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Case Report

A Challenging Case of Severe Recurrent Respiratory Papillomatosis in Childhood Requiring 28 Surgical Interventions: A Case Report

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ABSTRACT

Recurrent respiratory papillomatosis (RRP) is a chronic human-papillomavirus associated disease characterized by recurrent benign papillomatous growth in the respiratory tract. Despite its benign histology, it may follow an aggressive clinical course requiring repeated surgical intervention. We report a 8-year-old girl with severe RRP managed over 6 years. Symptoms began in infancy with a weak cry and progressed to persistent hoarseness, noisy breathing, chronic cough, and recurrent episodes of airway obstruction. She underwent her first direct laryngoscopy and clearance biopsy at 2 years and 6 months of age. Since then, she has required 28 surgical procedures, averaging four per year. Two procedures were performed emergently due to airway compromise, and the patient required three postoperative intensive care admissions and one tracheostomy. Histopathology confirmed squamous papilloma without dysplasia or malignant transformation. The disease imposed significant educational, psychological, and financial burdens. This case highlights the aggressive nature of juvenile-onset RRP and the challenges of long-term management, particularly in resource-limited settings.

Key words: Recurrent respiratory papillomatosis, human papillomavirus, laryngeal papilloma, direct laryngoscopy, tracheostomy

INTRODUCTION

Recurrent respiratory papillomatosis (RRP) is a chronic disease of the aerodigestive tract that occurs in both children and adults. It is characterized by the growth of benign squamous papillomas within the respiratory epithelium. RRP is traditionally classified into juvenile-onset and adult-onset forms, depending on whether symptoms begin before or after 12 years of age, respectively. [1, 2] It is caused predominantly by low-risk human papillomavirus (HPV) types 6 and 11, with HPV-11 being associated with a more aggressive clinical course and earlier onset of disease. RRP tends to take a more aggressive clinical course in children and can be fatal because it tends to recur and spread throughout the respiratory tract. [3–5]

The prevalence of this disease is likely variable depending on the age of presentation, country, and socioeconomic status of the population being studied, but is generally expected to be between 1 and 4 per 100,000. [2, 6] In an 11-year retrospective review conducted in Enugu, Nigeria, Mgbor et al. analyzed 54 cases of laryngeal papillomatosis and reported that 64% occurred in pediatric patients (≤15 years), with the majority presenting with upper airway obstruction requiring emergency tracheostomy. [7]

Vertical transmission of HPV during vaginal delivery is believed to be the principal mode of acquisition in children, particularly in cases where the mother has an active genital HPV infection. [8]

Clinically, most commonly presents with progressive hoarseness, weak cry, stridor, and varying degrees of respiratory distress. Less common presenting symptoms include chronic cough, recurrent pneumonia, and failure to thrive, especially in infants with an upper respiratory tract infection. A diagnosis of asthma, croup, allergies, vocal nodules, or bronchitis is entertained before a definitive diagnosis is made. [1, 3]

Management of RRP remains primarily surgical, to maintain airway patency and preserve voice quality rather than achieving a cure. [3, 5]

We report a case of severe RRP in an 8-year-old girl managed at the Department of Otorhinolaryngology, Federal Teaching Hospital Katsina, Katsina, Nigeria, who underwent 28 direct laryngoscopic clearance procedures over 6 years, highlighting the challenges of long-term management and the significant morbidity associated with aggressive disease.

CASE REPORT

An 8-year-old female who resides with her parents in Katsina State, Nigeria. She has been managed for juvenile-onset RRP (JORRP) since early childhood. She is the fifth child of her mother. Maternal history revealed a supervised pregnancy carried to term and delivered via spontaneous vaginal delivery without prolonged labor or instrumental assistance. There was a history of maternal vaginal infection with discharge during the pregnancy. HPV testing was not performed for the mother, and she has not received the HPV vaccination due to limited resources.

The illness was first noted in infancy when the mother observed a weak cry, which progressively evolved into noisy breathing, persistent hoarseness, chronic cough, and difficulty in breathing by the age of 2 years.

There is no history of neck trauma, prior instrumentation, foreign body inhalation, corrosive ingestion, asthma, or allergic disease, and no family history of similar illness. No history of fever or drenching night sweat. She is fully immunized according to the National Programme on Immunization schedule.

Developmental history revealed normal attainment of age-appropriate milestones. The child achieved head control at 3 months, sat without support at 6 months, walked independently at 13 months, and had normal speech development for age.

On examination, flexible laryngoscopy revealed multiple papillomatous lesions involving the false vocal cord and anterior commissure, resulting in airway narrowing. Examination of the oral cavity, oropharynx, nose, and ear was normal. X-ray soft tissue neck showed opacities around the laryngeal region with a narrowed air column.

Histopathological examination of tissue obtained during direct laryngoscopy and clearance biopsies demonstrated a papillomatous lesion lined by acanthotic stratified squamous epithelium overlying a fibrocollagenous stroma with congested vascular channels, consistent with laryngeal squamous papilloma, with no evidence of dysplasia or malignant transformation.

She underwent her first direct laryngoscopy and clearance biopsy at the age of 2 years and 6 months. Since then, the disease has followed an aggressive course with frequent recurrences. Symptoms typically reappeared approximately 3 months after each procedure, with the patient remaining symptom-free for an average duration of 8 weeks.

She has required an average of four surgical interventions per year, with a total of 28 direct laryngoscopy and clearance biopsy procedures performed to date.

Two of these procedures were undertaken as emergency surgeries due to significant airway compromise. The patient required three postoperative admissions to the intensive care unit and underwent one tracheostomy during the course of the disease.

Most direct laryngoscopy and clearance biopsies were performed using cold steel techniques with forceps. A single procedure was carried out using a microdebrider, after which symptom recurrence occurred in less than 6 weeks. Adjuvant therapy with acyclovir and systemic prednisolone was administered and was associated with a modest prolongation of the interval between surgical interventions.

The illness has significantly affected her education, resulting in frequent school absenteeism and missed examination periods. It has also imposed a substantial financial burden on the family, and both the patient and her caregivers experience considerable fear and anxiety related to repeated surgical procedures. Despite multiple interventions, she continues to have persistent hoarseness; however, her speech remains intelligible, with no significant impairment in communication.

She is currently clinically stable, with preserved airway patency following the most recent intervention. Nevertheless, due to the early onset and aggressive nature of the disease, the risk of recurrence remains high. The long-term prognosis is variable, with some patients achieving remission after puberty, while others may continue to require periodic interventions (**Table 1**; **Figures 1–5**).

DISCUSSION

RRP is characterized by the formation of exophytic proliferative lesions of connective tissue covered by epithelium anywhere in the respiratory tract, with a predilection towards the squamocolumnar junction, primarily involving the larynx, especially the vocal cords. [9] There is a bimodal age distribution: JORRP occurs in children below 12 years, and adult-onset RRP (AORRP) occurs after 12 years of age. [2, 9]

HPV is the established etiology for JORRP, with HPV types 6 and 11 being the most frequent strains. HPV types 6 and 11 are associated with condyloma acuminata. [2, 8, 9] Therefore, infants of mothers with genital warts carry a high risk of acquiring HPV vertically through vaginal birth. The characteristic triad increasing the likelihood of JORRP includes being the firstborn, vaginal delivery, and having a young mother. [8]

Table 1: Timeline of surgical interventions.

Age (years)	Approximate number of procedures	Clinical indication	ICU admission	Tracheostomy	Surgical technique
2.5	1 (first surgery)	Initial airway obstruction	Yes (1 episode)	No	Cold steel
3–5	12	Recurrent hoarseness and stridor, including emergencies	Yes (2 episodes)	Yes (1 episode)	Cold steel; microdebrider (1 procedure)
5–7	10	Recurrence	No	No	Cold steel
7–8	5	Recurrence	No	No	Cold steel
Total	28 procedures		3 ICU admissions	1	Predominantly cold steel

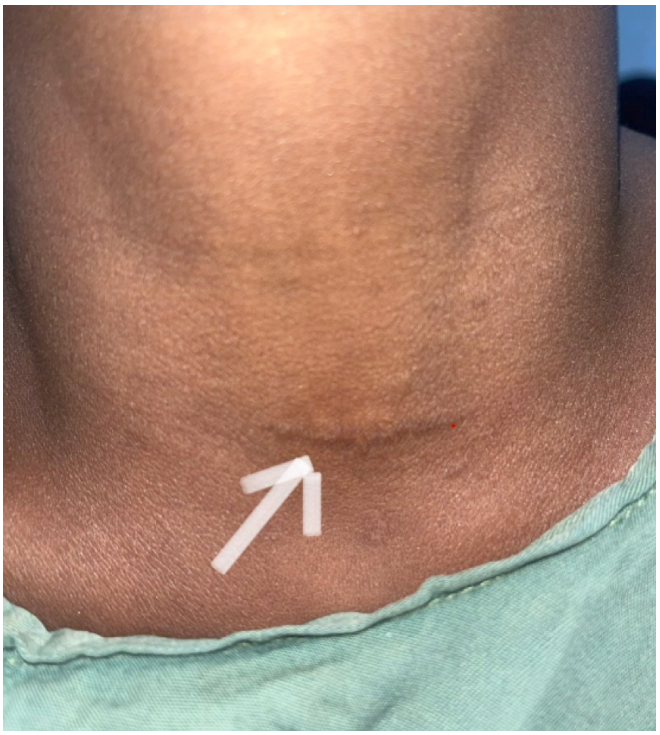

Figure 1: Healed tracheostomy scar on the anterior neck (arrow) following tracheostomy performed during the course of treatment.

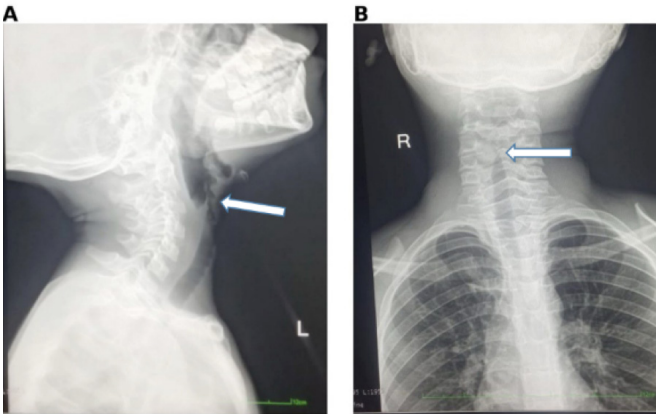

Figure 2: Soft tissue neck radiographs of the patient demonstrating laryngeal airway narrowing. **(A)** Lateral view showing irregular soft tissue opacities within the laryngeal region with narrowing of the upper airway column. **(B)** AP view showing relative narrowing of the upper airway at the laryngeal level.

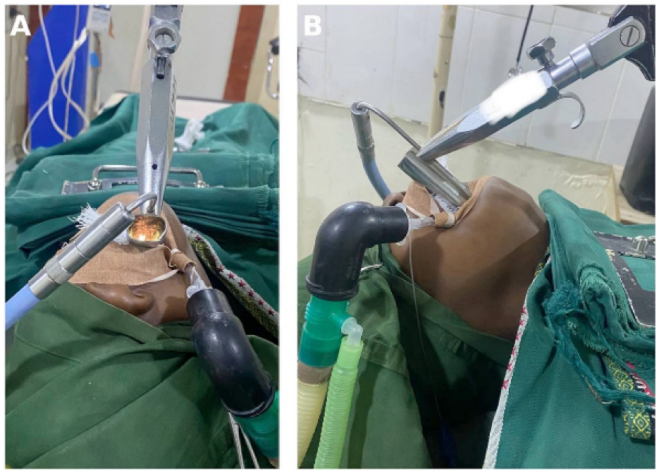

Figure 3: Intraoperative setup during direct laryngoscopy and clearance biopsy. **(A)** Patient positioned for direct laryngoscopy with suspension laryngoscope in place and operating light source attached. **(B)** Side view showing suspension laryngoscopy with anesthetic ventilation circuit during airway exposure.

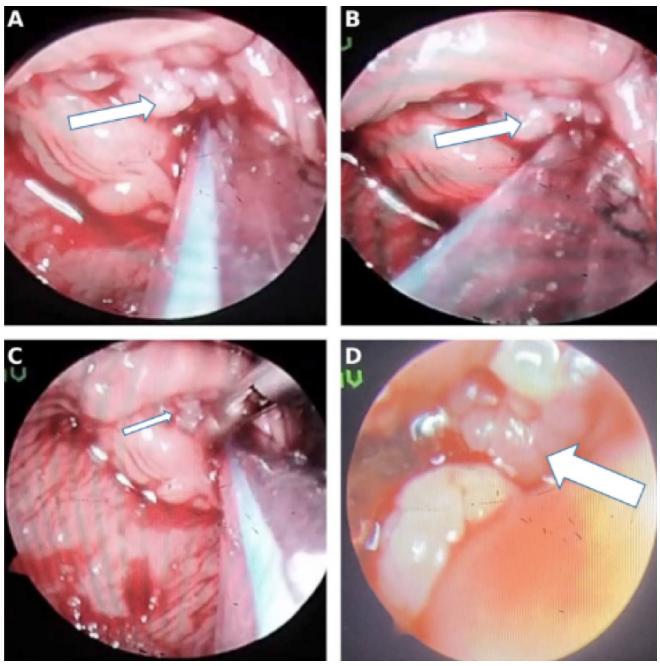

Figure 4: Endoscopic views of the laryngeal inlet showing papillomatous lesions. **(A–D)** Multiple exophytic papillomatous growths involving the supraglottic and glottic region were observed during direct laryngoscopy and surgical clearance.

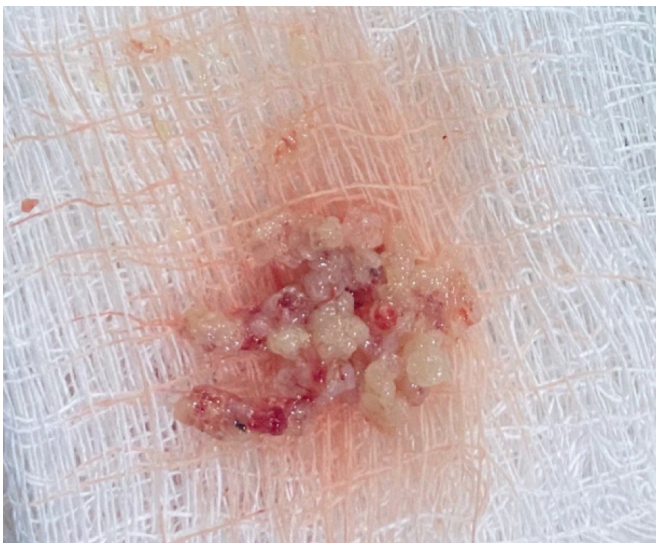

Figure 5: Gross appearance of excised laryngeal papillomatous tissue obtained during direct laryngoscopy and clearance biopsy showing small, multiple whitish, lobulated papillomatous fragments.

A history of maternal vaginal infection during pregnancy in this case further supports this possibility, although definitive virological confirmation was not feasible. The relative risk of developing RRP after vaginal delivery ranges from two to seven in 1000, corresponding to an odds ratio of 231 to 400 compared with children born vaginally without condylomata, showing a low risk of transmission. [10] The HPV DNA has been identified in the upper airways of as many as 25% of normal, unaffected children. This has fueled debate on the benefit of offering caesarean section routinely to mothers with anogenital condylomata. [10] JORRP occurs in one of 109 children delivered via caesarean section to a mother with HPV infection of the anogenital region. This suggests other modes of transmission, among which is the hematogenous spread of HPV to the fetus while in utero, as HPV has been demonstrated in cord blood. [11]

Although HPV typing was not performed in this case due to a lack of availability, the disease's clinical severity, including 28 surgical procedures over 6 years, multiple emergency airway interventions, ICU admissions, and the need for a tracheostomy, raises a strong suspicion of a potentially more aggressive HPV subtype. [1, 2]

The patient in this report had persistent hoarseness, noisy breathing, and difficulty breathing. Most reviews show that hoarseness of voice is the principal presenting symptom among pediatric patients. [9, 11] Stridor, which is the second common symptom, begins as inspiratory and then progresses to biphasic. [11] Early onset of symptoms is generally thought to indicate either higher viral activity or greater susceptibility in the child's immune system, both of which can lead to faster regrowth of the papillomas after they are removed surgically. [5]

The natural history of RRP is highly variable; patients may experience a lifelong remission after initial disease, whereas some will require periodic surgeries ranging from days to weeks in addition to adjuvant medical therapies. [2, 6] The treatment of RRP remains primarily surgical, with the main goals being to maintain airway patency and preserve voice quality rather than achieve complete eradication of the disease, and the current standard of care involves surgical excision aimed at complete removal of the papillomas while preserving normal anatomy. [1, 3]

In patients who have anterior or posterior commissure disease or highly aggressive papillomas, the goal may be sufficient removal to clear the airway while preserving normal structures so as to avoid complications of subglottic and glottic stenosis, web formation, and resulting airway stenosis. [5] Indications for adjuvant therapy generally include the need for four or more surgical procedures per year, rapid regrowth, or distal airway involvement, and have demonstrated benefit in reducing surgical frequency in severe cases. [9, 11]

Our patient required an average of four procedures per year and experienced episodes of emergency airway obstruction, which is consistent with severe disease. Although formal Derkay severity staging was not documented at initial presentation, the overall clinical pattern in this case aligns with severe disease based on established criteria. [12] The severity of disease in this patient can be attributed to multiple factors, including early age at onset, frequent recurrences, and episodes of airway compromise necessitating emergency intervention and tracheostomy. These are well-recognized predictors of aggressive JORRP. In addition, limited access to advanced adjuvant therapies such as bevacizumab or cidofovir may have contributed to continued disease recurrence, highlighting the impact of resource constraints on disease control and long-term outcomes. [13]

Cold steel excision remains widely practiced, particularly in resource-limited settings; however, microdebrider and laser techniques have been associated with improved precision and reduced tissue trauma in some reports. [5, 7, 14] Pasquale et al. reported improved voice quality, less operating room time, less mucosal injury, and a cost benefit for the microdebrider compared with the CO2 laser. [14]

In our case, only one procedure was performed using a microdebrider, and recurrence occurred within 6 weeks, suggesting that surgical modality alone does not necessarily alter the biological behavior of aggressive disease. This aligns with evidence that recurrence is primarily driven by persistent HPV infection rather than incomplete excision. [15]

Tracheostomy is generally avoided in RRP due to the risk of distal spread along the tracheostomy tract and potential seeding of the lower airway. [16] Nevertheless, it may become unavoidable in cases of severe airway obstruction, as occurred in this patient.

Malignant transformation in RRP is uncommon, affecting fewer than 1% of juvenile-onset cases. However, prolonged disease duration, a high number of surgical procedures, and infection with HPV-11 are recognized as possible risk factors. [2] In this patient, repeated histopathological examinations have consistently shown a benign squamous papilloma with no evidence of dysplasia or malignant transformation, which is reassuring.

In addition to its physical effects, RRP carries a substantial psychosocial and economic burden. Severe cases are well known to cause repeated surgeries, exposure to anesthesia,

frequent hospitalizations, missed school days, and significant parental stress and anxiety. [1, 2] The financial burden described in this case reflects the chronic and resource-intensive nature of management, particularly in low and middle-income settings where access to specialized care is restricted.

HPV quadrivalent vaccination has shown substantial effectiveness in reducing the incidence of cervical cancer, adenocarcinoma in situ, and cervical intraepithelial neoplasia 1 to 3, vulvar and vaginal intraepithelial neoplasias grades 2 to 3, and genital warts associated with HPV 6, 11, 16, and 18 infection and may indirectly reduce the future incidence of RRP through decreased maternal transmission. [5] Emerging evidence also suggests a potential therapeutic role of HPV vaccination in reducing recurrence in affected individuals. [13, 17] Neither the patient nor her mother had received the HPV vaccine, representing a missed opportunity for prevention and emphasizing the critical need to broaden HPV vaccination coverage.

CONCLUSIONS

Although RRP is histologically benign, it may follow a highly aggressive clinical course. Early age at onset, frequent recurrences, and the need for repeated surgical interventions are key indicators of severe disease. This case highlights the significant medical, psychosocial, and economic burden associated with long-standing RRP, particularly in resource-limited settings. Improved access to advanced adjuvant therapies and expanded HPV vaccination coverage may help reduce disease severity and prevent future cases. Long-term follow-up remains essential for monitoring airway status and detecting potential complications.

CONSENT

Written informed consent was obtained from parents for the publication of this case report and all associated images.

AUTHORS' CONTRIBUTION

All authors have significantly contributed to the work, whether by following the case at the bedside, conducting literature searches, drafting, revising, or critically reviewing the article. They have given their final approval of the version to be published, have agreed with the journal to which the article has been submitted, and agree to be accountable for all aspects of the work.

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CONFLICT OF INTEREST

None.

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