

## CASE REPORT

# Bilateral tongue fibrous hyperplasia: A case report and literature review.

## Hiperplasia fibrosa bilateral de língua: relato de um caso e revisão da literatura.

Gabriel Oliveira Cavezzi<sup>1</sup>  | Orlando Cavezzi Junior<sup>2</sup> 

### OPEN ACCESS

#### Institutional Affiliation

<sup>1</sup> Universidade Estadual de Londrina, Faculdade de Odontologia, Londrina, Paraná, Brazil.

<sup>2</sup> Private practice, Avaré, São Paulo, Brazil.

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#### Correspondence:

Gabriel Oliveira Cavezzi – Rua Joaquim Lacerda nº60 Apto 402 Londrina, Paraná Brazil CEP 86015-260 Tel. 55-14-996307421 email: [gabcavezzi@gmail.com](mailto:gabcavezzi@gmail.com)

#### ABSTRACT

Focal fibrous hyperplasia (FFH) is a pathology characterized by tissue enlargement as a result of increasing the number of inflammatory cells which receive chronic mechanical stimuli of low intensity that may affect gums, lip, palate, jugal mucosa and tongue. The aim of this article is to report a case of FFH on the lateral border on both sides of the tongue associated with the use of the Hyrax appliance in a hebiatric patient, whose chief complaint was chewing discomfort. This case was diagnosed clinically and by histopathology analysis and treated by surgical excision as well as the removal of Hyrax appliances. Follow-up care provided the recovery of the patient's quality of life.

#### KEYWORDS

Reactive Hyperplasia, Focal Fibrous Hyperplasia, Orthodontic Appliance, Hyrax appliance, Tongue

#### RESUMEN

La hiperplasia fibrosa focal (HFF) es una patología caracterizada por el aumento de tamaño de los tejidos como consecuencia del aumento del número de células inflamatorias que reciben estímulos mecánicos crónicos de baja intensidad que pueden afectar a encías, labio, paladar, mucosa yugal y lengua. El objetivo de este artículo es reportar un caso de FFH en el borde lateral a ambos lados de la lengua asociado al uso del aparato Hyrax en un paciente hebiátrico, cuyo principal motivo de consulta era la molestia masticatoria. Este caso fue diagnosticado clínicamente y por análisis histopatológico y tratado mediante escisión quirúrgica así como la retirada de los aparatos de Hyrax. La atención de seguimiento permitió la recuperación de la calidad de vida del paciente.

#### PALABRAS CLAVE

Hiperplasia reactiva; Hiperplasia fibrosa focal; Aparato de ortodoncia; Aparato Hyrax; Lengua.

### Clinical Relevance

The clinician must always be aware of the possible occurrence of atypical lesions, especially on the lateral border of the tongue. Although histological investigations are almost always underestimated by clinical aspects in such lesions, it is mandatory to perform a histopathological analysis.

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## CLINICAL RELEVANCE

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## INTRODUCTION

The oral cavity may act as one of the common sites for development of any injury due to constant stimuli and, therefore, reveals a variety of clinically similar diseases that often presents a diagnostic challenge, especially, soft tissue enlargements. These lesions are usually hyperplastic reactions (i.e., reactive hyperplasia) and are not considered as neoplasms.<sup>1</sup> Although benign in nature, they do have a tendency toward recurrence with incomplete removal of the lesion or the local irritants involved at the site.<sup>2</sup> These lesions occur most routinely on the buccal mucosa, gingiva, lips, and tongue but can arise at any other soft tissue site. The reactive hyperplasia can be classified into four groups: Focal Fibrous hyperplasia (FFH), Pyogenic Granuloma (PG), Peripheral Ossifying Fibroma (POF) and Peripheral Giant Cell Granuloma.<sup>3</sup>

The histopathological features of these lesions are entirely distinct, but significant overlaps are detected among them. Accordingly, this may be associated with the connective tissue response to varying intensities of irritation.<sup>2</sup> The focal fibrous hyperplasia (FFH) is also known as irritation fibroma, traumatic fibroma, fibrous nodule, or fibroma, which is often due to trauma or local irritation as the causative factor.<sup>1</sup> Clinically, the FFH is generally painless, has a variable growth rate, may be pedunculated or sessile, firm consistency mass with a smooth surface, and is usually the same color as the surrounding mucosa.<sup>3</sup> The FFH is more common among females than males and most common in the third to sixth decades of life.<sup>1-3</sup> Microscopic examination of the FFH reveals a nodular mass of fibrous connective dense tissue covered by stratified squamous epithelium. The lesion is not encapsulated, and the fibrous tissue merges gradually into the surrounding connective tissues. The collagen bundles may be arranged in a radiating, circular, or even casual fashion. The covering epithelium typically demonstrates atrophy of the rete ridges due to the underlying fibrous mass.

Secondary trauma may exhibit hyperkeratosis on the surface of the lesion and the scattered inflammation may be seen, most regularly beneath the epithelial surface. Frequently this inflammation is chronic and consists mostly of lymphocytes and plasma cells.<sup>1</sup>

The treatment of choice is conservative surgical excision along with the causative irritant removal. Nevertheless, the follow-up should be done regularly. The recurrence is rare, but the excised tissue must be sending to the histopathological analysis because other benign or malignant tumors may mimic the clinical appearance of a FFH. This paper shows a case of a manifestation of focal fibrous hyperplasia on the lateral border on both sides of the tongue which was excised by surgical blade along with removal of Hyrax appliance successfully.

## CASE DESCRIPTION

A 13-year-old Caucasian male was referred by his orthodontist to our dental private service with a chief complaint of growth on both sides of the tongue (Figure 1). No relevant medical history was reported. The intra-oral examination showed an exophytic, sessile, and fibrotic growth on the lateral border of on both sides of the tongue and the presence of Hyrax appliance in the upper dental arch. The lesions had a defined boundary, and smooth surface, and the color was similar to adjacent mucosa. The extent of lesion on the left side was  $3,0 \times 1,5 \times 0,5$  cm in its largest axis, whilst the lesion on the right side was  $1,0 \times 0,7 \times 0,7$  cm in its largest axis.

The patient complained that the growth was interfering with chewing and was uncomfortable but painless. Based on clinical findings, a provisional diagnosis of fibrous hyperplasia was given. Before initiating therapy, the surgical treatment plan was discussed and approved by the patient and his parents. After providing all explanations, the informed consent was taken. Furthermore, the parents authorized the publication of the present clinical case respecting the confidentiality of the patient's personal information. After obtaining hematological investigations, an excisional biopsy was performed under local anesthesia with 2% lidocaine with 1:100,000 epinephrine. After anesthetizing around the lesion by infiltrating, the overgrowth mass was held with an Allis tissue grasping forceps, and it was excised completely from its base using a surgical scalpel blade 15C. The right-side wound was sutured using a 4.0 black mononylon suture (Ethicon –

J&J- Brazil) while the left-side wound was sutured using a 4.0 black silk suture (Ethicon – J&J- Brazil). The excised specimens were fixed in 10% formalin and were sent for histopathological analysis. Post-operative instructions were given to the patient and analgesics were prescribed.

The post-operative course was uneventful and one week later the sutures were removed.

The follow-up was three months (Figure 2). The histopathological analysis showed parakeratinized stratified squamous epithelium with thin epithelial extensions that project into the underlying connective tissue (i.e., rete ridge or rete pegs) and the stroma was composed of fibrous connective tissue, chronic inflammation and few blood vessels that confirmed the diagnosis of FFH (Figure 3).



**Figure 1.** Clinical feature showing swelling on the right side of the face. B, surgical excision of the lesion. C, immediate postoperative period. D, clinical feature after 12 months of follow-up, showing normal healing and no signs of recurrence.



**Figure 2.** Three months of the postoperative follow-up period.

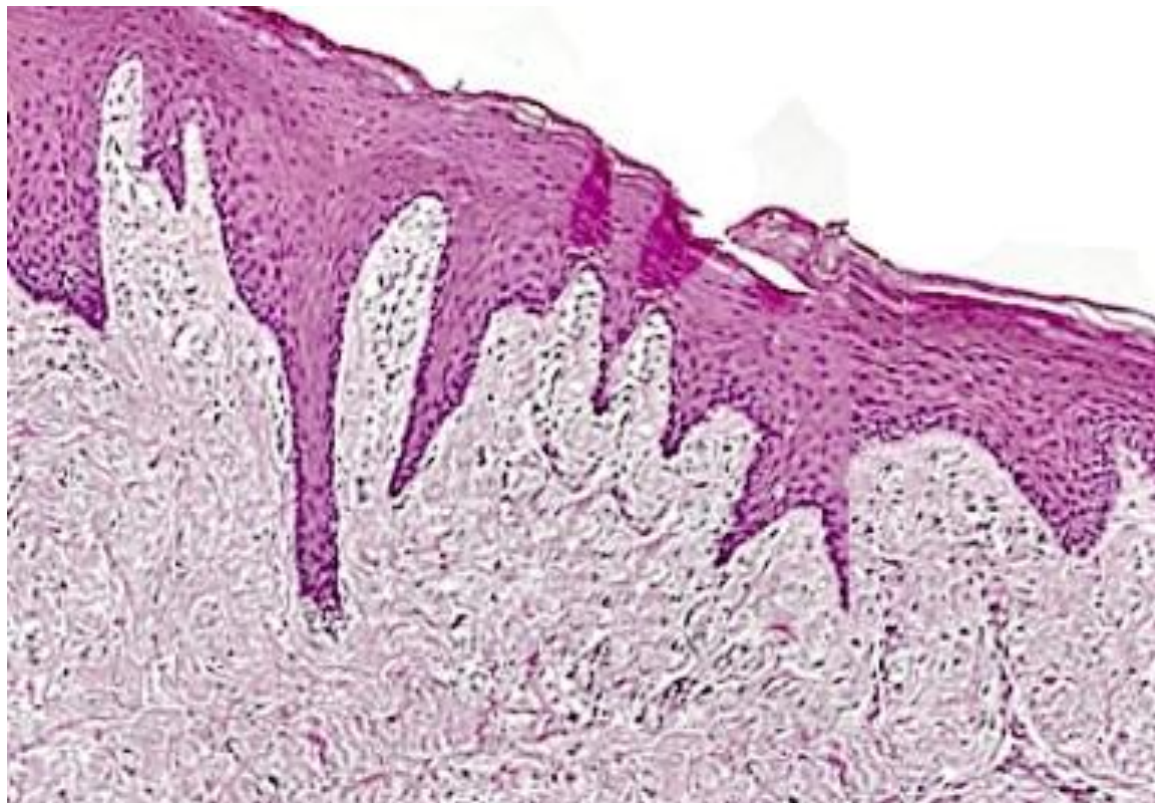


Figure 3. Figure 3. Histological aspect of growth (H&E) stained section (10X).

## DISCUSSION

Oral soft tissue lesions are frequent in clinical practice, and reactive lesions probably represent one of the most common conditions in the oral cavity because of the high frequency of tissue injuries. Focal fibrous hyperplasia arises as a result of a chronic repair process that comprehends granulation tissue and scar formation, resulting in a submucosal fibrous mass.<sup>4</sup> This term is preferable rather than the term “fibroma.” Because it is not a real neoplasm, but focal hyperplasia of fibrous connective tissue induced by local trauma or chronic irritation.<sup>5</sup> Although the term focal fibrous hyperplasia must be more suitable for the clinical appearance and pathogenesis of this entity, it is not commonly used. In this case report, we describe a surprisingly negative effect of Hyrax appliance use in a 13-year-old Caucasian male that provoked a reactive lesion named fibrous hyperplasia on the lateral border on both sides of the tongue.

This lesion is frequently a well-defined, slowly growing that can affect any age of life, but cases are concentrated from second to sixth decade of life.<sup>2-3-5-14</sup> As regards the female-male ratio, the literature points out a prevalence of females.<sup>2-3-5-16</sup> In most cases, the female population is usually greater than the males because women are more likely to seek dental assistance than men.

If women are more susceptible to having oral reactive lesions, this is still unclear. In contrast, a study from India and Iran reported a male predominance.<sup>2-5</sup> While another reported no gender predilection.<sup>6-10</sup> Albeit, the occurrence of FFH in the tongue is not uncommon, some studies have shown the low prevalence of FFH in the tongue<sup>6-12-17-18</sup> whilst others not.<sup>2-5-7-9-13-19-20</sup> Furthermore, the preponderance of Caucasians is remarkable.<sup>21</sup> Clinically, the FFH can be sessile, wide base, or occasionally pedunculated, resembling a stalk.<sup>22</sup> The growth, in this case, was attached to the underlying tissue with a wide base and was excised with a safety margin. Mostly growths appear as an elevated nodule with similar color to the adjacent normal mucosa, smooth surface, and is usually painless as seen in this case report. Although the literature showed a painful symptomatology.<sup>9-10</sup>

The size of FFH lesions can vary from a few millimeters to several centimeters. However, most lesions are lesser than 2.0 cm.<sup>9-10-14-15-23</sup> The size of this lesion is related to the exaggerated degree of one or more components of the inflammatory reaction and the healing response. Accordingly, represents an overexuberant repair process with the proliferation of scars. Most of these lesions occur

limitrophe to the oral mucosa surface, where irritating factors are quite common and therefore are subject to continuous chewing trauma.

The use of Hyrax appliance generally takes 4 to 6 months and probably this lesion emerged during this period caused by the trauma of the appliance. Therefore, the duration of disease from the initial notice of the lesions until the date of referral to our service had passed at least six months. Although easy to examine, most oral lesions are difficult to provide exact diagnosis clinically. Thus, it is necessary to obtain a differential diagnosis detailing the medical and dental history of the patient further attentive history of the lesion.

The differential diagnosis of FFH should include increasing attention when the site is the tongue, even at an early age. It must consider other lesions including neurofibroma, neurilemmoma, giant cell fibroma, peripheral giant cell granuloma, peripheral ossifying fibroma and benign and malignant salivary gland tumors.<sup>22</sup> Nevertheless, the FFH lesion is relatively effortless to solve and should be removed to rule out other pathologic processes, so histologic analysis must be necessary to reach exact diagnosis. The most common way to remove the FFH is by scalpel incisions, along with complete removal of local irritants and follow-up care. Recurrences of this lesion are uncommon or rare. However, Manchanda et al.<sup>10</sup> had 3 recurrences (2.5%) in 117 cases of focal fibrous hyperplasia, while De Santana Santos et al.<sup>9</sup> had two recurrences (1.0%) in 193 cases. Hence, attention should be given at the time of complete removal of the lesion and the local irritants involved at the site.

In addition, it is important to point out some limitations applicable in terms of diagnosis, treatment, and post-surgical controls. The diagnosis of this condition can be difficult due to the nonspecific clinical features of this lesion, especially on the lateral border of the tongue, making the diagnosis challenging but, in this case, is noteworthy to emphasize the presence of triggering factor of the lesion that was the presence of the Hyrax appliance. It is essential to rule out malignancy to avoid misdiagnosis and inappropriate treatment. The biopsy is mandatory for the assessment of histopathology to confirm the diagnosis. Additionally, the treatment can be complex by the proximity of the lesion to critical structures. The delicate nature of the tongue, a highly vascular and mobile organ, makes the procedure laborious as seen in this case. Post-surgical controls are also essential to monitor for any signs of recurrence or complications, which may require additional interventions. Consequently, the limitations in

the diagnosis, treatment, and post-surgical controls of FFH highlight a comprehensive care for patients with this condition.

## CONCLUSION

Focal fibrous hyperplasia is not an uncommon lesion in the tongue, but the present case brought a singular manifestation on the lateral border on both sides of the tongue of the patient that wearing a Hyrax expander. Proper history taking, careful clinical examination and histological analysis are keys to make a proper diagnose of the lesion and planning the modality of treatment. The lesion was successfully excised by scalpel and complete removal of local irritants, with follow-up care to prevent the recurrence.

## CONFLICTS OF INTEREST

The authors declare no conflict of interest.

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