SOLITARY JUVENILE LARYNGEAL PAPILLOMA

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INTRODUCTION

Laryngeal papillomas are undoubtedly one of the most challenging benign conditions the laryngologist has to face. The pathogenesis as well as reasons for their often sudden disappearance is poorly understood. Many different modalities have been used in the management of patients with laryngeal papillomas, and the multitude of methods seems to reflect the inability of individual method to control the condition. Some contradictions may exist in the classification of laryngeal papillomas, as most authors differentiate between juvenile multiple papilloma & solitary adult papilloma-with the addition that juvenile papilloma may in some cases start in even adults.

Though McKenzie about a century ago, noted that the papilloma was the most common benign tumor of the larynx-here we report a rare case of solitary juvenile laryngeal papilloma which had unusual presentation and behaviour.

CASE REPORT

A 15 years old male was admitted with respiratory distress and history of hoarseness and difficulty in breathing for last 6-7 months. Emergency tracheostomy was done. The local examination revealed, proliferative pinkish mass

Fig. I : Preoperative photograph of the patient showing widening of laryngeal frame work.

Fig. II : C. E. C. T. ; Large heterogeneously enhancing mass completely filling the laryngeal inlet and causing splaying of hyoid bone.

Fig. III : C. E. C. T. ; Mass in the subglottis.

extending from oropharynx completely filling the laryngeal inlet; no normal laryngeal structures could be visualised, The whole laryngeal framework was widened and there was not cervical lymphadenopathy (Fig. I). Biopsy of the mass revealed it to be laryngeal papilloma. Fresh C. E. C. T. of the mass showed large heterogeneously enhancing mass in supraglottic, glottic and subglottic compartment extending from the level of hard palate to the cricoid cartilage and causing splaying of hyoid bone, displacement of both carotids and parapharyngeal spaces laterally (Fig. II & III). Since as per CT-SCAN, none of the normal endolaryngeal structures could be seen, therefore the patient was undertaken for surgery with the impression that we

Fig. IV : Peroperative photograph showing tumor being delivered in toto.

Fig. V : Postoperative specimen (inlet) and micro photograph of the specimen showing papillomatosis of epithelium overlying sparsely inflamed fibrovascular stroma (x40).

might have to sacrifice his larynx. Accordingly consent for total laryngectomy was taken. Midline laryngopharyngotomy was done after giving Sorenson's incision. The mass was found to be attached to both aryepiglottic folds, and pharyngoepiglottic folds; true & false cords of either side were visualised clearly, subglottic part of the mass was lying free in lumen. The mass was removed in toto (Fig. IV). Histopathology of the mass was benign laryngeal papilloma (Fig. V). Patient was decanulated two weeks after surgery, laryngoscopy done at intervals of four weeks revealed larynx to be normal. The patient is absolutely asymptomatic after three years of follow up.

The past history and medical record of the patient revealed that the patient had undergone direct laryngoscopic examination followed by lateral pharyngotomy about a year back. The mass was removed completely and the histopathology of the same was laryngeal papilloma. Patient was asymptomatic for two to three months only, after the surgery.

DISCUSSION

Papilloma is the most common benign neoplasm of the larynx. In New & Erich's series, papilloma accounted for 82 percent of true neoplasms, and in Holinger and Johnston's study, they accounted for 92 percent. Solitary juvenile papilloma are considered rare (Newmann et al 1980). Even in Lindburg, Oxlund and Elmond's (1986) series there was not even a single case of solitary juvenile papilloma.

There has been much debate over the cause of papillomas and although the issue remains unsettled, the evidence is overwhelmingly in support of viral etiology particularly the DNA human papilloma virus (Ulmann E.V. 1923, Geismann et al 1983). It has also been suggested that compromised immune function may predispose a patient to development of papillomas, some researches have suggested that papillomas may be related to the presence of maternal genitalwarts. However, maternal condylomas are not likely to be responsible for papilloma arising in adults. Nevertheless human papilloma virus type 6 has been found in juvenile and multiple adult papillomas as well as in condylomas, (Mounts P, Shah & Kashima H. 1982).

Historically papillomas have been divided into two groups - Juvenile onset and Adult onset. Despite the identical histological appearance of these groups, their clinical behaviours are markedly different. Papillomas of the juvenile onset group, often manifest as multiple lesions, which may recur rapidly or spread diffusely following excision (Strong, Vaughan et al, 1982). Adults onset variety is much less aggressive often occurring as a solitary lesions with a lesser propensity to recur after removal. Kleinsasser and Cruz (1973) believe adult onset group can be further subdivided into keratinizing lesions, which are solitary nonrecurrent and possibly precancerous and non keratinizing lesion which tend to behave more like the juvenile onset papilloma. Lindburg, Oster, Oxulund and Elbrond (1986) classified the papilloma into four groups.

- I. Juvenile multiple papillomas
- II. Juvenile solitary papillomas
- III. Adult multiple papillomas
- IV. Adult solitary papillomas.

Caper, Bailey & Michaels (1992) classified them into three subgroups. Solitary which is a single papilloma, including large confluent growths which appeared as one lesion, multiple that is two or more distinct lesion in the larynx,

separated by clinically normal epithelium. Florid, which are those cases where there is involvement of every part of the larynx, with an aggressive mode of growth necessitating frequent removal over a lengthy period.

In adult onset, majority of patients are male, in contrast to juvenile onset group in which most of the cases are female. (Quiney, et al & Irwin et al) Female to male ratio in juvenile variety is 2:1 in contrast to 2:1 male predominance in adult type. Vocal cord is the most frequent site of the disease in the larynx, and both vocal cords are equally susceptible. But any part of the larynx may be affected like suprahoid or subhyoid supraglottis or subglottis. Subglottis is least likely site to be involved. The lesion in severe cases may extend to oropharynx, hypopharynx or trachea. Histologically, papillomas appear as benign squamous lesion with and arborizing core of highly vascular connective tissue surrounded by an outerlayer of nonkeratinizing squamous epithelium. Clinically, the papillomas present as pedunculated exophytic mass with a warty appearance that varies from pink to red (non-keratinized) to white (keratinized). The large number of treatment modalities advocated for this disease proves to the inadequacy of most of them. Simple endoscopic removal has been the most frequently employed treatment, especially in those cases in which the papilloma occurred as solitary lesion. Other recent therapies include laser excision, radiation therapy, ultrasound, immunomodulation (BCG, autogenous vaccines, interferon), electrocautery cryosurgery, chemotherapeutic drugs and topical application (acids, podophyllin, alcohol and estrogens). The majority of more elaborate modalities have been directed at treatment of the juvenile onset papilloma because of its more refractory nature. In managing extensive cases, care must be taken to prevent damage to normal laryngeal structure, to avoid post operative structure & webbing. Occasionally, laryngectomy has been done for the severe and uncontrolled disease (Robbins and Howard 1983). Carcinomatous changes in respiratory tract papillomas in children is rare but squamous carcinoma of the larynx may develop following radiotherapy of laryngeal juvenile papillomatosis. Some authors doubt, however, whether malignant transformation of adult laryngeal papillomas does

occur (Batsakis, 1979). Papillomas that grow or recur rapidly, especially after therapy may show varying degrees of maturation disturbance within the epithelium.

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