CASE REPORT

Adenomatoid hyperplasia of the palatal minor salivary glands: A forgotten diagnosis?
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Abstract
It is not uncommon to find lesions on the hard palate in daily practice, but what is uncommon is the encounter of rarities. One such rarity is the adenomatoid hyperplasia (AH) of the minor salivary glands, which is hyperplasia of the normally occurring salivary glands. This rare entity may occur anywhere in the oral cavity; but happens to occur more frequently on the palate. Two cases of AH occurring on the palate in elderly individuals are being reported.

Keywords: Adenomatoid hyperplasia, minor salivary gland, salivary hyperplasia

Introduction
Adenomatoid hyperplasia (AH) is a rare disorder of the minor salivary glands.[1] The earliest description of an entity which could possibly might have been this AH was given by Hendrick and Tyldesley.[2] Later, in 1971 Giansanti et al. studied the lesion histopathologically and identified this entity.[1] In 1981, Arafat et al. coined the term AH.[2] Although it has been more than 4 decades since its recognition, not many clinicians are cognizant about this entity. The true nature of AH is still unknown, and undetermined making it a lesion less explored and hence is the least considered differential diagnosis.[3] Many believe that it's a hamartomatous proliferation of minor salivary glands while others consider it as a hyperplastic lesion.[1,4]

Case Reports
Case 1
A 64-year-old male patient visited with a chief complaint of missing maxillary teeth. History revealed that he had undergone extraction of maxillary teeth about 6 months back following mobility. His medical, surgical, and drug histories were unremarkable. On examination of the oral cavity, the maxillary right and left canines up to molars were missing, bilaterally. Incidentally, the posterior hard palate on the left side showed the presence of a well-defined, round, and nodular swelling which measured about 4 mm × 4 mm in size and had a slight yellowish hue. It was firm in consistency and nontender [Figure 1]. On questioning the patient about him being aware of the lesion, he denied the same. A maxillary occlusal radiograph failed to reveal any pathology of the underlying bone. With a presumptive diagnosis of fibroma, the patient was convinced for an excisional biopsy. The sections of the excised specimen under H and E stain showed the presence of normal salivary gland aggregates with no features of benign or a malignant disorder. The impression was that of hyperplasia of minor salivary glands [Figure 2]. Based on the above findings, a final diagnosis of AH of the palatal minor salivary gland was arrived at. The patient was referred for prosthetic rehabilitation following healing of the biopsy wound. The patient is under follow-up, and there has been no recurrence after 2 years.

Case 2
A 70-year-old male patient visited with a complaint of ill-fitting maxillary denture since about 6 months. The patient had undergone extraction of all his teeth due to mobility and decay and had been wearing the dentures since about 4 years. The patient was medically fit and did not suffer from any systemic illness. On examination, well preserved maxillary and mandibular alveolar ridges were noted. Furthermore, an incidental finding of a well-
defined, sessile, slightly yellowish, elevated nodule was noted on the left side hard palate, close to the midline, measuring 2 mm × 3 mm in size, and being covered with intact mucosa. On palpation, it was nontender and firm [Figure 3]. The patient was unaware of its presence and was totally asymptomatic. A maxillary occlusal radiograph failed to reveal any underlying bony abnormalities. With a clinical impression of fibroma/fibrous hyperplasia, the patient was educated and motivated for a biopsy, and the lesion was excised under local anesthesia. The lesion under H and E stains showed aggregates of normal salivary glands with no evidence of a capsule or malignant changes. The histopathologic impression was that of minor salivary gland hyperplasia. Based on these above features, a final diagnosis of AH of palate was arrived at. The patient received dentures which were fabricated following uneventful healing of the biopsy wound. There has been no recurrence since a year following its excision.

**Discussion**

**Etiology**

The etiology of AH is still unclear. Various factors proposed include idiopathic, local trauma, chronic irritation, denture wear, and smoking.[1-6] Many investigators have also refuted the above factors.[3] In this case, one of the patients had been a denture wearer whereas the other was not. Hence, denture wearing cannot be totally considered as the etiological agent.

**Epidemiology**

The available literature suggests that AH is common in the 4th through 6th decades of life with a wide age range.[1-4] Male preponderance has been noted[3,5] and Caucasians seem to be affected more.[1,2] Asians have a low prevalence according to a few authors[1,3] while others have negated this.[4] Both the patients here were males and in 5th and 6th decades of life.

**Clinical features**

Clinically, AH appears as sessile, nodular lesion, most commonly occurring on the palate - hard and soft, while some uncommon locations include lower labial mucosa, retromolar area, buccal mucosa, and the tongue.[1,3,4] The lesion is mostly asymptomatic but occasionally may be painful.[3] AH may vary in size from a few millimeters to larger lesions up to or over 3 centimeters.[1,2,5] The lesion may be present for a few months or years[5] but most often is routinely discovered by clinicians.[1,4] Rarely, patients having noticed the lesion may also report slow enlargement.[1,4] The mucosa over it will be intact and has been said to be of normal color whilst some authors have reported the mucosa to be slightly erythematous or bluish.[1,4] On palpation, AH will be firm and nontender.[4]

**Differential diagnosis**

Clinical differentials may include the common lesions such as the fibroma, and neurofibroma or the minor salivary gland tumors.[1,2] Those which occur on the lower lip may resemble a mucocele.[4] In addition, hemangioma and lymphangioma also have been considered.[5] Most often, the lesion at first clinical examination has been diagnosed as minor salivary gland tumor.[1-5]
Adenomatoid hyperplasia

Investigations
Radiographic investigations, often done to rule out erosion of underlying bone, have been routinely done but yielded negative results.[1,3,4] Incisional/excisional biopsy, the next in line of investigations is performed for histopathological examination.[1,4] Under the microscope, the H and E sections show normal salivary acinar lobules, with some showing acinar hyperplasia and dilated ducts with mild chronic inflammatory infiltrate.[1,4] These help differentiate AH from other benign or malignant salivary pathologies.[1]

Management
Most often, excisional biopsy is therapeutic, and no special management protocol would be necessary.[1,3] The lesion generally does not recur, and there are no reports of malignant transformation.[1,8]

Conclusion
Although decades have passed since the original description of AH, clinicians seem to have totally ignored this benign hyperplastic salivary lesion and often fail to consider in the differentials. An honest attempt has been made to portray this innocuous lesion through the case report and to recapitulate this almost forgotten entity.

References