Case Report: Enlarging symmetrical masses of the palate of idiopathic etiology [version 1; peer review: 1 approved, 1 approved with reservations]

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Abstract
We report the case of a 33 year-old woman who came to our attention with slowly enlarging exophytic masses of the palate, histologically characterized by sub-epithelial fibrous proliferation with packed collagen bundles and increased fibroblasts number. We describe the condition of idiopathic fibrous hyperplasia, its diagnosis and its surgical treatment, which in our case was carried out with the aid of a custom made thermal printed plaque used as a scaffold.

Keywords
fibrous hyperplasia, palate, idiopathic, gengival hyperplasia
Introduction
Idiopathic fibrous hyperplasia is a rare benign condition, characterized by a slow and progressive increase in gingival volume\(^1\). It manifests as a rosy swelling of hard consistency while, at histological examination, it is characterized by a proliferation of fibroblasts in a myxomatous stroma. We describe and discuss a case of idiopathic fibrous hyperplasia of the palate.

Case
A 33 year-old woman came to our attention with slowly enlarging exophytic masses of the palate, which had begun to grow 2 years before and caused her disturbances of phonation as well as in swallowing solids and liquids. The patient did not take any drugs; however she had been a frequent user of nonsteroidal anti-inflammatory drugs for the last 3 years because of chronic back pain. Remote personal and family histories were negative, except for recurrent gastric nuisance and back pain.

Clinical and rhinoscopy examination demonstrated bilateral and symmetrical hypertrophic tissue, of hard consistency and rosy color, at the posterior-later area of the palate, with a tendency to coalesce medially. This tissue was contiguous to the adjacent gingiva (Figure 1).

Computerized tomography scan showed such lesions to be limited to the mucosal palate, with no underlying bone involvement (Figure 2).

An incisional biopsy demonstrated a sub-epithelial fibrous proliferation with packed collagen bundles and increased fibroblasts number (Figure 3). We concluded for a diagnosis of localized idiopathic fibrous hyperplasia. We treated the patient with a personalized approach using surgical resection and insetting of a thermal-printed palate plaque (Figure 4). We obtained good functional results at 20 days post-op (Figure 5), and no sign of recurrence at the 12 months follow up (Figure 6).
Secondary forms associated to pregnancy, scurvy, leukemia and drugs are also known. Among the latter, various chemotherapy agents can elicit secondary forms, including ipilimumab and vemurafenib. However, no reports link gingival fibrous hyperplasia to the drugs administered in our patient. The localized form has its onset from the second decade, does not generally recur after surgery, and normally is not associated to genetic predisposition, although investigations to exclude syndromes commonly associated to gingival fibromatosis should always be carried out in our experience; these include Laband, Rutherfurd, Cross and Ramon syndromes. In both the localized and the generalized forms, local factors such as dental plaques, caries, and the action of chemical substances and their metabolites might contribute to the onset in susceptible patients. The precise pathogenic mechanism of idiopathic forms is unknown, but it appears to confine to the gingival and mucosal fibroblasts with no involvement of the periodontal ligament or the palate underlying bones.

Regardless of the etiology, excess tissue removal is the treatment of choice in localized fibrous hyperplasia, either by scalpel or by CO₂ laser. Our personal approach includes the use of a thermal-printed palate plaque (Figure 4), to be left in place for 20 days after surgery (Figure 5), which in our experience yields excellent hemostasis by exerting compression and, at the same time, functions as a scaffold that promotes and guides second intention healing, preventing recurrence of exuberant tissue growth. The long term (12 months) results reported in our case testify to the efficacy of such an approach.

Consent
Written informed consent for publication of their clinical details and clinical images was obtained from the patient.

Competing interests
No competing interests were disclosed.

Grant information
The author(s) declared that no grants were involved in supporting this work.

Discussion
Two main subtypes of fibrous hyperplasia are known: generalized and localized. Generalized form has a genetic predisposition, appears at decimal or definitive dental eruption, and usually demonstrates a tendency to recur after surgery.

References
Open Peer Review

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This Case Report is of scientific interest and well written. A more detailed explanation of therapy would be appreciated. About the thermal printed palate plaque for instance: what material is it made of? Is it a mold or 3D printed? Is it placed immediately after surgery and left in place 24 hours a day? Is thermal plaque therapy a novel approach or are there any references about this kind of therapy?

Is the background of the case's history and progression described in sufficient detail?
Yes

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?
Yes

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?
Yes

Is the case presented with sufficient detail to be useful for other practitioners?
Partly

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Plastic surgery, burns, aesthetic surgery

We confirm that we have read this submission and believe that we have an appropriate level
of expertise to confirm that it is of an acceptable scientific standard, however we have significant reservations, as outlined above.

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I believe the article is well structured and describes an interesting presentation of a rare benign condition.

I must congratulate the authors on the quality and quantity of the photographic material. The case is really well documented with clinical pictures, histologic images and radiologic findings.

I have personally treated a couple of these cases but often experienced short to medium term recurrence. The alternative treatment described by the authors (the use of a guiding plaque after removal) is interesting and seems to warrant better long term outcome.

Is the background of the case’s history and progression described in sufficient detail?
Yes

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?
Yes

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?
Yes

Is the case presented with sufficient detail to be useful for other practitioners?
Yes

Competing Interests: No competing interests were disclosed.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.
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