World Journal of Surgery and Surgical Research

Case Report
Published: 02 Apr, 2024

Is There a Place for Endobronchial Stent Treatment in Laryngeal Papillomatosis? Case Report and Literature Review

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Abstract

Recurrent respiratory papillomatosis is a challenging chronic airway disease that occurs in both children and adults. It is caused by the human papillomavirus, more than 90% caused by HPV types 6 and HPV 11. The clinical presentation is nonspecific and most patients present with hoarseness, stridor or dyspnea. Although there is no definitive treatment for RRP, the objective of treatment focuses on improving voice quality and maintaining airway patency, in our case it was achieved through the placement of a Y-shaped stent placed in airway added to adjuvant treatment with Bevacizumab. Surgical management is the mainstay of treatment and around 20% of patients benefit from adjuvant therapies.

Keywords: Papillomatosis; Airway; Y-stent

Introduction

Recurrent Respiratory Papillomatosis (RRP) is a disease of recurrent warty lesions of the upper aerodigestive tract. Papillomas are the most common benign neoplasm of the larynx in children [1]. It is considered a manageable rather than curable disease due to its high recurrence rates and unpredictable clinical course [2]. Current management strategies primarily focus on improving voice quality and maintaining airway patency.

There is a bimodal age distribution, with the juvenile form occurring most commonly in children under 5 years of age and the adult-onset form occurring between 20 and 40 years of age. The juvenile form is considered highly aggressive with a high recurrence rate compared to the adult form. In the adult form, it predominates in men, while there is no gender difference in the juvenile form. The triad of first-born, vaginal birth and child of a young mother is found in 75% of children who present PRR.

Recurrent respiratory papillomatosis is caused by the Human Papillomavirus (HPV). This DNA virus is well known and can cause both papillomas and condylomas and cancer of the cervix, anus, mouth or throat. There are hundreds of strains of HPV. The most common strains found in PRR are HPV 6 and 11.

HPV 11 has been found to be more aggressive, requires more frequent surgical debridement's, and is more likely to have extralaryngeal involvement with a higher risk of requiring tracheostomy

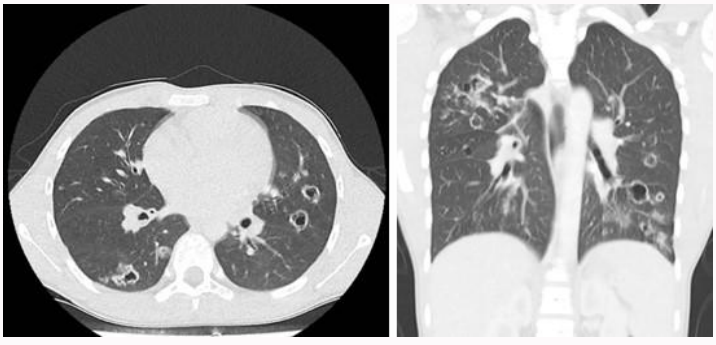

Figure 1: Chest tomography with evidence of multiple cavitated images in both lung fields.

OPEN ACCESS

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Received Date: *07 Mar 2024*
Accepted Date: *27 Mar 2024*
Published Date: *02 Apr 2024*

Citation:
Zinni MA, España MI, Carrabs A, Bustos ME. Is There a Place for Endobronchial Stent Treatment in Laryngeal Papillomatosis? Case Report and Literature Review. World J Surg Surgical Res. 2024; 7: 1529.

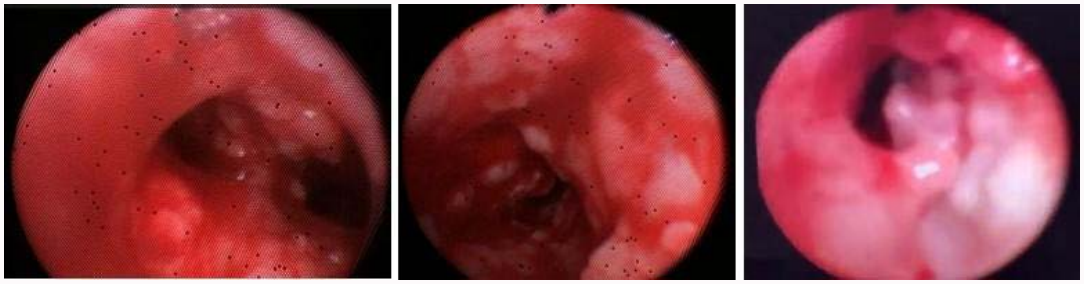

Figure 2: Repeated fibrobronchoscopies with evidence of papillomatosis in the airway.

and treatment with adjuvant therapy.

In most children, HPV infection occurs during childbirth, since the mother's genital tract acts as a reservoir for HPV; cesarean delivery can prevent HPV transmission in mothers with active condylomas, however, exists 12% of PRR cases transmitted through the placenta [1].

RRP is usually located in the larynx and subglottic region, while involvement of the trachea has been documented in 5% of cases and the lower respiratory tract is involved in less than 1% of cases, only 1.8% of patients present lung lesions [3].

The first warning symptom is always dysphonia. Occasionally, dyspnea appears, which is usually progressive, but can decompensate rapidly during the course of an upper airway infection [4].

The vocal cords are the main site of papilloma growth, leading to hoarseness being the main symptom. Inspiratory stridor, which progresses to biphasic stridor, is the second presenting symptom. Other symptoms are: Dysphagia, dyspnea, chronic cough, growth retardation and respiratory tract infections. Aggressive forms of RRP can manifest as acute respiratory distress [2].

Although infection occurs at birth or before, most patients do not present symptoms immediately [4].

Patients with previous invasive procedures, tracheostomy, HPV-11 infection, and age <3 years are a high-risk group for developing extralaryngeal papilloma.

The natural evolution can be unpredictable, it can remit spontaneously, remain stable or show aggressive growth [4].

Although RRP is clinically a benign disease, transformation to dysplasia and invasive carcinoma can occur. The age of onset of the disease is a significant risk factor for dysplasia or squamous cell carcinoma [1].

Other risk factors for malignant transformation include persistent infection with HPV types 16 and 18.

The diagnosis of RRP consists of visualization of the papilloma with a rigid or flexible laryngoscope with biopsy for confirmation [2]. In histology, it appears as exophytic, branched and sessile pedunculated masses of reddish or pink color with a highly vascularized lobed surface, with white-pink, warty characteristics (similar to "raspberries" or "bunches of grapes"). Single or multiple form and its growth can cause airway obstruction [5].

Helical CT can help show some characteristic features of RRP lung lesions, revealing single or multiple, multilobular, well-defined, solid nodular or polypoid lesions, of various sizes, with a centrilobular distribution, scattered throughout the lungs with a tendency to the confluence [1].

Figure 3: Fibrobronchoscopy that shows correct Y-Stent placement.

Figure 4: Control fiber bronchoscopy after treatment with Bevacizumab.

Considering the challenging treatment of RRP, prevention should be the primary focus of physicians [2].

The current standard treatment for RRP is surgical excision of symptomatic lesions. The goal of surgery is to reduce and remove the papilloma with preservation of the surrounding normal structure.

Laser surgery is one of the newest modalities to treat papilloma has hemostatic properties and superior visualization compared to cold instruments or the use of microdebrider.

Tracheostomy is avoided in patients with RRP, if possible, as it creates an iatrogenic squamocolumnar junction that may be a source for papilloma development. If a tracheostomy is required for patient safety, early decannulation is recommended.

Although surgical intervention is the main treatment for RRP, about 20% of patients with RRP require adjuvant therapy [1]. Adjuvant therapies are indicated when more than four interventions

per year are required, there is extralaryngeal involvement and/or rapid growth with respiratory obstruction and in children under 2 years of age [4].

Multiple adjuvant therapies have been used including cidofovir, interferon, bevacizumab, and PD-1 inhibitor.

Bevacizumab (Avastin) is one of the newest adjuvant therapies to control PRR. It is a recombinant monoclonal antibody directed at vascular endothelial growth factor (anti-VEGF) that leads to reduced angiogenesis [1].

Case Presentation

We present the case of a 17-year-old male patient diagnosed with laryngeal papillomatosis at 7 months of age (history of laryngeal dysphonia and stridor), requiring tracheostomy at 2 years of age. Mother with no history of genital papilloma, born by cesarean section. He underwent treatment with Interferon, Cidofovir and Bevacizumab. Patient experiences multiple episodes of respiratory superinfection and progressive dyspnea. Because it was treated in a pediatric institution when he turns 14, he is referred to our institution.

A chest tomography was performed which showed involvement of the lung parenchyma with 5 mm nodular images at the level of the right upper lobe and bilateral basal level (Figure 1).

We continued with performing a fiberoptic bronchoscopy where tracheobronchial papillomatosis with subtotal occlusion of the lumen was observed, and also complete obstruction of the larynx and vocal cords without allowing the passage of the endoscope. On several occasions, partial resection of tracheal and bronchial lesions with recurrences was performed, so it was decided to place a Y-stent to ensure an airway. Because larynx permeabilization was not achieved, it was decided to extract the end through the tracheostomy orifice, thus allowing oxygenation through the end externalized by tracheostomy, and securing the end of the stent with a cannula device. After the procedure, the patient showed immediate improvement in respiratory symptoms (Figure 2, 3).

It is discussed in committee and it is decided to start new therapy with Bevacizumab. Subsequent controls are performed, improvement of papillomatosis and laryngeal permeabilization is observed (Figure 4).

Discussion

Recurrent respiratory papillomatosis is the most common tumor of the larynx and a common cause of pediatric hoarseness. The main problem with PRR is its tendency to repeat itself. RRP recurs due to the histological characteristics of the papilloma and its clinical course depending on the virus subtypes. Furthermore, the high recurrence rate may be due to the difficulty of completely removing the papilloma [2].

Children whose RRP is diagnosed at a younger age (less than three years) are 3.6 times more likely to have more than four surgeries per year and almost twice as likely to have multiple anatomical sites affected.

The most common symptoms are hoarseness, chronic cough, dyspnea, recurrent upper respiratory infections, pneumonia, dysphagia, stridor or growth retardation, which tend to be more serious in children due to the rapid growth of the lesions [3].

It is essential to recognize voice disorders and evaluate the airway in any infant or child with persistent dysphonia, in order to establish an accurate diagnosis and appropriate treatment. Delay in diagnosis can have negative clinical consequences due to disease progression [4].

RRP is considered a manageable rather than a curable pathology. The papilloma can grow so extensively that the airways can become blocked and, if left untreated, can lead to death. Surgery remains the fundamental treatment practice.

Airway stents, also known as tracheobronchial prostheses, are tube-shaped devices with a hollow lumen that are inserted into the airways [6], respiratory tract, can be used to treat a variety of large airway diseases, it is a palliative therapy or a bridge to curative therapy for patients with various types of airway diseases. Silicone stents are one of the most common stent types used in clinical practice as they are firm and durable, have a lower frequency of granulation tissue, and are easier to reposition and remove using rigid grasping forceps. There are several types of silicone stents, including Y-shaped, they are mainly used for diseases of the main carina, although they can also be used for injuries that affect the secondary carina. Many treatments may be required as the papilloma recurs. Considering the challenging treatment of PRR, prevention should be the main focus. Treatment should be individualized and based on shared decision making [2].

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