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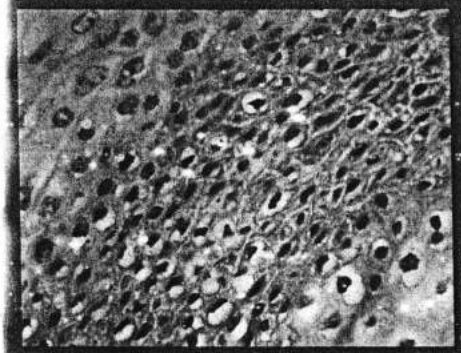

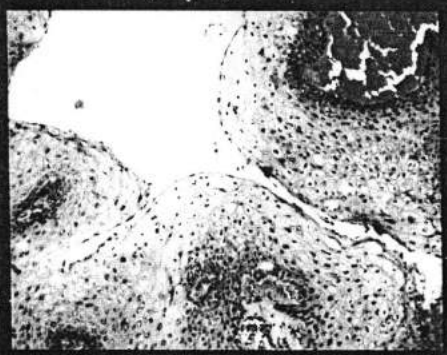

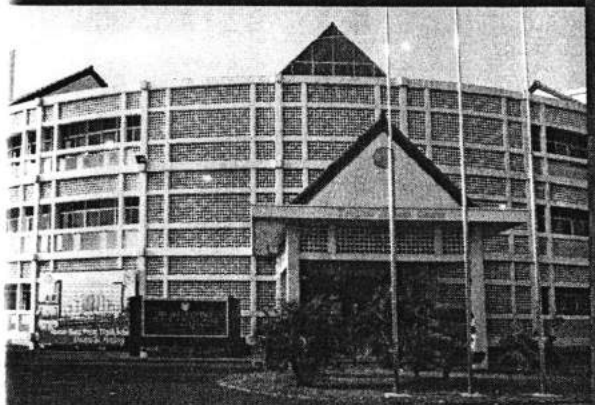

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CONTENTS

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Case Report

RECCURENT LARYNGEAL PAPILLOMA

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ABSTRACT

A case of respiratory papillomatosis was reported. The patient suffered from the disease since eight months old with chief complaint progressive hoarseness and dyspnea. It was diagnosed with respiratory papillomatosis and scheduled for performing tracheotomy and continued with the first microlaryngeal surgery (MLS). Decanulation was taken after 2nd surgery of removing papillomas. Finally was reported she got serial of surgery for 22 times during 18 years of age. It was costly and deteriorating quality of life. The problem remains persisted because of frequent recurrences and need for repetitive surgeries. Specimen biopsy for histologic examination was shown the signs of HPV infection, papilomatic coated squamous epithel with mild dysplasia and coilocytosis. The threatening of upper airway obstruction is the main important reason for patient's coming. The patency of airway assessed by Direct Laryngoscopy then the next treatment was decided with schedule of Micro Laryngeal Surgery (MLS). Finally the MLS treatment is just only for temporarily recovery. A further research to define the proper treatment in the future is required, especially for prevention of the diseases related to the viral causes of infection.

Key words: *recurrent, respiratory papilloma, HPV, microlaryngeal surgery*

INTRODUCTION

Respiratory papilloma has been known since the 17th century ago. This disease was first discovered by Marcellus donalus as "*warts in the throat*" that grows on the throat area. Papillomas may grow on the mucosa throughout the respiratory tract. Vocal cords are a common predilection obtained. The growth of tumors usually occurs in multiple and tends to grow recurrent.[1,2]

Papilloma of the nose is rarely obtained. Moreover, it can also be found type of sinonasal papilloma (inverted papilloma) and carcinoma can mimic the form of papilloma in the nose area.[3]

Patients were commonly found in difficulty to breath, dyspnea state, because of airway obstruction and indicated for performing tracheotomy. The disease leads to the expansion of papilloma growth and tends to increase morbidity. Considering from this point, early diagnosis is very important and need some efforts to avoid tracheotomy in patients.[4]

Since now Papilloma is remain a problem because of its frequent relapses and potential to threat airway obstruction that endangers the lives of patients. This problem more over complicated, cause there is no right treatment to overcome this problem so far. Even though various theories have been published but the results are not satisfactory yet.[5]

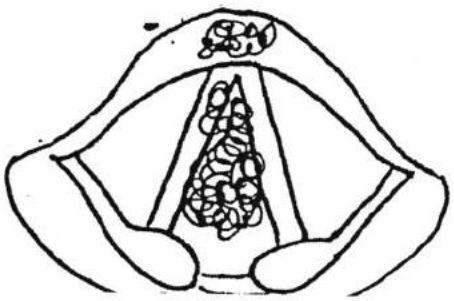
The purpose of this paper is to report a case of laryngeal papilloma in our Departmen, Departement of Otorhinolaryngology Head and Neck, Airlangga University, Dr. Soetomo General hospital.

CASE REPORT

March 17, 1994, in ENT outpatient, a young woman (RAF) 8 months old, was referred from a doctor, ORL-HNS specialist at the Dr. Soedono Hospital, Madiun city. She complaint with hoarsness since 3 months before. She looked cahectic.

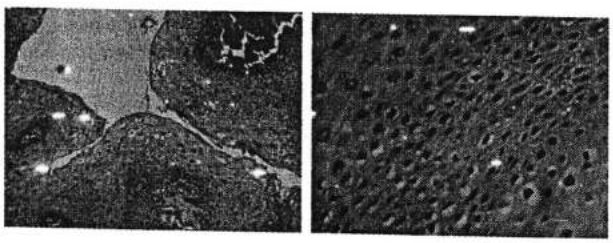
Examination on the ear and nose and thorat, showed no abnormalities. On direct laryngoscopy examination with the following results; Anamnesis hoarseness, sometimes dyspnea, coughing was not found and ate and drank well. Physical examination found mild stridor and intercostals

retractions. Direct laryoscope showed bump of mass which colour white pellucid, uneven, lookslike papilloma pharynx and larynx, in the glottis and supraglottis. It planned for tracheotomy and extirpation with Micro Laryngeal Surgery (MLS).



Picture 1. A scheme of direct laryngoscopy showed mass in the larynx, glottis and supraglottis

One day later the patient was performed tracheotomy and followed with MLS one month later with the following result, was seen mass bumps, which color is translucent white alike papilloma, located in the pharynx, posterior middle aritenoid, cricopharynx at 3, 6 and 9 hours. Then the tumor is extracted until it was clean and performed histopathologic examination.



Picture 2. Specimen from papilloma in the vocal cord biopsy when performing MLS, showing tissue sections shaped papilomatic coated with squamous epithel with mild dysplasia and coilocytosis, epithel in the surface and shown stroma with fibrous.

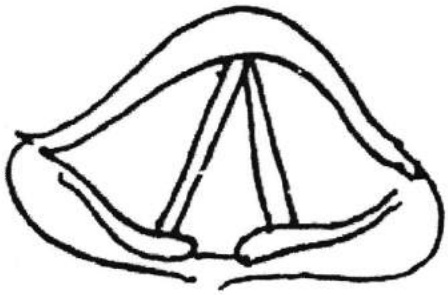
Patient came at May 11, 1994 (13 days later), without any complaints. On examination found trakeocanul installed and functioning properly. The pathologic anatomy result (No. L. 1598/94) with the conclusion: papilloma with coilocytosis (signs of HPV infection).

On September, 1994, four months later papillomas were still in the pharynx and larynx. Second MLS was planned next one month.

Tha patient returned 1 month after the MLS, there was no complaint and the examination didn't find growth of papilloma again. Decanulation was planned.

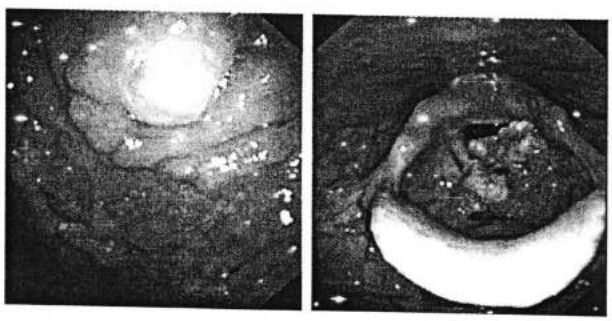
On December, 1994 (one month later), still found little papillomas in the oropharynx and one month later the situation remain similar. Decanulation was performed.

On February 15, 1995, papilloma became the less prominent. Likewise the following months, the situation remains the same until the month of November 1995 (20 months since the patient first came).



Picture 3. Endoscopic examination (illustration) on larynx, minimal growth of papilloma, and airway is wide enough

Recently status coming, the patient was 18 years old. She had been performed MLS for 22 times surgeries. We recorded the endoscopic examination at August, 2011. The picture wass shown below and after that examination, she performed the 22nd MLS for removed the papillomas.



Picture 4. Endoscopic examination on larynx. Left picture: papillomas growth on the pharynx, right: papillomas in the glottis.

Picture 5. Endoscopic examination after performed the 22nd, showed minimal papillomas in the larynx.

DISCUSSION

It was found a patient with papillomas which age were 8 months old when in the first arrived. Most patients with papilloma have age under 5 years.[6]

In adults, men are tends common occured, but the incidence in children is almost the same.[8] In this case, the patient is a women.

Tumors can grow along the respiratory tract and mouth (aero-digestive tract) and predilection the most common is in the larynx (97.9%–100%).[3,4] The growth of papilloma of the nose, are often in the histopathologic form of inverted papilloma (47%) and fungiformis papilloma (50%) than the cylindrical papilloma (3%).[15]

One of the factors causing papillomas is due to a viral infection. Any signs of HPV infection are found in both patients in the form of coilositosis cells, so that convince suspicion the virus as the etiological factor of disease.[1,9] This can cause by transmission from mother during delivery (60%).[4] But, the gynecological examination form the mother of the patient didn't found signs of condyloma. This possibility can occur because the patient's mother may have recovered from her illness at the time when examination performed (some time later after giving birth).

At first, papilloma is often confused with suspicion of allergic disease, asthma or croup.[5] Similar with the 2nd case, the complaint of runny nose and frequent epistaxis has suffered since 2 years before. Papilloma was diagnosed after one year later after the appearance growing mass in the left nose. Three months later, there were complaints of sound breathing and short of breath. Patients referred with the airway inflammation. But, the thoracic X-ray showed no abnormalities. Finally, the direct laryngoscope showed multiple masses in the pharynx and larynx, suggest papillomas.

Papilloma can show remission with increasing age.[6] In this case, a minimal tumor growth after 20 months later and the MLS has done frequently. Following the Papilloma growth getting fewer and steady, therefore, the tracheo-canule could be pulled out. Based on studies about papilloma that grows outside the larynx, it gives a better response to treatment (MLS).[5]

Serial of Microlaryngeal surgery (MLS) were performed repeatedly to they that need to excise the tumor, because of that, airway is free and sounds normal again. Decreasing of papilloma is expected to facilitate the body's defense system to eradicate the residual lesion, and then would accelerate healing. As is well known, larger size of the tumor, there is a lot of virus and more difficult to control.[17]

All patients performed emergency tracheotomy at the first time came at the emergency room (second case) and tracheotomy preparation for MLS a day after the examination of direct laryngoscope (first case). Actually, tracheotomy could be avoided if the patient came and diagnosed earlier. This procedure will cause a wound that may facilitate the implantation of new lesions in lower respiratory tract. Expansion to the tracheobronchial founded approximately 83% after tracheotomy.[2] This is a concern, especially in the second case where there is growth of laryngeal papilloma, with using tracheocanule can cause new lesions caused by friction of the canule.

Therefore, it's needed to evaluate the subglottic and trachea due to the expansion of laryngeal papillomas. The first case, where the larynx is clear from papilloma, there is no papillomas growth in tracheobronchial region although it has been performed tracheotomy. This is corresponding with the state that the papillomas growth in the trachea is always preceded by a laryngeal papilloma after tracheotomy.[8] We only have two papilloma patients without tracheotomy in our hospital. Examination on 11 and 13 months after first MLS didn't found any papillomas extension to the tracheobronchial. In 14 patients who performed tracheotomy, several of them were found down expansion to the tracheobronchial after 2nd or 3rd MLS (approximately 6–12 months). After that, interval time between MLS more short (1–2 months), even in one case the papillomas expansion has reached the left bronchus after the 23rd MLS (34 months later).

Tracheotomy is necessary when there is upper airway obstruction with grade III Jackson or show signs of respiratory failure. Meanwhile, when in grade I-II, could performed MLS with insufflations anesthesia techniques. However, this technique has never been applied so far, so tracheotomy performed for procedures such as in the case of the first MLS.

Decanulation done as early as possible when conditions are stable and papilloma growth stopped for at least 6 months. Likewise in the first case, growing of the papilloma was slight then pulled out the canule performed 10 months later and next 10 months showed minimal lesion.[5]

In addition, there are also two patients who have been decanulation after 6th and 10th MLS (2 yr and 3yr 5mo). Until tracheal, papilloma growth has stopped.

The existence of a large papilloma growth (diffuse, multiple) possibly because patient with low immune state (since the age of 8 months has reccurence of cough) thereby increasing aggressiveness of the disease. One factor in accelerating the remission of disease is to increase the immunity of patients, namely how to immunotherapy such as vaccination and administration of interferon.[10,16] This treatment is not yet a standard treatment at our institution.

Inverted papilloma of the nose, which its epithelial growth folding in to the stroma. HPV virus is a one of suspected etiology factor, these tumors are potentially associated with multiple papillomas along the respiratory tract and mouth. This is consistent with the results of studies using PCR techniques (Polymerase Chain Reaction) which have found HPV virus types 6, 11, 16 and 18 in the genital tract and respiratory tract. In the genital tract HPV types 6 and 11 found in the exophytic condyloma, but types 16 and 18 are found on flat condyloma with a high degree of dysplasia and invasive carcinoma. Similarly in the respiratory tract, HPV types 6 and 11 associated squamous papilloma and inverted papilloma, while HPV types 16 and 18 are found in squamous carcinomas.[18]

An important thing to differentiated from squamous papilloma is the nature of the invasive and the tendency to malignancy in inverted papilloma. Therefore, patients with inverted papilloma need to be having long-term follow-up of recurrence and risk factors of transformation towards malignancy. Interval changes of malignancy ranging from

5 to 20 years, with the incidence of 1.5 to 2%.[19] There was a report the occurrence of malignancies at the age of 20 years from one patient papillomas since childhood and has performed tracheotomy, ie bronchogenik carcinoma. Eventually the patient died after occured metastasis. Some experts associate inadequate incision and exposure to carcinogens such as radiation with materials, cigarette smoke with risk factor of reccurence and malignancy. Histologic examination found a representation of atypical epithelium and dysplasia.[17]

Aggressiveness of papilloma growth may be explained by histopathology examination, among others associated with the type of papilloma, the degree of cell atipia, mitotic index, the ratio of neoplastic epithelium with the stroma, and the presence of inflammatory cells.[13,15] It required a clear description of histopathology analysis results by including the factors mentioned above. Likewise, signs of viral infection should be included, for example coilocytosis, nuclear inclusion bodies or multinucleated epithelial cells.[15] Where possible to do on a regularly, such information will be able to add the epidemiological data that may be useful in overcoming this disease.

CONCLUSION

It has been reported a case of recurrent Laryngeal papilloma, threatened the airway and lead to obstruction in the larynx.

Tracheotomy should be avoided if patients can come earlier and early diagnosis is established.

The problem was still persisted with the high recurrence in children and treated temporary by Micro Laryngeal Surgery.

Inverted papillomas might have a greater risk for the occurrence of malignant transformation, then long-term follow-up is required.

Following study is necessary to explore further about pathogenesis of suspected viral infection in pregnant patients as resources to find a strategy in the epidemiological approach to disease prevention.

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