Gingival salivary gland choristoma: An unusual case report

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INTRODUCTION

The gingival salivary gland choristoma (GSGC) is a highly unusual tumor-like mass alteration constituted by normal salivary gland cells in an abnormal location, reported for the first time in 1964 by Moskow and Baden.1 To our knowledge, only 12 cases of this entity (including present case) are reported to date, the majority observed as asymptomatic, solitary, smooth-surfaced tumor-like masses, measuring 0.5–1.5 cm with no osseous involvement. This case involves a 38-year-old female, with a pink symptomatic polypoid nodule on the posterior mandibular alveolar ridge mucosa, measuring 6 mm in diameter where no osseous abnormalities are shown. An excisional biopsy was performed. Microscopically, the specimen was constituted by dense fibrous connective tissue containing mucous minor salivary glands, intraductal calcification and adipose tissue clusters. Furthermore, inflammatory infiltrate foci were seen. An immunohistochemical technique was used as the support for the diagnostic methodology. The diagnosis of gingival salivary gland choristoma was established. Some development theories are discussed, referring to a pluripotential capacity of the gingiva. This case reflects the importance of not underestimating innocuous lesions that could represent more serious or unusual entities. In addition, histopathological analysis is mandatory to achieve a correct diagnosis and management of soft-tissue enlargements of oral mucosa.

Keywords: Alveolar ridge, gingival choristoma, heterotopic tissue, polypoid nodule, salivary gland

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CASE REPORT

A 38-year-old female, was referred to the oral medicine service at the University of Guadalajara. At the diagnosis interrogation, the patient reported a 3-month symptomatic enlargement and did not mention any antecedents of importance. After obtaining consent from the patient, we proceed to intraoral exploration that revealed a polyposic nodule of approximately 6 mm in size, with smooth surface, firm consistency and similar coloration to the normal adjacent oral mucosa, the lesion was located on the posterior alveolar ridge mucosa close upon a root remnant of the mandibular right second molar [Figure 1a]. A periapical radiography was applied, where no osseous or dental root abnormalities are shown [Figure 1b]. An excisional biopsy was performed under local anesthesia, and the obtained tissue was submitted to the histopathological analysis.

Microscopically, the specimen was constituted by a nodular mass of dense fibrous connective tissue [Figure 2], containing mucous minor salivary glands [Figure 3a], medium caliber arteries and lymphatic vessels. Adjacent to salivary acini, intraductal calcification [Figure 3b] and adipose tissue clusters [Figure 3c] could be observed. In the overall stroma, mild inflammatory infiltrate foci with predominance of lymphocytes were observed.

Although it is not necessary to establish the diagnosis, an immunohistochemical technique was performed to assess the proliferative activity of gland cells through the evaluation of Ki-67 expression, where it was observed that the majority of glandular cells show lack of expression [Figure 4]. In addition, positive expression of Ki-67 was found in the 2% and 4% of the nuclei of acinar and stromal cells, respectively. Based on the clinical and histopathological findings, the diagnosis of GSGC known as heterotopic salivary gland tissue was established. After the excisional biopsy, postsurgical care was indicated to the patient. The relief of occasional pain was achieved, and there was no recurrence after 12 weeks of follow-up.

DISCUSSION

According to the clinical and radiographic findings, this injury can be diagnosed with a reactive origin tumor-like mass due to the close relationship of the alveolar ridge with the occlusal contact, similar to an irritation fibroma, which usually is a well-delimited smooth surface where a firm consistency is observed on the nodule. This can vary from

Table 1: Gingival salivary gland choristomas reported to date

| Author/year | Age (years) | Gender | Region            | Histopathologic findings                                      |
|-------------|-------------|--------|-------------------|----------------------------------------------------------------|
| Traeger(19)/1961 | 19         | Male   | Posterior maxilla | Gingival cyst and lobule of mucous salivary gland               |
| Moscow and Baden(19)/1964 | 35         | Male   | Posterior maxilla | Mucous and sebaceous salivary gland and lipoid material         |
| Moss-Salentijn and Applebaum(19)/1972 | N/A | N/A   | Posterior mandible | Mucous salivary gland                                          |
| Wilson and MacEntee(19)/1974 | 77        | Male   | Anterior mandible | Papillary cystadenoma and lobule of mucous gland                |
| Izumi et al.(19)/1976 | 11 months | Female | Anterior maxilla  | Cystic glandular epithelium                                    |
| Ide et al.(19)/1983 | 9          | Female | Anterior maxilla  | Mucous salivary gland and excretory ducts                      |
| Moscow and Baden(19)/1986 | N/A | N/A   | Anterior mandible | Mucous salivary gland and gingival cyst                        |
| Brannon et al.(19)/1986 | 21         | Male   | Anterior mandible | Mucous salivary gland                                          |
| Ledesma-Montes et al.(19)/1998 | 43        | Female | Anterior mandible | Mucous salivary gland lobules                                  |
| Gheena et al.(19)/2011 | 45        | Female | Anterior mandible | Mucous salivary gland lobules                                  |
| Gheena et al.(19)/2011 (two cases) | 51        | Male   | Anterior maxilla  | Mucous salivary gland, adipose tissue and focal ductal calcification |

N/A: Not available
millimeters to a few centimeters and the growth is attached to the mucosa by a sessile or pedunculated base, and mild symptomatology can be associated due to constant trauma.\[13\]

According to the data, it is uncertain how minor salivary glands cells have interacted to create mucinous acini in the mandibular alveolar ridge. Researches explain some theories about the mechanism of the development of GSGC. One of these theories describes that the gingival epithelium shows a pluripotential quality, so the unusual location of this minor salivary gland tissue demonstrates that there is an ectopic formation, thus creating a morphogenesis of minor salivary glands.\[8\] Therefore, the fact of finding adipose tissue in the present case reinforces the possible theory of the pluripotential capacity of the gingiva. Another theory debates that in the normal salivary gland tissue development, a fraction of glandular tissue placed in the oral mucosa becomes “trapped” in the gingiva and consequently producing an ectopic growth, leading to the formation of GSGC.\[2\]

In general, it is known that the choristoma is a mature tissue growth with no significant mitotic activity. In the present case, we found positive Ki-67 in some acinar cells. There are a few studies that evaluate index proliferation in adult salivary glandular tissue. In one study, Ki-67 positive cells were expressed in acinar and ductal cells, with a frequency of 8% and 1%, respectively.\[14\] In a more recent work, Aure et al. evaluated the proliferative capacity of the salivary glands in murine mice. They found that homeostasis and maintenance of the adult salivary gland cells are through the duplication of differentiated secretory cells.\[15\]

CONCLUSION

The gingival mucosa is constantly under chronic irritation, chewing forces, trapped food remains, poorly-adjusted restorations, dental calculus, as well as the oral microbiota which under atypical conditions may become pathogenic. That is why in gingiva, there is a tendency for multiple entities to appear which are associated with chronic trauma and external irritation and could be a factor to present symptomatology. It is important that the professional of oral care do not underestimate the innocuous lesions, even polypoid nodules could represent more serious or unusual entities. Whereby a histopathological analysis is mandatory to achieve a correct diagnosis and management of soft-tissue enlargements of oral mucosa.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initial(s) will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.
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Conflicts of interest
There are no conflicts of interest.

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