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Case Report

Recurrent respiratory papillomatosis in a 5-year old child from sokoto, Nigeria: A masquerader of childhood bronchial asthma

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ABSTRACT

Recurrent respiratory papillomatosis (RRP) is a rare, chronic disease caused by the human papilloma virus. A delay in appropriate intervention could be caused by the symptomatology being similar to bronchial asthma. A 5-year-old boy presented to the pediatric emergency unit of Usmanu Danfodiyo University Teaching Hospital with a 2-year history of recurrent difficulty in breathing, cough and noisy breathing. He had previously received several treatments for asthma at Primary Health Care (PHC) facilities. On examination, he was dyspneic with a respiratory rate of 60 cycles/minute, oxygen saturation was 86% on room air, and chest auscultation showed vesicular breath sounds. Initially, acute severe asthma was diagnosed. However, further examination revealed wart-like lesions protruding below the pharynx, and a diagnosis of respiratory papillomatosis was considered. He had an emergency tracheostomy and video-assisted laryngoscopy with excision of the lesions. The histological report confirmed a laryngeal papilloma. His clinical condition improved after removal of wart-like lesions, and he was discharged afterwards. He had three relapses and surgical removal (September and December 2023, June 2024). Currently, he is stable and is being followed up at the Pediatric and Otorhinolaryngology Clinic.

1. Introduction

Respiratory papillomatosis is a rare disease of the upper airway with significant morbidity and risk of mortality from airway obstruction.[1–3] It is caused by human papilloma virus (HPV) types 6 and 11 usually transmitted from mother to child during the peri-partum period.[4] Respiratory papillomatosis can progress to recurrent respiratory papillomatosis (RRP) which is characterized by exophytic wart-like lesion of the airway with the potential to spread throughout the respiratory tract, in addition to minimal risk of malignant transformation.[2,5] The course of RRP is said to be variable depending on whether it's the aggressive or non-aggressive type, as the aggressive type is associated with rapid recurrence.[2] Two sub-types of RRP exist: the juvenile onset RRP and the adult onset RRP, based on diagnosis before or after 12 years of age respectively.[2] The risk factors for the development of juvenile onset RRP include vaginal delivery, being a first-born, low socio-economic status and maternal age younger than 20 years.[2,6] Clinical manifestations include nonspecific airway symptoms such as progressive hoarseness, chronic cough, shortness of breath, stridor and wheezing.[5,6] Conditions such as bronchial asthma, viral croup, foreign body, laryngeal hemangioma and laryngomalacia can mimic RRT.[1] Respiratory papillomatosis is usually associated with

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delays in presentation, diagnosis as well as treatment.[5]

2. Case Presentation

We report the case of A.A, a 5-year-old boy who was brought by his parents on self-referral to the Emergency Paediatric Unit of Usmanu Danfodiyo University Teaching Hospital (UDUTH) Sokoto, Nigeria, in April 2023. He had a 2-year history of recurrent cough and difficulty in breathing with associated hoarseness and noisy breathing. Complaints had progressively worsened since onset and became persistent 2 months prior to presentation. He had disturbance in sleep pattern but no histories of fever, nasal discharge, foreign body aspiration, refusal of feeds, poor weight gain, or allergy. He had received several treatments for bronchial asthma and pneumonia at PHC facilities as well as over the counter medications and traditional concussions with no significant improvement. There was no family history of similar complaints. He had no past histories of vaccine preventable diseases, hospital admission or surgery. During the 3rd trimester of pregnancy, the mother had febrile illness with associated itchy rashes in her mouth, on the knees and the calf, sparing the genital area. He was delivered at term via spontaneous vaginal delivery at a Primary Health Care (PHC) facility. The parents' age could not be ascertained. They had no formal education, the mother was a housewife and the father was a peasant farmer. Physical examination revealed an ill-looking child, dyspnoeic with tracheal tugging, stridulous breathing and oxygen saturation (SPO_2) of 86% on room air. His weight and Height were 15kg and 104cm respectively. His pulse rate was 160 beats/min; blood pressure 100/60 millimetres of mercury; Heart sounds were S1 and S2 with no murmur. His respiratory rate was 60 cycles/minute, percussion note was resonant, air intensity was markedly reduced, breath sounds were vesicular with no added sounds and he had normal heart sounds. Other systems were normal on examination. An initial diagnosis of acute severe asthma with differential of upper airway obstruction from foreign body aspiration was made.

2.1. Management and outcome

The patient was commenced on intranasal oxygen, he was nebulized using salbutamol (3 courses) and had intravenous hydrocortisone with no significant improvement. Further examination revealed warty lesions protruding from beneath the pharynx. The pharynx and tonsils were not inflammed. The diagnosis of Respiratory papillomatosis was entertained and consult was sent to the Otorhinolaryngologist. The Otorhinolaryngologist reviewed and the diagnosis of upper airway obstruction from recurrent respiratory papillomatosis with differential of foreign body aspiration was made. He had normal complete blood count and metabolic panel (Table 1). Human immunodeficiency virus DNA polymerase chain reaction was negative. He had emergency tracheostomy and video assisted direct laryngoscopy which revealed multiple grapelike warty lesions (Figure 1) that were subsequently excised under general anaesthesia.

Parenteral antibiotics (Ceftriaxone and metronidazole), Dexamethasone and Diclofenac were given post operatively to prevent wound infection, laryngeal oedema and pain control respectively. Histology report of the laryngeal tissue biopsy revealed fragments of tissue focally lined by non-keratinized stratified squamous epithelium thrown into papillary fold, some of which are inverted. The composing cells are bland round to oval having euchromatic nuclei and moderate eosinophillic cytoplasm in fibrocollenageous stroma with a diagnosis of laryngeal papilloma (Figure 2 & 3). The parents were counseled on the recurrent nature of RRT and He was subsequently discharged after one week on the ward. He has had recurrence of the lesions and surgical excision twice (September & December 2023, June 2024). He is currently stable and on routine follow-up at the Paediatric outpatient and Otorhinolaryngology clinics.

Figure 1: Endoscopic view of a laryngeal papillomatosis patient at the Usmanu Danfodiyo University Teaching Hospital Sokoto, Nigeria, April 2023 showing multiple grapelike warty lesions (Black arrow)

3. Discussion

Recurrent respiratory papillomatosis is a rare Paediatric condition of the larynx.[1,6] Although benign, it can cause significant morbidity and mortality from airway obstruction.[1–3] The age and gender of the index patient fell within the predominant age and gender as reported in the literature.[2] The implication of Laryngeal papillomatosis occurring at this young age is that our patient is at risk of recurrence as documented in a previous study,[7] and needs to be followed up on a regular and long term basis. Being

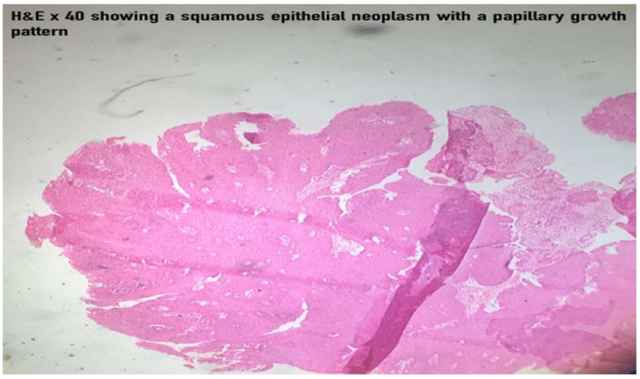

Figure 2: Histology report of a laryngeal papillomatosis patient at the Usmanu Danfodiyo University Teaching Hospital Sokoto, Nigeria, April 2023. Figure shows squamous epithelial neoplasm with a papillary growth pattern

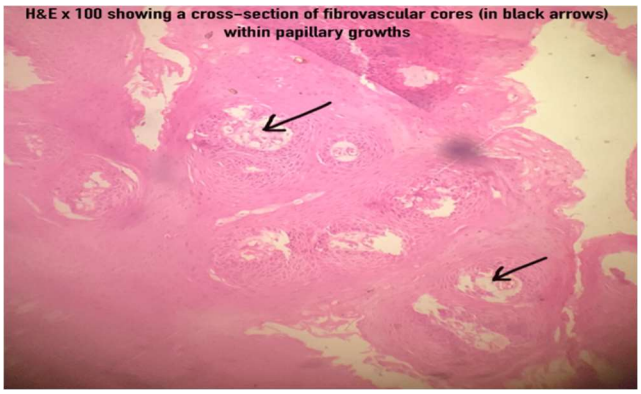

Figure 3: Histology report of a laryngeal papillomatosis patient at the Usmanu Danfodiyo University Teaching Hospital Sokoto, Nigeria, April 2023. Figure shows fibrovascular cores within papillary growths (black arrows)

Table 1: Laboratory data of the patient with laryngeal papillomatosis at the Usmanu Danfodiyo University Teaching Hospital Sokoto, Nigeria, April 2023.

Metabolic panel	Blood count
Sodium 141mmol/l	White blood cells 11.2 $X10^9$/L
Potassium 4.9mmol/l	Lymphocytes 2.9 $X10^9$/L
Chloride 100mmo/l	Granulocytes 7.5 $X10^9$/L
Bicarbonate 23mmol/l	Haematocrit 39.6%
Urea 5.6mmol/l	MCV 81.3%
Creatinine 0.7mg/dl	MCHC 26.1pg
Glucose 4.2mmol/l	Platelets 245$X10^9$/L

Key: MCV= Mean corpuscular volume, MCHC= Mean corpuscular haemoglobin concentration

the 4th child contradicts the commonest birth order reported by Afolabi et al from Ilorin and an earlier hypothesis of first-born child being more at risk of developing RRP because primigravid mothers are more likely to have a long second stage of labor thereby prolonging exposure of the child to HPV in the birth canal.[3,8] Although the socio-economic status of the caregiver in this case is a risk factor for acquiring sexually transmitted diseases such as HPV infection,[2] the absence of genital warts in the mother of our patient makes the mode of transmission of HPV quite unclear and at variance with earlier documentation.[2,4,6]

The clinical presentation of our patient was in consonance to report from another study and as documented in the literature.[3,5,6] The average duration of symptoms before diagnosis was 24months in our case and can be considered late presentation. The delayed presentation of our patient might have been due to inability of health workers at the PHC's to recognize the diagnosis of respiratory papillomatosis in addition to ignorance on the part of the parents, considering their socio-economic status. Socio-cultural predisposition, low literacy level, religious beliefs, self-medications, delayed referral from the general practitioners have been documented as reasons for late presentation in previous reports.[3,5,9] Experiencing frequent recurrence as observed in this case who has had three episodes in ten months is a pointer to the possibility of an aggressive type of RRP as previously documented.[2]

The management goals are to secure the airway, to improve and maintain an acceptable voice, and to facilitate remission while limiting morbidity and complications.[10] No cure has been identified for RRP and treatment is usually directed at relief of airway obstruction.[11] The primary treatment of RRP is surgical removal of the papilloma. Our patient had emergency tracheostomy to relieve the obstruction as well as video assisted direct laryngoscopy with surgical excision of the lesions and is currently stable. This conforms to the intervention that was reported by other studies[3,6] and is said to be reliable and affordable considering our resource constraint setting where CO_2 laser as an intervention practiced in the well-resourced countries is not yet available.[3] The key to excellent outcome in the management of respiratory papillomatosis remains early diagnosis, proper and adequate treatment and serial monitoring with follow-up also, importantly is the HPV vaccination so as to reduce HPV infection in women particularly during pregnancy as this is expected to reduce the transmission of HPV thereby reducing the chances of developing RRP.[11,12] Quadrivalent HPV vaccine has also been used in persons with active RRP and has been shown to significantly lower the frequency of RRP, the number of times a patient will need surgery as well as the possibility of spontaneous regression.[11,13,14]

Other adjuvant therapies have been used in small scale trials with variable outcomes of RRT. Some examples

include systemic and intralesional Bevacizumab (a humanized monoclonal antibody that inhibit vascular endothelial growth), Metformin (a biguanide with possibly anti-proliferative effects), Intralesional Cidofovir (an antiviral agent that interferes with viral replication by inhibiting viral DNA synthesis) and pegylated interferon.[11,15–17] These therapies are indicated in moderate-severe disease with the goal of reducting progression, severity, recurrence rate, or burden of disease. However, the systemic side effects associated with the aforementioned pharmacological therapies such as nephrotoxicity, lactic acidosis, oncogenic effects, bleeding, neutropenia as well as non-availability and high costs of therapy makes surgical intervention a more feasible option for patients in low-resource settings like ours.

4. Conclusion

Respiratory papillomatosis though rare, could be mis-diagnosed as childhood bronchial asthma. High index of suspicion is required to avoid delay in appropriate intervention. Counseling and follow up is required on a regular and long term basis.

5. Authors Contribution

All authors contributed to the completion of this work. The final manuscript was read and approved by all authors.

6. Ethical Considerations

Ethical approval to conduct the study was obtained from Usmanu Danfodiyo University Teaching Hospital (UDUTH) Health Research and Ethics Committee. Authors obtained permission from the caregivers to publish the clinical details of the patient.

7. Source of Funding

None.

8. Conflict of Interest

None.

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