## **Case Report**

# **Recurrent Oral Squamous Papilloma in a Pediatric Patient: Case Report and Review of the Literature**

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Squamous papillomas are common lesions of the oral mucosa. They are benign proliferating lesions often painless, slow growing and with a cauliflower appearance. However, its clinical appearance which sometimes mimics exophytic carcinoma, verrucous carcinoma or condyloma acuminatum raises concern when it occurs in the oral cavity. Squamous papilloma occurs predominantly in 30- to 50-year old's. However, they may be seen in children <10 years and accounted for 8% of all oral tumors in children. There is no sex predilection. It has a predilection for the tongue and soft palate, but may occur on any other surface of the oral cavity. Oral squamous papillomas have been associated with infection by the human papilloma virus (HPV). The present report is a case of a recurrent squamous papilloma of the hard palate in a 5-year-old patient with a review of the literature.

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## INTRODUCTION

ral squamous papillomas are common lesions of the oral mucosa of squamous epithelial origin presenting as a papillary or verrucous exophytic mass.<sup>[1]</sup> It is the 4<sup>th</sup> most common benign epithelial lesion associated with human papilloma virus (HPV) types 6 and 11.<sup>[2]</sup> The most common site of occurrence is the tongue and soft palate, however, other sites in the oral cavity may be affected.<sup>[1,2]</sup>

The etiology of oral squamous papilloma is unknown but it has been associated with HPV 6, 11 and trauma. HPV has been found to have the ability to invade the nuclei of cells in the spinous layer resulting in proliferative tissue growth.<sup>[3]</sup> A recent report in the scientific literature, however, suggests that presence of HPV may be merely an incidental finding unrelated to the development of a squamous papilloma.

Mode of transmission for children has been reported as ingestion of viral particles of infected cells from the birth canal, whereas in adults through sexual contact.<sup>[1]</sup>

Oral squamous papilloma may present as a more sinister lesion mimicking lesions like exophytic carcinoma, verrucous carcinoma or condyloma acuminatum.<sup>[4]</sup>

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Diagnostic techniques in the detection of HPV are based on the morphological effects of the virus on tissue, also on the presence of HPV DNA, which can be detected using the polymerase chain reaction (PCR).<sup>[5]</sup>

There is a very limited literature on squamous papilloma of the oral cavity in children from Nigeria and sub-Saharan Africa. We therefore report this case in a child to increase the body of knowledge available on this subject in our environment.

## CASE SUMMARY

A 5-year-old female patient reported to the Pediatric Dentistry Outpatient Clinic of the Lagos University Teaching Hospital in February 2015, with the chief complaint of a growth on the anterior palate of 3 months duration. Growth started as a small nodule that has gradually increased in size to its present size, she also complained of pain while eating. There was no history

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of paresthesia or numbness associated with the growth and no similar lesions were present elsewhere. Family history was not significant. There was no medical or dental history of significance.

Clinical examination revealed a warty pedunculated growth on the anterior hard palate of size around  $10 \times 7$  mm. Swelling was pink in color, firm in consistency and did not interfere with the occlusion. A provisional diagnosis of oral squamous papilloma was made. A complete surgical excision up to 1 mm margin to the depth of the submucosa and a small area of surrounding normal tissue was done and specimen was submitted for histopathologic analysis [Figure 1a and b].

Histopathologic reports showed hyperkeratinized stratified squamous epithelium with thin fibrovascular connective tissue core. The connective tissue contained small endothelial lined vascular channels and a few chronic inflammatory cells consisting mostly of lymphocytes. Koilocytic cells were also seen [Figure 2].

These findings are consistent with the strict histopathologic criteria for the diagnosis of oral squamous papilloma suggested by Carneiro *et al.*<sup>[4]</sup>

In February 2017, patient presented with a recurrence of the lesion on the palate at a site distal to the initial lesions, and was about  $6 \times 5$  mm in size and was excised [Figure 3a and b]. Histopathologic examination revealed hyperplastic parakeratotic stratified squamous



Figure 1: (a and b) Photomicrograph showing the initial lesion before and after excision

epithelium thrown into papillomatous projections. A few virally modified cells suggestive of koilocytes were seen in the epithelium. Within the connective tissue were seen endothelial lined vascular channels and sprinkles of chronic inflammatory cells infiltrate [Figure 4].

### DISCUSSION

Squamous papilloma presents a pedunculated lesion with cauliflower-like surface. They present as single multiple or diffusely involving broad areas of the oral mucosa. In this case it presented as a single cauliflower-like lesion as reported by Abbey *et al.*<sup>[5]</sup>

Oral squamous cell papilloma has been reported to be the most common oral tumor. In 2000, Rafindadi *et al.*<sup>[6]</sup> reported oral squamous cell papilloma as the most commonly seen oral tumors in Nigeria, Al-Khateeb *et al.*<sup>[7]</sup> also reported the same.

Oral squamous cell papilloma is more common in the palate.<sup>[1,8]</sup> This is what we also found in our patient. However, Major *et al.*<sup>[9]</sup> reported the labial mucosa as the most common site, while Flint *et al.*<sup>[10]</sup> reported the tongue. Das and Das<sup>[11]</sup> and Abbey *et al.*<sup>[5]</sup> have reported these lesions in the uvula, tongue, lips and gingiva.

Although the exact etiology of squamous papilloma is unknown,<sup>[8]</sup> it is believed to be related to HPV especially HPV 6 and 11. However, trauma has also been implicated in the etiology of papilloma.<sup>[8]</sup> HPV is said to have the capability to invade the nuclei of the cells in the spinous layer thereby inducing a series of proliferative alterations resulting in growth.<sup>[3]</sup>

Etiology of HPV in children is controversial, however, various mechanisms have been proposed. In children younger than 1 year of age with papilloma lesions, maternal–fetal or maternal–neonatal HPV



**Figure 2:** Photomicrograph showing the initial lesion showing hyperkeratinized stratified squamous epithelium with thin fibrovascular connective tissue core (H and E,  $\times 10$ )

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Figure 3: (a and b) Photomicrograph showing the recurrent lesion before and after excision

transmission has been implicated. It may also result from hematogenous spread from a recent infection or reactivation of a latent infection in the mother.<sup>[3,12,13]</sup>

In older children, transmission may be attributed to either auto (autoinoculation by hand to genital area and genital area to hand or mouth, non-sexual contact or perinatal transmission) or heteroinnoculation.<sup>[13]</sup> However, Yoshpe<sup>[13]</sup> reported the possibility of sexual abuse as a mode of transmission when other routes have been ruled out. Puranen *et al.*<sup>[14]</sup> also proposed horizontal transmission through saliva or other contacts for children whose mothers are HPV negative. However, we could not ascertain the exact mode of transmission in our case, even though we queried sexual abuse in our patient because of the recurrence, there was no evidence to back it up.

Diagnosis involves various techniques such as cytology, biopsy, immunohistochemistry and molecular techniques.<sup>[2,3]</sup> As it can be challenging because of its clinical similarities to other epithelial lesions, like condyloma acuminatum and verruca vulgaris. In our case, the diagnostic method we used was biopsy. Although we also wanted to use the PCR techniques, but lack of adequate facilities prevented us.



Figure 4: Photomicrograph stratified squamous epithelium thrown in to papillomatous projections with a central connective tissue core

Carneiro *et al.*<sup>[4]</sup> in their own study, gave some strict histopathologic criteria for the diagnosis of oral squamous papilloma. The strict histopathologic criteria were as follows: squamous epithelium arrayed in finger-like projections, normal maturation pattern and presence of hyperparakeratosis in the epithelium, koilocytosis as a result of perinuclear cytoplasmic vacuolization of cells of the spinous layer of the epithelium, producing perinuclear pale/clear halos and pyknosis and the occasional presence of basilar hyperplasia. Our histopathologic findings fulfill these criteria. Surgical removal is the treatment of choice by either routine excision or laser ablation. Other treatment modalities include electrocautery, cryosurgery and intralesional injections of interferon.<sup>[13]</sup>

Oral squamous papilloma are common oral lesions, even though they are rarely seen in children. They have a predilection for the tongue and palate and are associated with infection by HPV 6 and 11 variants. We suspected sexual abuse in the present case, but our investigations did not confirm such. Surgery, electrocautery and cryosurgery are treatment options.

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#### Conflicts of interest

There are no conflicts of interest.

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