Multiple Laryngeal Papillomatosis in an Adult Masquerading as a Laryngeal Malignancy: A Case Report

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Abstract

Multiple laryngeal papilloma is a disease common in childhood. It presents with papilloma involving the supraglottis and glottis with complaints of progressive hoarseness, stridor and dyspnea. Adult onset papilloma are usually solitary and mainly involve the glottis, with the main symptom being hoarseness. Multiple laryngeal papilloma in an adult is rare and has the potential of malignant transformation. The treatment of papillomatosis is surgical excision or ablation. Regular follow up is essential both to examine for recurrences as well as malignancy. This is a case report of an adult male with multiple laryngeal papillomatosis, treated surgically with microdebrider.

Introduction

Multiple laryngeal papilloma is characterized by the presence of multiple squamous papilloma in the larynx. This may affect any part of the respiratory tract but the larynx is the most common site. This is typically seen in the pediatric age group and is the most common benign neoplasm of the larynx in the pediatric population, hence it is termed as juvenile laryngeal papillomatosis. The human papilloma virus is associated with the etiology of this disease. Usual clinical presentation is seen with hoarseness, stridor and respiratory distress, depending on the site and extent of laryngeal involvement of the disease. Surgical excision is the treatment of choice for this condition, rarely a tracheostomy is required to secure the airway. Excision may be done with microlaryngoscopy using cold instruments, laser, electrocautery or microdebrider. A microdebrider is an effective tool for complete clearance of laryngeal lesions, provided

2013 Volume 6(1)
biopsy is taken for histopathological examination to rule out malignancy. Here we report a case of multiple laryngeal papilloma in an adult male, who presented with stridor and respiratory distress which was managed by tracheostomy followed by excision and clearance using a microdebrider.

**Case Report:** A 55 year old agriculturist presented to the emergency department of our hospital with breathing difficulty for 2 days. He also had a history of a hoarse voice for 4 years and noisy breathing since last three months. He gave a history of smoking beedi for last 30 years. His vitals were within normal limits. There was biphasic stridor. Voice was hoarse. There was no cyanosis. Oxygen saturation was 95% on air. Indirect laryngoscopy examination was done which showed a large exophytic mass lesion obstructing the glottic chink. A clinical diagnosis of laryngeal malignancy was made. He was explained the necessity for an alternate airway and a tracheostomy was performed after taking written informed consent (See right-hand picture). Direct laryngoscopy was done after establishing the airway, which revealed multiple papillomas involving both the true and false vocal cords, arytenoids, and extending to the anterior and posterior commissure completely obstructing the laryngeal airway. The largest of the papillomas was attached to the right false vocal cord. Biopsy was taken from multiple sites, as we had a clinical suspicion of malignancy. Histopathology of the biopsy showed stratified squamous epithelium with hyperkeratosis, acanthosis, papillomatosis with fibrovascular core, suggestive of squamous papilloma (See pictures below).

There was no evidence of malignancy among the tissue examined. Excision of the papilloma was done under microlaryngoscopy with complete clearance of the lesion using a microdebrider under general anesthesia (See video to the right). The patient withstood the procedure well. Postoperative period was uneventful. Histopathological examination of the specimen revealed squamous papilloma without any evidence of malignancy. Tracheostomy was closed after one week. There was no difficulty in decanulation. Patient was discharged two days after decanulation. He has been under regular follow up for the past two years with no recurrence of the lesion (See video to the right).

View YouTube Video of the surgery and the post-op appearance of the larynx: [https://youtu.be/ROU3CaC88uQ](https://youtu.be/ROU3CaC88uQ)
Discussion

Multiple laryngeal papillomatosis is characterized by recurrent squamous papilloma in the respiratory tract, hence the name recurrent respiratory papillomatosis (RRP) which has widely replaced the term juvenile laryngeal papillomatosis. This is predominantly seen in the larynx and is the most common benign neoplasm of the larynx in the pediatric population.\(^1\) It is mainly seen in children, but rarely can be seen in adults also. The causative organism of this disease is Human Papilloma Virus (HPV) Types 6 and 11. Babies born vaginally to mothers with genital warts have a high risk of developing laryngeal papilloma.\(^3\) They present with hoarseness, but more advanced cases present with stridor and respiratory distress.\(^2\) Other presentations: Chronic cough, choking, recurrent pneumonia, dyspnea, and dysphagia. Adults may present with a globus sensation.

Based on the age at onset, clinically there are two forms of RRP: Juvenile onset and adult onset.\(^3\) Juvenile onset RRP (JORRP) is seen most commonly between 2 and 4 years of age. There is no sex predilection. This is generally more aggressive than adult-onset RRP (AORRP) which is diagnosed most commonly between the ages of 20 and 40 years, has a slight male predilection, and is overall less common than JORRP.\(^2\)

RRP has a predilection for anatomic sites that are junctions between squamous and ciliated epithelium like limen vestibuli, the nasopharyngeal surface of the soft palate, the midzone of the laryngeal surface of the epiglottis, the upper and lower margins of the ventricle, the under surface of the vocal folds, the carina, and at bronchial spurs. Longstanding cases of laryngeal papillomatosis may lead to involvement of bronchopulmonary segments and lung parenchyma.\(^4\)

Multiple laryngeal papillomatosis may be misdiagnosed as asthma, croup, allergy, laryngitis, bronchitis or carcinoma. Even in our case we had made a clinical diagnosis of carcinoma, because of the age, history of chronic smoking, clinical presentation with hoarseness, stridor and respiratory distress, and an exophytic mass completing occluding the glottis. Tracheostomy was planned accordingly, and papilloma could be appreciated only after doing a thorough direct laryngoscopy examination. However we have no regrets for performing the tracheostomy in this patient, as the disease was well controlled and we could close the tracheostomy within one week after surgery.

Histologically, papilloma is composed of multiple fingerlike projections of nonkeratinized stratified squamous epithelium overlying a vascularized core of connective tissue stroma. Lesions rarely undergo malignant degeneration; however, the papilloma virus is oncogenic and varying degrees of atypia are common.\(^5\)

Techniques of RRP surgical resection have evolved remarkably in the last 30 years. Goals of treatment are assuring an adequate airway, improving voice and eradicating disease. Tracheostomy needs to be avoided as long as possible to prevent implantation of papilloma at the tracheostomy site. Endolaryngeal microsurgery is the treatment of choice. Papilloma can be excised using cold instruments, laser, electrocautery and microdebrider.\(^6\) Cold instrumentation is useful in adults. However, use of a microdebrider is a major development in the last few years and is safer and more accurate than laser. This can even remove tracheal lesions when used with a rigid endoscope. Pasquale, et al.\(^6\) did a prospective study of comparison of microdebrider versus CO2 laser for removal of recurrent respiratory papillomatosis.
in nineteen patients. They concluded that microdebrider is safe and more cost effective that CO₂ laser. El-Bitar and Zalzal in their retrospective study on the use of microdebrider and CO₂ laser in the management of recurrent respiratory papillomatosis, concluded that microdebrider is less consuming and user friendly than laser, as the disease warrants repeated procedures.⁷

The research is still on regarding the medical management of laryngeal papilloma. The drugs which are proved to be of some use in this regards are alpha Interferons, Indole 3-Carbinol, Antivirals like Cidofovir and retinoids like Isotretinoin. However none of these are proved useful when used alone.⁸

Conclusion

Juvenile laryngeal papillomatosis is known for recurrence after surgical excision and the patient may require multiple surgical procedures. The microdebrider is an effective tool for complete and precise clearance of papilloma. Alpha interferons, retinoids and antivirals like cidofovir are increasingly being used in the treatment of multiple laryngeal papilloma as adjunctive treatment to prevent recurrence. Malignant transformation is more common in adult onset laryngeal papilloma requiring histopathological examination of the specimen and regular follow up after excision.

References


