



Laryngeal Papillomatosis: A Case Report

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Abstract

Laryngeal papillomatosis (LP) is a relatively rare benign tumoral proliferation of the larynx, usually seen in children. It is due to infection with Human Papilloma Virus (HPV), and the first symptom is chronic dysphonia. There is currently no consensus on the treatment regimen for LP. Several treatments are used, but their efficacy is difficult to determine due to small patient cohorts. In a developing country, the treatment is based on peeling by laryngeal microsurgery. Its capricious evolution requires regular monitoring of patients. The possible prevention through the use of prophylactic vaccines is the most significant recent development. We report the case of a child who was dysphonic and had laryngeal dyspnea, in whom the peeling of a papilloma with forceps under direct laryngoscopy was performed, the histological study affirmed the diagnosis and made it possible to eliminate a neoplasm.

Keywords: *Papillomatosis, Larynx, Treatment*

Introduction

Laryngeal papillomatosis results in ripening lesions on the vocal cords (**Figure 1**) and is generally observed in children. It results from infection with a human papillomavirus (HPV), mainly subgroup 6 or 11 ^[1]. The latter are less oncogenic than the 16/18 and 31/34 subgroups. However, HPV 11 is more aggressive and would be more carcinogenic than HPV 6 ^[1]. The diagnosis of papillomatosis is suspected in children presenting with dysphonia for more than three weeks. It is strongly suggested by the endoscopic appearance and must be confirmed by a histological study ^[2]. The risk of laryngeal papillomavirus infection is airway obstruction and dissemination to the respiratory tract. In addition, there is a risk of cancerization in the event of laryngeal involvement ^[3]. There is no curative treatment. The treatment involves iterative endoscopies under general anesthesia to remove the papillomas visible with forceps, CO₂ laser or microdebrider. The sequelae are of the dysphonia type due to the scars on the vocal cords.

Case report

Our patient is a 5-year-old child, first-born to a 30 years old mother, after a pregnancy followed to term; the delivery was vaginal, with good adaptation to extra-uterine life. The child is well vaccinated according to the national immunization program. The beginning of the symptomatology goes back to the age of 2 years, by the modification of the voice and hoarse cries, but the family did not

come to consult for this disorder because it seemed minor. It was only at the age of 4 that the parents took him to consult for dysphonia which was progressive. Speech therapy sessions were indicated for the patient, but without improvement. Faced with the worsening of the symptoms and with the onset of laryngeal dyspnea, the patient was referred to us for care.

On admission, the child was conscious, afebrile, presented with aphonia, laryngeal stridor at rest, dyspnea such as inspiratory bradypnea with drawing and hominess, signs of intense respiratory struggles: suprasternal, supraclavicular and subcostal. Room air saturation was 93%, heart rate 107 beats per min. The patient was put in condition with monitoring. The otorhinolaryngological examination by nasofibroscope revealed a Florida laryngeal papilloma in the form of whitish tufts of ripening papillomas organized in clusters on the vocal cords (**Figure 1**). The glottic floor was affected, hence the presence of dysphonia in our patients. Furthermore, there was no tracheal or pulmonary involvement. Strong peeling was performed (**Figure 2**). With good immediate progress postoperatively, the patient became eupneic, and the saturation increased to 99% in ambient air. The histopathology study of the excision fragments confirmed the diagnosis of a laryngeal papilloma by showing a proliferation of papillomatosis architecture made up of well-differentiated acanthotic squamous cells forming a thickened hyperplastic coating. No atypia and mitosis (**Figure 3**) were observed. The study of the serotype responsible for papillomatosis is in progress. The patient is currently in regular follow-up with his surgeon.



Fig 1: ripening lesions on the vocal cords



Figure 2: Strong peeling was performed

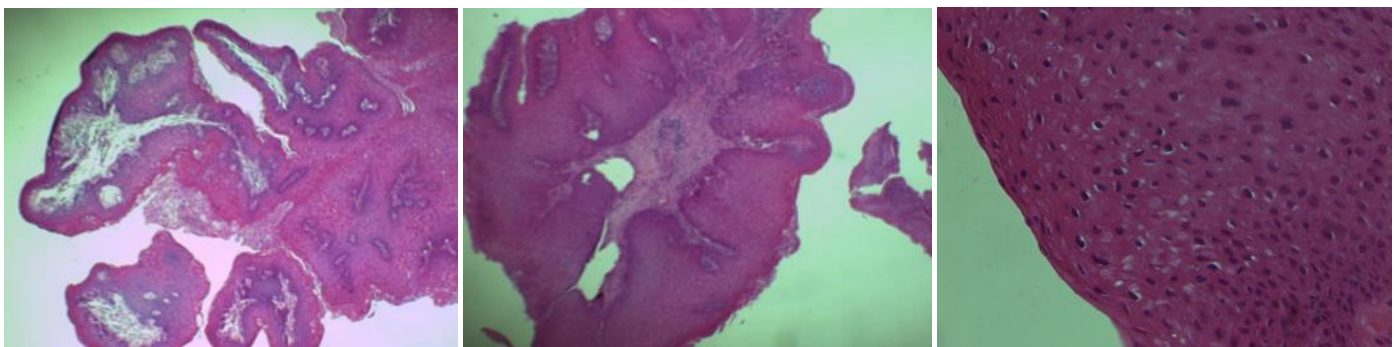


Figure 3: No atypia and mitosis

Discussion

Laryngeal papillomatosis is a benign tumoral proliferation of the larynx. The etiopathogenesis remains uncertain, but the role of HPV infection has been confirmed in recent studies [2]. It is the most common benign tumor of the larynx in children [2]. It is a rare condition in developed countries. Cristensen [3] reports 0.2 to 0.7 cases per 100,000 inhabitants in Sweden. Bomholt [4] reports 2 cases per year per center in the United States. Malick in Senegal finds 3 cases per year on average [5]. In Morocco, the incidence is on average 2.5 cases per year [6]. A male predominance is most often found in the majority of published studies. This condition is usually described in children [7].

The deadline for the first consultation is most often delayed. Kpemissi et al. [8] note a delay of three to six months. This period may be longer because papillomatosis is often assimilated to asthma or chronic laryngitis treated repeatedly in pediatric emergencies.

The first symptom is chronic dysphonia, hence the rule that states that one must examine the larynx of any patient with dysphonia for more than 15 days. It is strongly suggested by the endoscopic aspect and must be confirmed by a histological study [2]. Seen late, the patient presents with dyspnea of the inspiratory bradypnea type, with indrawing and horniness [9].

The management of laryngeal papillomatosis is only symptomatic and palliative. No curative treatment is currently available. Endoscopy under general anesthesia makes it possible to establish the lesion assessment; it shows tufts of papillomas with a raspberry, mature-like, greyish-pink appearance, in clusters. These lesions usually start on the floor of the ventricles, on the anterior commissure and on the anterior third of the vocal cord and can extend to the entire larynx or even the hypopharynx and the tracheobronchial tree. Peeling is done with forceps under direct laryngoscopy in suspension. Histological study of papilloma's is

necessary, particularly in adolescents where malignant transformation is to be feared [9]. Nevertheless, the anatomopathological examination must be carried out after each peeling. The HPV types implicated in this condition are HPV 6 and HPV 11, which represent the two HPV types most commonly associated with condyloma acuminata. It is well established that children with laryngeal papillomatosis have been infected from the mother during childbirth [9].

Tracheotomy remains the only emergency rescue alternative [10]. However, this life-saving gesture would significantly compromise the long-term prognosis of the disease. Many authors report that tracheostomy is a factor in the dissemination of papillomatosis in the tracheobronchial tract. Although its role is controversial, it should be avoided [11-14].

Endoscopic CO2 laser vaporization under general anesthesia is a treatment alternative that is increasingly used today [15]. The microdebrider has recently been proposed for the treatment of laryngeal papillomatosis. Bleeding would be greater intraoperatively, but the follow-up is just as simple as the CO2 laser [16].

There are unfortunately serious forms, involving the vital prognosis. The forms at risk are those that begin very early, before 1 year (they extend to the lower airways), and those due to HPV 16 or 18. Drug treatments have been or are being tried in these latter forms. For the past year, a clinical trial has been evaluating intralesional injections of bevacizumab [17]. There are also treatment trials with HPV vaccines. Death can occur by invasion of the lower airways, but also by malignant transformation [18].

Several authors report the value of adjuvant treatment in the aggressive forms defined according to the criteria of Doyle et al. [19]. Interferon alpha-2a in daily or weekly subcutaneous injections remains to be discussed given its poorly tolerated side effects such as asthenia, headache, fever and alopecia. Other published studies

report the usefulness of cidofovir despite its high cost. Indeed, cidofovir is a nucleotide analogue of cytosine with antiviral activity. It can be used by intra-lesional injection after surgical removal of the papillomas or by in situ injection within the papillomas [2]. However, the results of this technique depend on the concentration of the product, the interval between administrations and the number of injections [20]. Few side effects after intralesional use of Cidofovir have been detected. Side effects relate to nephrotoxicity and carcinogenic potential. The latter has been a source of debate to lead to the absence of risk added to the risk of spontaneous dysplasia within the papilloma's [21]. The use of HPV 6 and HPV 11 vaccines has been tried as an adjuvant treatment for recurrent and aggressive forms and in patients whose use of one or two adjuvant therapies has not produced any effects [19, 22]. Other studies emphasize the need to treat any associated gastroesophageal reflux with antacids [23]. The vital prognosis is involved in the diffuse forms and the totally obstructive forms of the glottic stage. The same applies to cases of cannular obstruction by mucous plug responsible for death, especially in children whose cannulas are small in diameter. Ondzotto et al. [9] reported two cases of death in their series due to mucus plug. In case of dissemination, the malignant transformation of pulmonary papillomatosis into squamous cell carcinoma is a rare complication. This malignant development has mainly been reported in patients whose papillomas had been treated by irradiation. It has been observed that HPV 11 infections have a more aggressive course than HPV 6 infections [1,25]. The functional prognosis depends on the frequency of peeling with forceps. Indeed, repeated trauma causes hoarseness. The unpredictable and frequent recurrences of this condition make its evolution capricious, which requires regular monitoring of patients.

Conclusion

Laryngeal papillomatosis is a relatively rare but dreaded benign tumor of the larynx. Its diagnosis is usually easy as long as a laryngoscopic examination is systematically carried out in the face of chronic dysphonia in children. Peeling, over several sessions, remains the only therapy available and accessible. No etiological treatment is currently available. Several adjuvant therapies are under development. Its capricious evolution requires regular monitoring. Histological study is essential given the risk of malignant transformation of papillomas.

List of abbreviations

LP: Laryngeal papillomatosis Human
HPV: Papilloma Virus

Ethics approval and consent to participate

Not applicable

Data Availability

Not applicable

Conflicts of Interest

The authors declare that there is no conflict of interest regarding the publication of this paper.

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Authors' contributions

This work was carried out in collaboration among all authors. NM and CM designed the study, wrote the protocol, wrote the first draft

of the manuscript. NM, and CM managed the literature searches. All authors read and approved the final manuscript.

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