



A Patient with Juvenile Recurrent Respiratory Papillomatosis Complicated by Laryngeal Stenosis after Laryngeal Microsurgery: A Clinical Case

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Abstract

BACKGROUND: The relevance of the problem of juvenile respiratory papillomatosis lies in the recurrent course of the disease in young children. This can lead to the development of respiratory stenosis, requiring repeated, in some cases, monthly surgical treatments for endolaryngeal removal of papillomas, which sometimes leads to persistent scar stenosis of the larynx. We present clinical case of achieving remission of recurrent juvenile papillomatosis of the larynx.

CLINICAL CASE: Patient, 5 years old, who applied in February 2016, was hospitalized in the ENT department with severe dyspnea, laryngoscopy revealed laryngeal papillomatosis with 3–4 grade stenosis. Over the past 10 months, the patient has undergone laryngeal surgery 6 times using cold instruments and a CO₂ laser due to papillomatosis. Taking into account the recurrent nature of the course of the disease, the patient is administered to consult an allergist-immunologist. Based on the anamnesis and the results of the tests, the immunologist-allergist diagnosed: Primary immunodeficiency, unspecified (insufficiency in the production of specific antibodies), persistent allergic rhinitis, in the stage of incomplete remission and prescribed treatment. The patient was operated twice from 2017 to 2018 for a developed post-operative complication – cicatricial stenosis of the larynx. With these surgical repairs, no recurrence of laryngeal papillomatosis was detected. Furthermore, in 2021, according to endovideolaryngoscopy, remission was confirmed. R In this clinical case, we managed to achieve remission of laryngeal papillomatosis in a patient after surgery and treatment by an immunologist. Allergen elimination and immunocorrection helped us achieve deep remission. Surgical treatment eliminated laryngeal stenosis, recovering breathing and voice quality. Furthermore, this clinical case demonstrates the importance of studying the immune status in patients with laryngeal papillomatosis.

CONCLUSION: The results obtained by us show the need for the use of immunomodulators in these patients. All children with prolonged hoarseness should undergo laryngoscopy, which will allow diagnosing laryngeal papillomatosis at earlier uncomplicated stages of the disease. Treatment of laryngeal papillomatosis should be comprehensive and in addition to surgical treatment should include differential immune correction.

RESULTS AND DISCUSSION: In the above clinical case, we managed to achieve remission of laryngeal papillomatosis in a patient after surgery and treatment by an immunologist. Allergen elimination and immunocorrection helped us achieve deep remission. Surgical treatment eliminated laryngeal stenosis, recovering breathing and voice quality. Furthermore, this clinical case demonstrates the importance of studying the immune status in patients with laryngeal papillomatosis. The results obtained by us show the need for the use of immunomodulators in these patients. All children with prolonged hoarseness should undergo laryngoscopy, which will allow diagnosing laryngeal papillomatosis at earlier uncomplicated stages of the disease. Treatment of laryngeal papillomatosis should be comprehensive and in addition to surgical treatment should include differential immune correction.

Introduction

Laryngeal papillomatosis is one of the global problems in the field of laryngology, since the exact etiopathogenesis of the disease is still not known. It is believed that about 10–60% of the world’s population is infected with the human papillomavirus (HPV) latently, but not all get laryngeal papillomatosis, the causative agent of which is type 11 and 16 of this virus [1]. Juvenile respiratory laryngeal papillomatosis suggests that infection occurs secondarily, through the natural birth canal of the mother. The virus penetrates through

the damaged area of the mucous membrane and affects the basal cells of the multilayered squamous epithelium [2]. Respiratory laryngeal papillomatosis is a benign neoplasm with a very low risk of dysplasia [3].

About 3.5% of neoplasms of ENT organs are laryngeal papillomatosis, which has more than 50 types of treatment [4]. The larynx and trachea are often affected by the HPV, and the oropharynx, nasopharynx, nose, and oral cavity are involved less often. HPV affects the distal airways in 48% of children. Only 3.3% of cases had lung damage. At the same time, the incidence of lung cancer among these patients was 16% [1]. It is also very rare to detect isolated papillomatosis of the trachea

and lungs without involving the larynx [5]. Low-frequency radiation laser ablation in the treatment of laryngeal papillomatosis has shown its effectiveness in practice [6]. Removal of papillomatosis with a blue laser is basically a new method and has shown its effectiveness, since it removes tissues with minimal carbonization and minimal visible impact on surrounding tissues [7].

Along with surgical methods of treatment, conservative treatment of laryngeal papillomatosis is also used, in which there is a slow growth of the tumor. For example, treatment with cidofovir significantly lengthens the remission time of recurrent respiratory laryngeal papillomatosis [8] [9]. The study showed that HPV vaccination reduces the development of recurrent laryngeal papillomatosis [10].

Study design

Clinical case.

Objective

Surgical repair of laryngeal stenosis and achieving remission of laryngeal papillomatosis.

Materials and Methods

Patient, 5 years old, came to us in February 2016 with the complaints of persistent hoarseness, asthma attacks, and dyspnea for 4–5 months. The child was hyposthenic, covered with cold sweat. Breathing was rapid, with a characteristic stenotic noise when breathing. During inhalation, there was a retraction of intercostal spaces, jugular, and supraclavicular pits. Cyanosis of the lips, mucous membranes, and skin were observed. Examination with fiber-optic laryngoscope showed a widespread growth of laryngeal papillomas complicated by stenosis of Grades 3–4 (Figure 1).



Figure 1: Endolaryngoscopic picture of the larynx before surgery

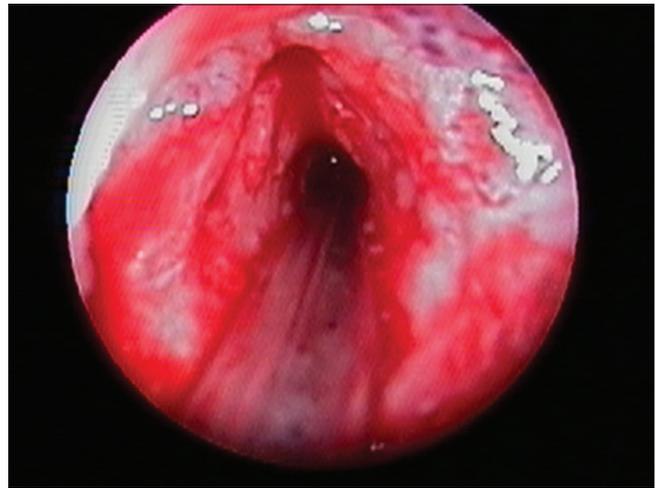


Figure 2: Endolaryngoscopic picture after surgery

The patient was hospitalized, emergency tracheostomy and endolaryngeal papilloma removal surgery were performed.

The patient contacted us again after 10 months. It turned out from the anamnesis that during this period of time, the child was subjected to endolaryngeal microsurgical repairs 6 times: With cold instruments and a CO₂ laser at intervals of 1.5–2 months due to recurrent papillomatosis (Figure 2) and decannulated after two surgical treatments. Diagnosis: Laryngeal papillomatosis, common form, recurrent course with 3–4 grade stenosis.

Taking into account the widespread growth of papillomas in the larynx, frequent relapse, the patient was recommended to consult an immunologist-allergist (December 2016). An immunologist was examined and the results of the tests showed the following:

Analysis from December 29, 2016	
Total IgE	44.45 MU/ml
CLIA HPV IgM	0.27
CMV IgG	26.53
HBsAg	2.0 MU/ml
ELISA-HPV IgM	Not detected
HPV: Human papillomavirus	
Skin allergic testing from December 29, 2016	
Pillow feather	+++
House dust mite	++
Sheep wool	++
Whole chicken egg	++
Aspergillus	++++
Test control	+

Based on the anamnesis and the results of the tests, the immunologist-allergist diagnosed: Primary immunodeficiency, unspecified (deficiency in the production of specific antibodies); allergic rhinitis, persistent form, moderate course, and stage of incomplete remission. Basic medical treatment was prescribed: (1) Lycopene 1 mg for 10 days under the tongue in between meals. Repeat the course in a month. (2) Inhalation of B2 fast-acting agonists (Berodual solution) through a nebulizer 12 drops. (3) Montelukast sodium 5 mg 1 tablet at night for 1 month, then a repeat course after 15 days. (4) Xyzal/Zyrtec 1 capsule × 2 times a day from the 1st days of ARVI for 7–10 days.

(5) Acyclovir 200 mg 1 tablet 4 times a day, after eating for 6 days (Aciclostad) (compliance with a fat-free diet, due to hepatotoxicity, nephrotoxicity of the drug).
 (6) Viferon 500.000 units during the 2 months.

Recommendations were also given on the observance of hypoallergenic life: Hygiene of the nose and eyes, wearing sunglasses, masks from April to September, to exclude contact with dust, feather, wool, dampness, chemicals (chlorine and chlorine-containing products), hygiene of the home, daily peak flow monitoring, and hypoallergenic diet. The patient underwent two courses of treatment with an allergist-immunologist in 2017.

The treatment made it possible to achieve remission within 3 months. Previously, relapses occurred monthly and surgical repair was required. Subsequently, the patient underwent endolaryngeal surgery once for laryngeal papillomatosis with minimal overgrowth. Also patient was undergoing endolaryngeal surgery on vocal cord twice due to a postoperative complication—cicatrical stenosis of the larynx (membranous vocal folds) from 2017 to 2018, where a remission of laryngeal papillomatosis was established. Further follow-up of the patient with a phoniologist and an immunologist-allergist is recommended.

The control fibrolaryngoscopy of the patient was performed in September 2021 (Figure 3). As shown in Figure 3, there is a post-operative scar and a slight deformation of the left vocal cord. At present, no recurrence of respiratory laryngeal papillomatosis has been detected. The patient's voice was sonorous and without hoarseness.

In 2017, a second examination was carried out by an allergist-immunologist and repeated tests were

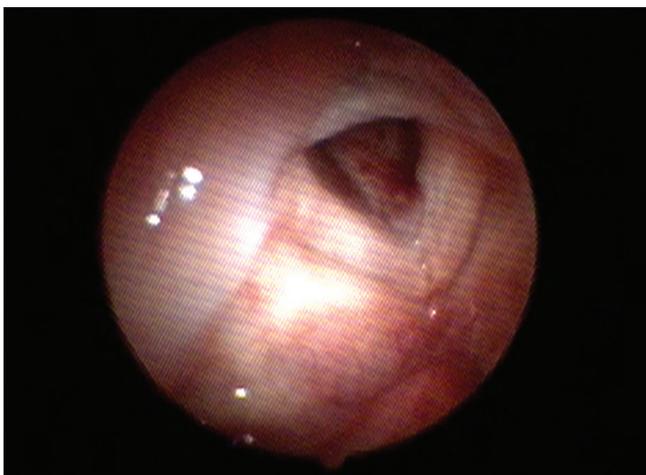


Figure 3: Endovideolaryngoscopy of the larynx

prescribed to assess the immune status. According to the analysis results: Total IgE – 30.45 MU/ml; HPV IgG – 9.93; and CMV IgG – 20.53; there is a decrease in indicators, which indicates the effectiveness of the treatment. According to clinical indicators, there is a significant improvement, which is indicated by

the absence of relapses. Further observation by an otolaryngologist, an allergist-immunologist, and repeated courses of treatment, if necessary, is recommended.

Conclusion

The study in 2021 showed that the patient is in remission. Children with prolonged hoarseness must undergo laryngoscopy. Furthermore, this clinical case shows the importance of studying the immune status of patients with laryngeal papillomatosis, which allows us to identify changes in cellular and humoral connections of the immune status – a decrease in all populations of T-lymphocytes and quantitative deficiency of B cells, a decrease in the main classes of immunoglobulins. These results make it reasonable to use immunomodulatory therapy in these patients. Treatment of laryngeal papillomatosis should be comprehensive and in addition to surgical treatment should include differential immune correction.

Ethics Statement

This clinical case was part of the clinical observation by the staff of our department.

Authors' Contributions

All authors declare that there are no conflicts of interest regarding the present study.

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