

Multiple Papillomatosis of Oropharynx in a Young Child-A Rare Presentation

Dr. Neha Salaria, Dr. Ritika Batra, Dr. Uma Garg

BPS Government Medical College for Women, Khanpur Kalan, Sonapat, Haryana

Correspondence Author: Dr. Neha Salaria, BPS Government Medical College for Women, Khanpur Kalan, Sonapat, Haryana, India.**Conflicts of Interest:** Nil**Abstract**

Chronic tonsillar hypertrophy is a common occurrence in children. Chronic tonsillitis presenting as bilateral papillomatous mass in a child is extremely rare. Multiple papillomas of the larynx and tracheobronchial tree are well known (Recurrent Juvenile Laryngeal Papillomatosis), however the occurrence of extensive papillomas of the oral cavity and oropharynx is hardly found in literature. Here, we report a rare case in an 8-year-old male child who presented in outpatient unit of ENT Department with recurrent fever, odynophagia and dysphagia since one year. The lesion was involving both palatine tonsils and part of soft palate on right side and had a cauliflower like appearance. The unusual clinical presentation of multiple papillomas involving soft palate and palatine tonsils and its pathologic significance are discussed in this rare presentation.

Introduction

Most of the neoplasms occurring in the palatine tonsils are malignant, benign tumors or tumor-like lesions are less common. Squamous papillomas account for the majority of these benign tumour like conditions. These are benign lesions of the squamous epithelium resulting in a papillary or verrucous like outgrowth. Lymphoid papillary hyperplasia is also a rare abnormality of the tonsils described mainly in the Japanese population. Other benign causes of enlarged tonsils are lymphangiectic fibrous

polyp, polypoid lymphangioma, angiofibroma, hamartomatous tonsillar polyp or squamous papilloma.

Chronic tonsillar enlargement represents the most frequent finding particularly in children as a result of any infection or inflammation. Under these circumstances, the tonsils became hypertrophied, resulting in a lymphoid proliferation, this being the single most common cause of surgery in children.

Multiple giant papillomatosis as a result of Human papilloma virus (HPV) are common in the larynx and may be recurrent. In the oropharynx, squamous papillomas generally appear as a single wart like growth which may be innocuous and an incidental finding or cause symptoms like odynophagia. However multiple extensive papillomatosis in the oropharynx are seldom reported. [1]

Case Report

An 8 year-old child presented to the ENT department of BPS Government Medical College with complaints of fever, odynophagia and dysphagia since one year. The parents also complained of a growth on the right side of his oral cavity which was initially pea sized but was constantly increasing in size over the last 6 months. He also complained of foreign body sensation in the mouth which increased during episodes of odynophagia. His past medical history, birth history and family history were unremarkable. The patient had bilateral Level II palpable cervical lymphadenopathy. He was mildly anaemic and rest of the biochemical parameters were within normal

limits. There was no evidence of decreased blood counts or impaired immunity.

On clinical examination there was a cauliflower like lesion involving part of soft palate and entire palatine tonsil on the right side whereas on the left side it was involving lower pole of palatine tonsil sparing the soft palate. On right side the mass was about 4 X 3cms in size while on left it was about 3 X 2 cms in size.(Figure 1,2) On both sides the lesion was pink in colour, soft in consistency & did not bleed on manipulation. Keeping in view the age of the patient the clinical possibility of a benign neoplastic lesion such mucosal papilloma and papillary lymphoid hyperplasia was made. Investigations including X-ray chest were normal & there was no evidence of immune suppression. Tonsillectomy was planned along with LASER excision of papillomatous mass from soft palate. After building up the patient and preanaesthetic check up the child was taken up for surgery under general anaesthesia. LASER assisted tonsillectomy was done along with complete excision of the mass from the soft palate. Hemorrhage was comparatively more yet could be controlled with conventional techniques.(Figure 3,4) The specimens were fixed in formalin was sent for histopathologic examination. Grossly the specimen showed exophytic growth. Microscopic examination revealed multiple papillae lined by hyperplastic squamous epithelium along with parakeratosis and focal koilocytic changes. Papillae were supported by delicate fibrovascular striae. Henceforth histological features were consistent with squamous papilloma with focal koilocytic changes. The patient has been monitored for more than one year with no evidence of recurrence.

Discussion

Papillomas are a group of localized polypoid masses as a result of hyperplastic exophytic lesions with a distinct verrucous or cauliflower like morphology. Certain studies

suggest a female gender predilection while others show the contrary.[2,3] In a study on 464 patients with oral squamous papilloma(OSP) it was found that papillomas were most abundant on the palatal complex, dorsum and lateral tongue borders, and lower lips, respectively.[3] Another study by Carneiro et al suggested that the most prevalent site was the tongue (41.7%), followed by the palate (33.3%), lip (16.7%) and labial commissure (8.3%). [2] None of these studies reported a papilloma involving the tonsillar fossa as in the case reported. This is probably the first case reporting a papilloma involving not only the soft palate but also both right anterior tonsillar pillar and bilateral tonsillar fossae till date.

Recurrent respiratory papillomatosis(RRP) is a disease of childhood in which papilloma's (wart like tissue) grows over any part of the respiratory tract. The most common site is the larynx including the vocal cords. Laryngeal papillomatosis is also nevertheless a rare condition seen in 1 and 4 per 100 000.[4] About 60-80% of the cases are seen in children under the age of three years in whom it has a propensity to have a more nefarious course. These grow not only to cause respiratory obstruction but are also recurrent in nature.

Papillomas are considered to be caused by a strain of HPV, more than 150 of which have been isolated till date. Human Papilloma Viruses are DNA viruses that infect the stratified epithelium and induce proliferative changes in it. Replication cycle of the HPV is completed in the outer epithelium and virions are shed alongwith shedding of these cells, hence exposure of immune cells to the viral antigens is limited which is thought to result in a more persistent infection. [5] Various strains are classified into high risk and low risk depending on whether they induce a benign or malignant growth in the normal mucosa. HPV 2 and 4 are known to cause skin warts while condylomas (genital warts), and respiratory papillomatosis are caused

by HPV 6 and 11 which are of low oncogenic potential. All these lesions are benign and can be treated by LASER ablation or surgical excision. In a study by Major et al 100% positivity for HPV 6 or 11 by PCR was found.[6] It is also postulated that in children usually there is coinfection by HPV 6 and 11 whereas in adults, respiratory papillomatosis is caused by a single variety.[7] HPV 16 and 18 are considered to be the more malicious strain causing cervical cancer in females in 95-98% of cases. [3]

The role of HPV in Head and neck SCC is not well understood, However studies indicate that HPV is 2 to 3 times more likely to be detected in precancerous lesions like leukoplakia, intraepithelial neoplasia and 4.7 times more likely in oral SCC than in normal healthy oral cavity mucosa.[1]. Studies also suggest that latent HPV is common in oral mucosa and HPV DNA is seen in as many as 80% individuals. [9]

The squamous papillomas represent 75% of the benign tonsillar masses as per literature. They are usually small innocuous papillary mucosal lesions occurring mainly in the palatal complex but seldom on the uvula and tonsil. They are usually discovered incidentally or they may cause unpleasant sensation in the throat. Most of the papillomas are asymptomatic, however it is thought that greater the length of the papilloma, more are the chances of the mass being symptomatic. The patient in the case reported had symptoms of odynophagia, dysphagia and foreign body sensation, this could be accounted for by the large size and multiple sites of the papillomatous mass. It is also reported that less than 25% of OSP's are more than 1 cm in length.[10,11] Moreover in the case presented, the papillomas were on the palatine tonsils which caused symptoms consistent with chronic tonsillitis. Extensive multiple papillomas as seen in the case presented raises suspicion of immunosuppression which must be ruled out.

Oral squamous papillomas have to be differentiated clinically from pyogenic granuloma, fibroma, verruca vulgaris, fibrous hyperplasia and verrucous carcinoma. [12]

Histologically tonsillar squamous papillomas are composed of well differentiated benign appearing squamous epithelial papillae on thin fibrovascular stalks. It has to be histologically differentiated from fibrous epulis, fibroepithelial polyps and papillary hyperplasia associated with candida. These lesions however have a prominent fibrous component and no viral change. [5] Viral presence is indicated by observation of koilocyte like cells in the spinous layer of the epithelium. The colour of these papilliform lesions may vary from pinkish to whitish which depends upon the thickness of the cornified layer. [2]

The treatment of choice for oral squamous papillomas is surgical excision which may be done with cold dissection, electrocautery, or Laser ablation. The prognosis is good as they do not have a propensity to recur unlike laryngeal papillomas.[10] Intra or Perilesional interferon has also been tried in HPV induced lesions in sites like eyelid where complete excision may not be achieved, however excision of the lesion is the definitive treatment of choice whenever possible.[13,14] Recently it has also been advocated that the quadrivalent HPV vaccines generally used in female population for prevention of Cervical cancer may also be of great significance in preventing oropharynx cancers and other lesions associated with HPV, however clinical trials are currently underway to prove this proposition.[15]

Conclusion

Oral squamous papilloma is a benign tumour which is generally innocuous. The occurrence of squamous papilloma on the tonsil is extremely rare.[11] In the case reported, not only was the tonsil involving the soft palate

but also the anterior tonsillar pillar on the right side alongwith involvement of both palatine tonsils. Due to such an extensive mass the patient had symptoms suggestive of chronic tonsillitis.No oral papilloma with such unusual picture and such extensive dimensions could be found on medline search. Hence such a presentation makes it an unusual entity.

References

[1]. Brodsky L, Siddiqui SY, Stanievich JF. Massive Oropharyngeal Papillomatosis Causing Obstructive Sleep Apnea in a Child. Arch Otolaryngol Head Neck Surg. 1987; 113(8):882–884. doi:10.1001/archotol.1987.01860080088024.

[2]. Carneiro TE, Marinho SA, Verli FD, Mesquita TM, Lima NL, Miranda JL. Oral squamous papilloma: clinical, histologic and immunohistochemical analyses. Journal of Oral Science. 2009; 51(3): 377-372.

[3]. Abbey LM, Page DG, Sawyer DR. The clinical and histopathologic features of a series of 464 oral squamous cell papillomas. Oral Surg Oral Med Oral Pathol. 1980 May;49(5):419-28.

[4]. Miller CS, Johnstone BM. HPV as a risk factor for oral squamous cell carcinoma: a metaanalysis, 1982-1997. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2001 Jun; 97(6): 622-35.

[5]. Larson DA, Derkay CS. Epidemiology of recurrent respiratory papillomatosis. APMIS. 2010 Jun;118(6-7):450-4. doi: 10.1111/j.1600-0463.2010.02619.x

[6]. Chong KT, Xiang L, Wang X, Jun EL, Xi L, Schweinfurth JM. High level expression of human epithelial beta defensins(hBD-1,2 and 3) in papillomavirus induced lesions. Virol J;2006(3)-75.

[7]. Major T, Szarka K, Sziklai I, Gergely L, Czegledy J. The characteristics of human papillomavirus DNA in head and neck cancers and papillomas. J Clin Pathol. 2005;80:63-66.

[8]. Eversole LR. Papillary lesions of the oral cavity: relationship to HPV. J Calif Dent Assoc.2000 Dec; 28(12):922-7.

[9]. Jaju PP, Suvarna PV, Desai RS. Squamous papilloma: Case report and review of literature. Int J Oral Sci 2010; 2:222-5.

[10]. Goodstein LA, Khan A, Pinczewski J, Young VN. Symptomatic Squamous Papilloma of the Uvula: Report of a Case and Review of the Literature. Case Reports in Otolaryngology. Volume 2012, Article I D 329289, 2 pages.doi:10.1155/2012/329289

[11]. Mundra RK, Verma J, Gupta A, Koshta V, Rathore SK. Tonsillar papilloma: A rare case. International Journal of Medical Science Research and Practice.2015; 2(3):162-163.

[12]. Wadhwa R, Kalra V, Gulati SP, Ghai A. A big solitary oropharyngeal papilloma in a child. Egypt J Ear Nose Throat Allied Sci 2012;13:131-2.

[13]. Lee BJ, Nelson CC. Intralesional interferon for extensive squamous papilloma of the eyelid margin. Ophthal Plast Reconstr Surg. 2012 Mar-Apr;28(2):e47-8.

[14]. Stellato G. Intralesional recombinant alpha 2B interferon in the treatment of human papillomavirus-associated cervical intraepithelial neoplasia. Sex Transm Dis. 1992 May-Jun;19(3):124-6.

[15]. Gillison ML. Human papillomavirus-related Diseases: Oropharynx Cancers and Potential Implications for Adolescent HPV Vaccination. J Adolesc Health. 2008 Oct; 43(4 Suppl): S52–S60. doi: 10.1016/j.jadohealth.2008.07.002.

Clinical Photographs



Figure 1. Papillomas seen on the soft palate (right side)



Figure 2. Papillomas involving soft palate, anterior pillar(right) and left tonsil. Right tonsil is obscured by the right sided mass.

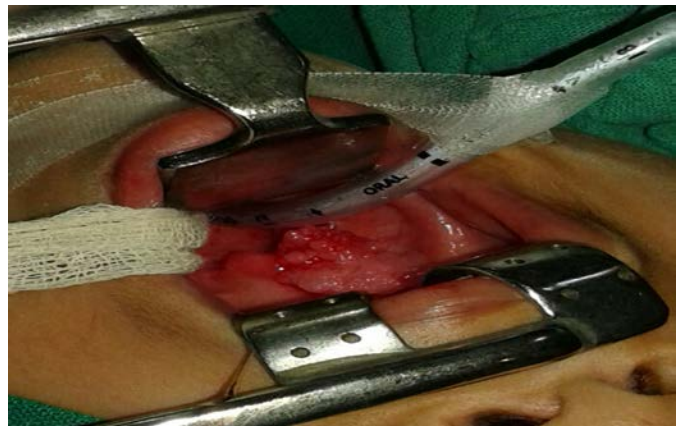


Figure 3. Intraoperative view revealing right tonsil papillomas alongwith soft palate and pillar involvement.



Figure 4. Showing healthy slough and postoperative site