
Recurrent laryngeal papillomatosis with bronchopulmonary spread in a 70-year-old man

Mohammad Hossein Rahimi RAD¹, Effat ALIZADEH², Behrouz ILKHANIZADEH³

¹ Department of Respiratory Medicine, Urmia University of Medical Sciences, Urmia, West Azerbaijan, Iran,

² Department of Biology, Faculty of Science, Shahid-Beheshti University, Tehran, Iran,

³ Department of Pathology, Urmia University of Medical Sciences, Urmia, West Azerbaijan, Iran.

ÖZET

Yetmiş yaşında erkek hastada bronkopulmoner yayılım olan rekürren larengeal papillomatosis olgusu

Larenkste siğil benzeri oluşumlarla karakterize olan rekürren larengeal papillomatosis (RRLP) çocuklarda ve genç erişkinlerde görülen nadir benign bir hastalıktır ve ileri yaşta erişkinlerde az sayıda olgu bildirilmiştir. Solunum yolu boyunca RRLP'nin yayılımı nadiren ortaya çıkar ve distal bronşlar, bronşiyoller ve akciğer parankiminin tutulumu oldukça nadirdir. Önceden iki kez cerrahi girişim yapılmış olan 70 yaşındaki erkek hastada trakeobronşiyal ve pulmoner yayılımı olan RRLP olgusunu bildiriyoruz. Erişkinlerde bu hastalık son derece nadir olmasına rağmen doğru tanı, klinik, radyolojik ve patolojik özelliklerin karakteristik kombinasyonu ile düşünülebilir.

Anahtar Kelimeler: Rekürren larengeal papillomatosis, rekürren respiratuar papillomatosis, pulmoner yayılım, papilloma virüs, erişkin, genç erişkin.

SUMMARY

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Mohammad Hossein Rahimi RAD¹, Effat ALIZADEH², Behrouz ILKHANIZADEH³

¹ Department of Respiratory Medicine, Urmia University of Medical Sciences, Urmia, West Azerbaijan, Iran,

² Department of Biology, Faculty of Science, Shahid-Beheshti University, Tehran, Iran,

³ Department of Pathology, Urmia University of Medical Sciences, Urmia, West Azerbaijan, Iran.

Yazışma Adresi (Address for Correspondence):

Mohammad Hossein Rahimi RAD, MD, Bronchoscopy Unit, Imam-Khomeini Hospital, Urmia, West Azerbaijan, Iran 81351 URMIA - IRAN

e-mail: mohamadrahimirad@gmail.com

Recurrent laryngeal papillomatosis (RRLP) which is characterized by wart like growths in the larynx is a rare benign disease seen in children and young adults and a few cases are reported in old aged adults. The spread of RRLP throughout the respiratory tract occurs rarely; and involvement of the distal bronchi, bronchioles, and lung parenchyma is very rare. We report a case of tracheobronchial and pulmonary spread of RRLP in a 70-year-old man after two previous surgeries. Despite the rarity of this disease in adults, the correct diagnosis may be suggested by a characteristic combination of clinical, radiographic, and pathologic features.

Key Words: *Recurrent laryngeal papillomatosis, recurrent respiratory papillomatosis, pulmonary spread, papilloma virus, adult, juvenile.*

Recurrent laryngeal papillomatosis (RRLP) is a rare benign disease seen in children and a few cases are reported in adults (1,2). Although RRLP is histologically a benign proliferation of stratified squamous epithelium and is usually limited to the larynx, but spread to tracheobronchial tree occurs in 2% to 29% and pulmonary parenchymal in less than 1% to 7% (1). Adult onset RRLP is usually associated with solitary lesions. These lesions do not spread as readily as juvenile forms do and recurrence rate is low after removal (1,3). In the present report, we report to tracheobronchopulmonary spread of RRLP in a 70-year-old male patient.

CASE REPORT

On July 2006, a 70-year-old man presented with chief complaint of cough and hoarseness. He had history of endoscopic operation for laryngeal nodule in 1996 and 1999. Histopathology of laryngeal biopsy in 1999 was reported papilloma with stratified squamous epithelium. On physical examination there was hoarse voice, without any mass in neck, trachea was in midline position. There were crackles with wheeze in lower zone of left hemithorax. Chest X-ray showed collapse and alveolar infiltration in left lower lobe (Figure 1). CT scan of chest revealed nodules in trachea in several slices (Figure 2), nodule in left lower lobe bronchus and fine nodules in left and right lower lobe (Figure 3). Fiberoptic bronchoscopy showed vegetations in left false vocal cord and anterior commissure clusters of grape like vegetations in subglottis, distal trachea, left main and lower lobe bronchi. Histopathology of forceps biopsy of left lower lobe vegetations showed fingerlike projections of



Figure 1. Chest X-ray showed collapse and alveolar infiltration in left lower lobe.

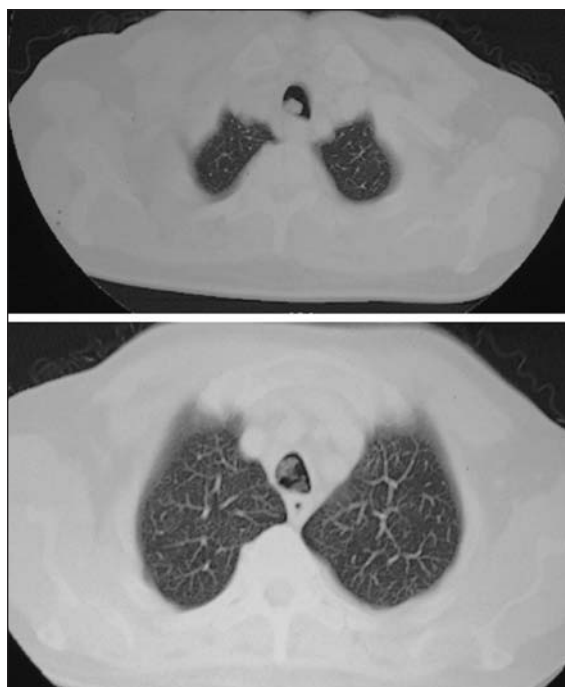


Figure 2. Axial CT slices demonstrate tracheal papillomas attached to the tracheal posterior and lateral walls.

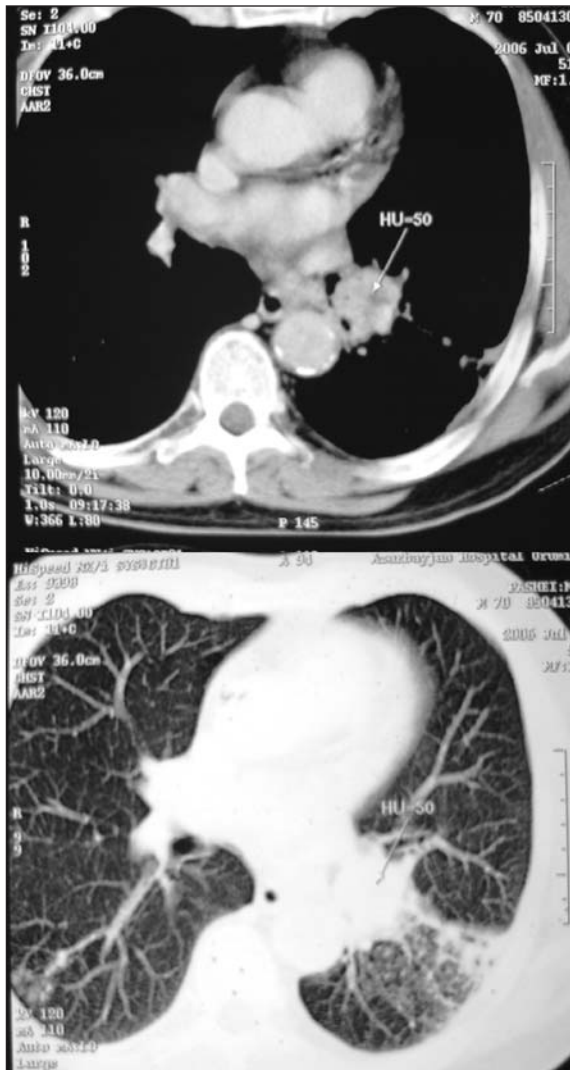


Figure 3. Papilloma in left bronchus, and multiple tiny nodules with a centrilobular distribution in right and left lung.

stratified squamous and ciliated columnar epithelium with a fibrovascular core compatible with papillomatosis (Figure 4).

DISCUSSION

RLP is a relatively rare disease but is still the most common pediatric neoplasm found in the larynx. The etiology of the disease is the human papilloma virus (HPV). The most common types of the virus that have been identified are HPV types 6 and 11 (4). Other less common presenting types include types 16 and 18; these have been associated more closely with malignant transformation (5,6).

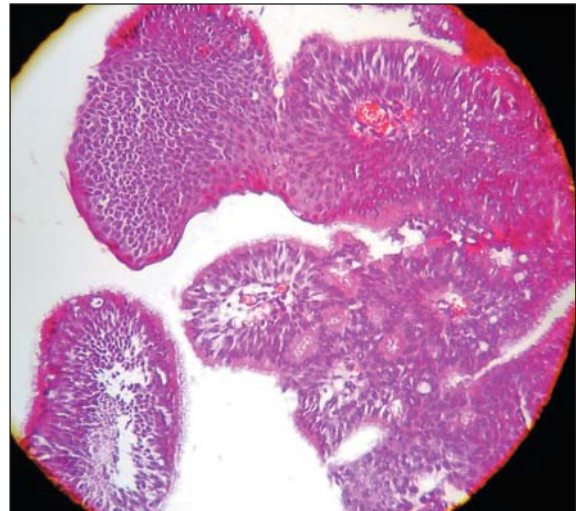


Figure 4. Photomicrograph of histopathologic bronchial mucosa specimen revealing a papilloma lined with mature squamous and ciliated epithelium, consistent papillomatosis tracheobronchial tree (HE, x50).

RLP is classified into juvenile and adult forms. Juvenile form presents as multiple lesions that are unpredictable in their response to treatment and tend to have a high rate of recurrence. In adults, RLP has a male predominance, are most commonly solitary, show a greater degree of inflammatory reaction, do not tend to spread and recur less frequently than juvenile form (3,4). The case is an old adult with recurrences, and distal tracheobronchial and pulmonary spread an uncommon finding in adults. Several possibilities for distal spread of RLP have been hypothesized: contiguous extension of papillomas, diffuse viral contamination, multicentric origin of papillomas and aerial dissemination of fragments (7). Tracheostomy and surgical manipulation are thought to induce spread of papillomas to the distal respiratory tree as appears to have occurred in the present case with two previous laryngeal surgeries. Formal tracheostomy should be avoided if at all possible in these patients (8).

The characteristic of RLP in children is the triad of progressive hoarseness, stridor and respiratory distress. In adult patients, hoarseness is the most common symptom as was in this patient. When it is extensive RLP may cause respiratory difficulty and even death (4). There are reports of upper airway obstruction misdiagnosed as asthma. Carroll et al. reported a 21 year old wo-

man presented with respiratory collapse to emergency department (9). Grobbelaar et al. reported three cases with pulmonary hypertension secondary to juvenile RLP (10).

The papilloma appears as an exophytic, branching, pedunculated or sessile mass, single or multiple, most frequently located on vocal folds, ventricular folds, the subglottis and laryngeal surface of the epiglottis. Histologically, papillomas appear as fingerlike projections of stratified squamous epithelium with a fibrovascular core. Basal cell hyperplasia and large vacuolated epithelial cells with a clear cytoplasm (koilocytosis) are typical. When RLP extended into the subglottis or tracheobronchial tree, the epithelium can be squamous or ciliated columnar epithelium (11). In the present case biopsy from larynx in 1999 was squamous papilloma, and biopsy from left lower lobe papilloma in 2006 showed both squamous and ciliated columnar epithelium.

Many methods of treating RLP including antibiotic administration, surgery, radiotherapy, autogenous vaccination, and chemotherapy, have generally been unsuccessful. However, some recent reports have stated that interferon alpha and antiviral agents such as cidofovir show promising results in the treatment of RLP (12-14).

RRLP is known to undergo malignant transformation. Spontaneous malignant degeneration in nonirradiated RLP has been reported in both the laryngotracheal and bronchioalveolar areas, the reported incidence being 2% to 3% and occurs within 10 years of diagnosis (3,7). It is thought to be associated with extrinsic factors such as smoking and radiation therapy. However, some cases have been reported in patients without this risk factors (15).

Conclusion: We described a case of RLP with tracheobronchial and pulmonary spread in an old age adult which is unusual. RLP should be considered as a differential diagnosis in tracheobronchopulmonary lesions especially in the patient with known laryngeal papillomas. This is particularly true if the patient has had a tracheostomy or surgery for papilloma. When RLP extended into tracheobronchial tree, the epithelium can be ciliated columnar epithelium or squamous.

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