variety, as only one case² had been reported at that time. To date, the case described here is the only other that has been reported.

CASE REPORT

A 46-year-old Asian man, fit and well, presented with hair growing from a point where the lingual frenum meets the anterior floor of the mouth. This had been growing for at least 3 years before referral. He had no history of earlier surgery at the site. The patient's only complaint was of the discomfort

of having hair present in his mouth (Figure 1).

Several grey hairs approximately 2 cm long could be seen. No mass could be palpated in the floor of the mouth. An ultrasound investigation of the sublingual region was performed but no abnormal mass or lesion was identified.

Despite this, a provisional diagnosis of hairs growing from the lining of a sublingual dermoid cyst was made. The patient was admitted for an excisional biopsy of the lesion.

Surgical access to the lesion was made intra-orally through the floor of the mouth and blunt dissection was used to excise the lesion. The incision site was closed with sutures and the wound healed uneventfully.

The gross appearance of the biopsy was of soft tissue measuring 1.0 x 1.0 x 0.5 cm with shafts of hair 2 cm long extending from the surface (Figure 2). The histopathological appearance showed an involution of surface epithelium, lined by pigmented epidermis containing sebaceous glands, sweat glands, and mature hair follicles (Figure 3). The rest of the lesion was normal mucosa and submucosa with no evidence of cyst lining at any point.

These findings, together with the

clinical examination and unremarkable ultrasound scan, were consistent with an oral 'follicular' choristoma.

DISCUSSION

Intra-oral hair may be found in people who have undergone reconstructive surgery with hair-bearing skin grafts or free flaps (Figure 4): if the graft takes or the flap remains vital the hair will continue to grow. Although this can be uncomfortable for the patient, the hair can be trimmed and tolerance develops.

A case has been reported in which oral hair could be seen growing unrelated to reconstructive surgery: Mitchell reported hair growing from the socket of a spontaneously exfoliated lower incisor.³ This anomaly was attributed to a mechanism in which a periodontal abscess associated with the exfoliated tooth migrated to the skin. It was postulated that facial hair from a deep midline chin cleft had invaginated into the sinus, possibly in the presence of distended sebaceous glands and hair follicles.

However, only one case other than this has been reported, this being



Figure 1. Intra-oral appearance at presentation: hairs are visibly growing inside the mouth.

V. Sood, BSc, BDS, FDS RCS (Eng.), Clinical Medical Student, Guy's, King's and St. Thomas' Schools of Medicine, Dentistry and Biomedical Sciences, Guy's Hospital, London.



Figure 2. Excisional biopsy of lesion revealed a soft tissue mass (approximately 1.0x 1.0 x 0.5 cm) with 2 cm long hairs extending from its surface.

unrelated to adjacent pathology or previous reconstructive surgery: Arwill *et al.*² reported a nodular lesion on the lingual aspect of the lower central incisors, associated with the gingivae, of a young girl. The lesion was excised and histopathological examination revealed a lesion consisting of hair follicles, sebaceous glands, melanocytes and keratocysts. Unlike the case being reported here, no hairs were evident clinically.

Dermoid cysts are not uncommon in the floor of the mouth and are known as sublingual dermoid cysts.⁴ They are known to contain dermal appendages, including hair follicles, in their epithelial lining and are thought to be the result of an abnormality of development of the branchial arches or pharyngeal pouches.

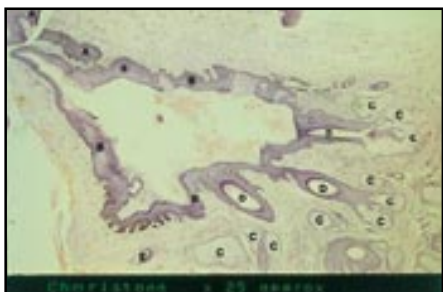


Figure 3. Histopathological slide of lesion. The histopathological slide is oriented with the surface epidermis (A) associated with the lesion at the bottom of the photograph. Involution of the epidermis (B) is present with associated sebaceous glands (C), mature hair follicles (D) and sweat glands (E). Unfortunately no shafts of hair are present in this plane of section.

Embryology of Oral Development

To explain the development of these lesions, it is worth reviewing the embryology of the mouth. The epithelium of the oral cavity is formed from the ectoderm derived from the stomatodeum and endoderm from the foregut. They are initially separated by the oropharyngeal membrane until this degenerates on the 28th day of development and both linings merge.⁵ There is disagreement as to where the dividing line of oral epithelium derived from ectoderm and endoderm lies.

One proposition for the development of the anterior floor of the mouth and tongue is that the tongue is separated by a downgrowth of ectoderm between it and the developing gingiva. This ectoderm subsequently degenerates with the formation of the linguogingival sulcus.⁶ Hence, the inner surface of the gingivae can be regarded as the dividing line between the ectoderm and endoderm.

Another theory suggests that, owing to the extensive changes occurring in oropharyngeal development, the line of division is difficult to trace, and that the presumed site of the original oropharyngeal membrane in the adult is an imaginary vertical plane from the posterior border of the body of the sphenoid bone through the tonsillar region of the fauces.⁵ This assumption is made as the anterior lobe of the pituitary gland (adenohypophysis) originates from Rathke's pouch anterior to the oropharyngeal membrane.

The derivatives of ectoderm and endoderm have been stated.⁷ *Ectoderm* gives rise in general to external organs and structures:

- the central nervous system;
- the peripheral nervous system;
- the sensory epithelium of the ear, nose and eye; and
- the epidermis, including the hair and nails.

In addition it gives rise to the subcutaneous glands, the mammary glands and the enamel of the teeth.

Endoderm gives rise to:

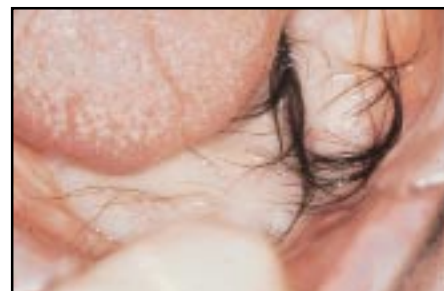


Figure 4. Hairs growing from a radial forearm free-flap graft used for oral reconstruction. (Courtesy of Mr D.A. Mitchell.)

- the gastrointestinal tract;
- the epithelial lining of the respiratory tract;
- parenchyma of the tonsil, thyroid, parathyroids, thymus, liver, and pancreas;
- the epithelial lining of the urinary bladder and urethra; and
- the epithelial lining of the tympanic cavity and eustachian tube.

From the wide variety of derivatives of endoderm and ectoderm, it is not unreasonable to question why a number of different tissues or structures do not develop in the oral cavity where the two embryonic tissues meet.

Development of 'Follicular' Choristoma

To explain the development of the oral 'follicular' choristoma, several mechanisms may exist:

1. It is probable that ectodermal epithelium can remain as islands in the floor of the mouth and not



Figure 5. Abundant Fordyce's spots in buccal mucosa. (Courtesy of Dr A. High.)

- degenerate.
2. Ectodermal epithelium in the oral cavity has the ability to form dermal appendages.
 3. If ectoderm can remain submucosally it has the potential to become a sublingual dermoid cyst, which can form dermal appendages.
 4. The spontaneous marsupialization of an undetected sublingual dermoid cyst with growth of hair from its lining.

Unfortunately, none of these mechanisms can account for why hair is not seen more often in the oral cavity, unlike other dermal appendages which

are more abundant in the oral mucosa – such as sebaceous glands (Fordyce’s spots) and melanocytes (Figure 5). However, this lesion should be included in the classification of oral choristomas.

ACKNOWLEDGEMENTS

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BOOK REVIEW

Oral Diagnosis. The Clinician’s Guide. By Warren Birnbaum and Stephen M. Dunne. Butterworth Heinemann, Oxford, 2000. (320pp., £17.99p/b). ISBN 0 7236 1040 1.

This book is aimed at undergraduate dental students and the newly qualified. The authors acknowledge feedback from their past and present students and this seems to have paid off. The text format, the easy to read bullet points and the summary start to each chapter were definitely approved of by the dental students at Liverpool. There is also a suggested short further reading list at the end of each chapter.

The opening chapter, ‘The challenge of diagnosis’ includes information such as establishing a rapport with the patients. Subsequent chapters deal very thoroughly with history-taking, examination and diagnostic tests. The dental diagnostic tests range from the routine, such as vitality, through to multiple biopsy techniques and an excellent section on venepuncture. Normal value ranges are given for haematological tests along with useful clinical information explaining abnormal results. Equally detailed consideration is given to dental and non-dental causes of pain. The trauma section is short but this is because the authors have purposely limited the length of the book. The chapters on infection, cysts, ulcers and white patches

are also excellent but there is a lack of photographs which is understandable as it would require a colour atlas to complement this text. The ‘Bumps, lumps and swellings’ chapter gives a differential diagnosis list, which is clear. Symptoms, signs and diagnostic tests are given for all of the discussed lesions. The final chapters discuss the oral changes associated with both systemic disease and medication.

Overall, this is a comprehensive and clearly written text which my students and the vocational trainees found helpful and user friendly. I would recommend this book without hesitation.

Martin Forde

GDP, Liverpool and Part-time Lecturer in Clinical Dental Sciences University of Liverpool Dental School

ABSTRACT

A CAUTIONARY TALE!

A Cautionary Tale. Case Report. P.N. Fatouris. *Australian Dental Journal* 2000; **45**: 53-54.

As well as presenting a most interesting case, this paper highlights the fact that general practitioners frequently encounter interesting and unusual cases. I believe that far more of these cases should be submitted for publication and discussion. We can all learn from the

experience, and mistakes, of others!

A patient presented to her general medical practitioner with a small painless lump on her cheek. The doctor made a diagnosis of sebaceous cyst, which was removed surgically under general anaesthesia with antibiotic cover. The lesion recurred, and the patient was referred to a plastic surgeon, who repeated the surgery, again under general anaesthesia and again with antibiotics.

When the lesion recurred yet again it was explored under local anaesthesia, with further antibiotic therapy. This

time a sinus tract was discovered leading to the apex of a crowned but symptomless upper first premolar. When root canal treatment was undertaken the facial lesion resolved within one week.

The case has a happy ending, however, as although the patient is left with considerable facial scarring and deformity, she accepts that she was the recipient of bad luck, not bad management, and is not seeking any redress through litigation.

Peter Carrotte
Glasgow Dental School