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## CASE REPORT

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# Laryngeal Papillomatosis with Pulmonary Spread: Case Report and Review

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### ABSTRACT

*Laryngeal papillomatosis is the most common benign tumour that affects the larynx in children. It is well known for its strong tendency to recur. Spread of the disease to the lung parenchyma is rare. There have been only a few reports in the literature that have addressed the radiological features of laryngeal papillomatosis. In this article, we present a case of laryngeal papillomatosis in a Chinese girl that involved distal pulmonary spread and recurrence, and we review the available literature on the subject.*

*Key Words:* Child; Laryngeal neoplasms; Neoplasm recurrence; Papilloma/pathology; Papillomavirus, human

### INTRODUCTION

Laryngeal papillomatosis is the most common benign tumour that affects the larynx during childhood. The disease is well known for its strong tendency to recur, although spread of the disease to the lung parenchyma is a rare event. There have been only a few published reports in the literature that have addressed the radiological features of laryngeal papillomatosis. In this article, we present a case of laryngeal papillomatosis in a Chinese girl that involved distal pulmonary spread and recurrence. We discuss the chest X-ray and computed tomography (CT) findings at presentation and follow-up, and we review the available literature on the subject.

### CASE REPORT

A 9-year-old girl who had been born by vaginal delivery presented to the Queen Elizabeth Hospital in the autumn of 2004 with a respiratory tract infection. She had a history of hoarseness that had started at age 5 months.

The patient had been initially treated conservatively in a hospital in mainland China until age 18 months, when she first developed respiratory tract infection, which

was complicated by airway obstruction. Tracheostomy was thus performed. She then moved to Hong Kong and continued her treatment at the Queen Elizabeth Hospital in August 1997. Laryngoscopy at first presentation to our hospital showed multiple nodular growths at the larynx, as well as airway narrowing. Tissue examination showed squamous cell papillomas. Human papillomavirus was subsequently isolated, and the diagnosis of papillomatosis was made. Laryngeal papillomatosis recurred despite repeated laryngoscopic removal of papillomas, carbon dioxide laser treatment, and interferon therapy.

Chest X-rays at first presentation revealed clear lungs. Subsequent X-rays showed multiple solid and cavitated nodules in both lung fields, which suggested that papillomatosis had spread to the lung parenchyma (Figure 1a). The CT scans demonstrated multiple small nodules along the larynx, trachea, and bronchi, which were the cause of airway narrowing (Figure 1b). There were also multiple nodules — both solid and cavitated — in both lung fields (Figure 1c). The size of the nodules ranged from 2 mm to 2 cm. The walls were mostly thin and smooth. Histological examination of the pulmonary lesion was not performed.

At age 8 years, X-rays and CT scans revealed a progressive course of disease (Figure 2). An increased number of nodules was noted, as well as evidence that the disease had spread. Follow-up chest X-ray and CT revealed infection and a collapsed lung segment of the

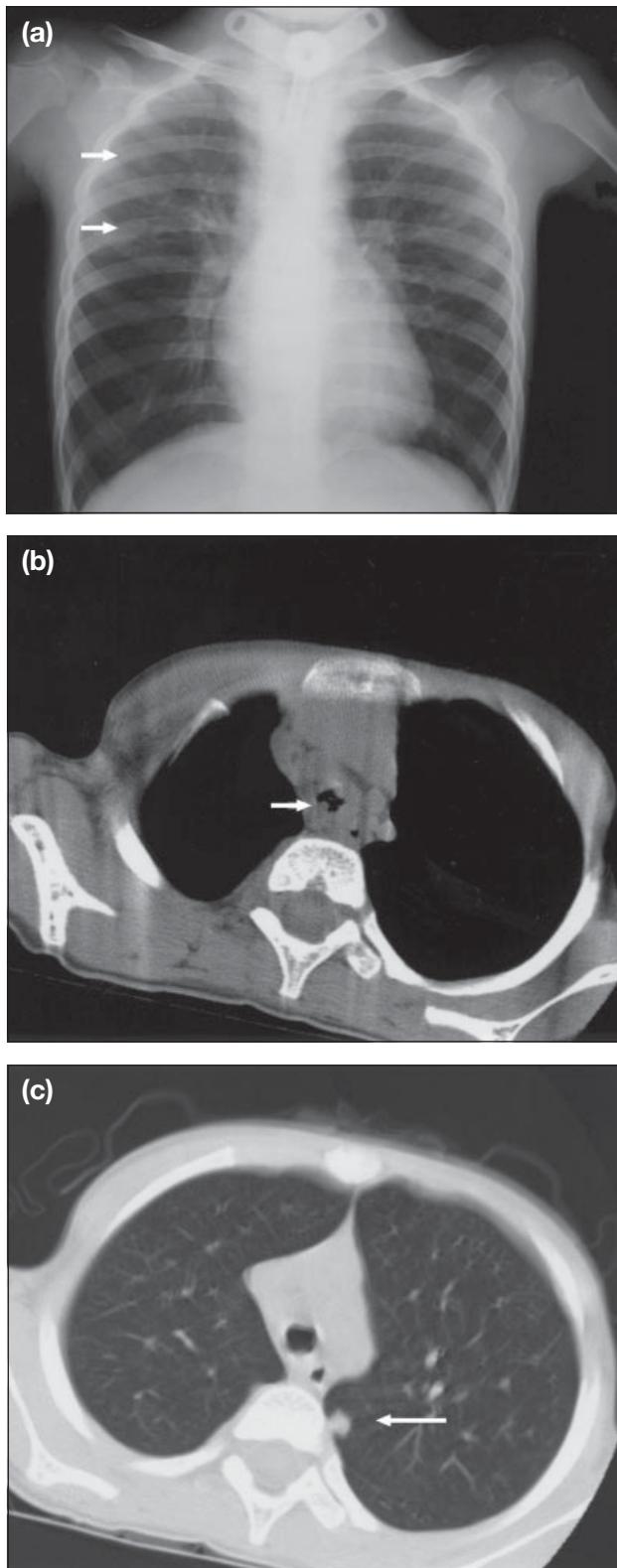
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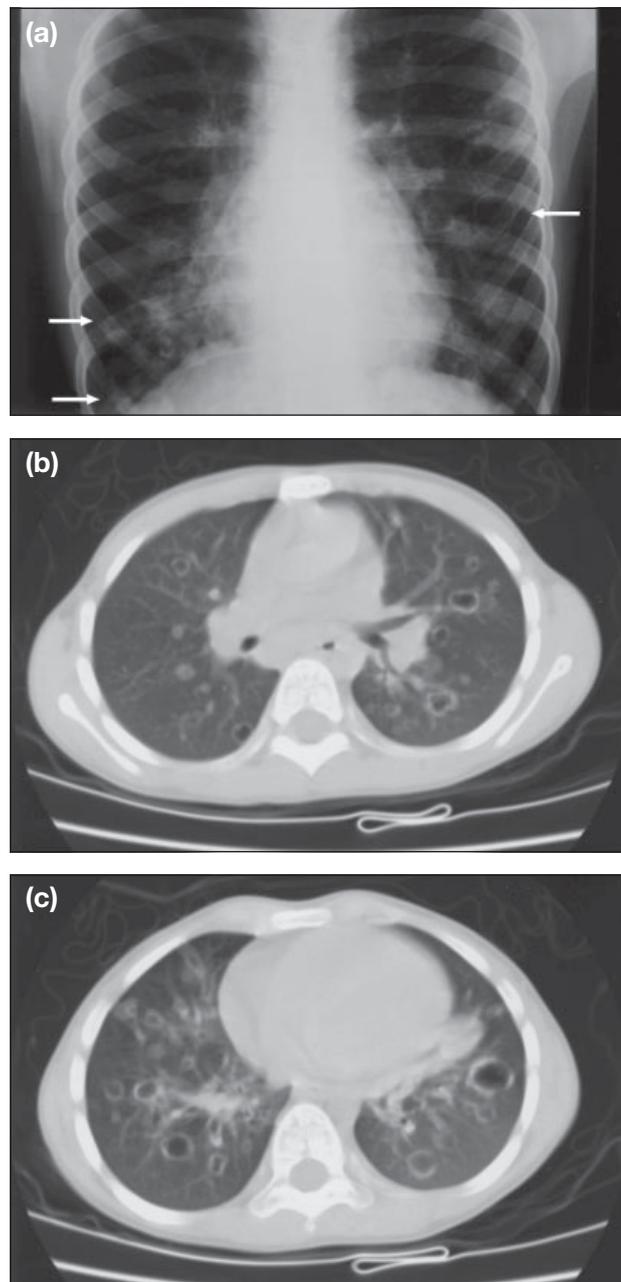
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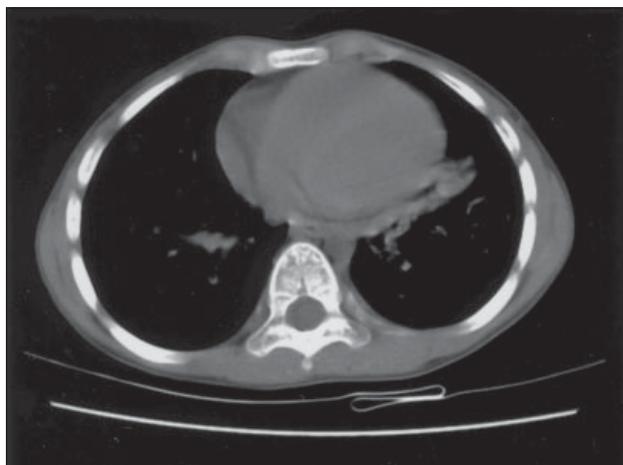
**Figure 1.** Radiological findings at age 3 years: (a) chest X-ray showing a few nodules (arrows) due to laryngeal papillomatosis in both lung fields (tracheostomy tube is also visible); (b) axial computed tomogram of the thorax (soft-tissue window) showing small soft-tissue nodular lesions and irregular wall thickening in the trachea (arrow); and (c) axial computed tomogram of the thorax (lung window) showing nodule in the left lung (arrow).



**Figure 2.** Radiological findings at age 8 years: (a) chest X-ray showing an increased number of nodules (arrows); and (b) and (c) computed tomograms showing more extensive disease, and solid and cavitated nodules with thin smooth walls in both lung fields.

lower lobe of the left lung (Figure 3). Despite progressively worsening radiological findings, the patient showed no signs of respiratory distress. She had only mild tachypnoea and cough during episodes of infection.

The patient continues to undergo regular surveillance and follow-up. Laryngoscopic tissue examination of the papillomas is conducted at least annually to monitor any malignant changes. Antibiotic treatment is given whenever she presents with respiratory infection.



**Figure 3.** Computed tomogram showing atelectasis in the left lung base.

## DISCUSSION

Recurrent respiratory papillomatosis is the most common laryngeal tumour in children.<sup>1</sup> Its former name was juvenile laryngeal papillomatosis because this disease involves not only the larynx, but also other sites of the respiratory tract. In addition, this disease can occur in adults.<sup>2,3</sup>

The majority of cases present before the age of 5 years, and up to 20% of cases occur in infancy.<sup>1,2</sup> Laryngeal involvement was found to be nearly 100% in several large studies.<sup>4,5</sup> The trachea is involved in 3% to 26% of cases, whereas pulmonary involvement is rare, occurring in about 1% to 3% of cases.<sup>4,6</sup> For juvenile-onset recurrent respiratory papillomatosis, the sex ratio is roughly equal.<sup>6,7</sup> However, pulmonary extension appears to be twice as common in males than in females.<sup>8</sup>

The causative agent of recurrent respiratory papillomatosis is the human papillomavirus, specifically types 6 and 11.<sup>3</sup> The virus may be contracted during delivery by vertical transmission,<sup>4,9</sup> which may probably be the case in the patient described in our case report.

Patients may present with symptoms such as hoarseness (as in our case), chronic coughing, dyspnoea, wheezing, and haemoptysis. More limited disease can be asymptomatic. Extensive disease may produce both obstructive and restrictive lung function.

The chest X-ray may show narrowed upper airways, with a clear chest if there is no pulmonary involvement. However, when pulmonary involvement is present, multiple solid and cavitated nodules of various sizes can be seen.<sup>1</sup> The cavitated lesions may have a thin or thick

wall. The lesions can be widely scattered or subpleural. Predominantly, they are found at the posterior aspect of the lower lobes. Sometimes, associated findings, such as bronchiectasis, atelectasis, and secondary infection, are noted.<sup>1</sup>

Differential considerations of multiple cavitated nodules in children include infective causes, such as fungal infection or septic embolism, but these patients usually appear ill and may be immunocompromised. Other possible diagnoses are bronchiectasis (but patients will have a predisposing factor such as easy aspiration or symptoms of cystic fibrosis) and adenomatoid malformation (but this should be present since birth). The presence of both upper airway papillomas and pulmonary cavitated nodular lesions, together with the absence of other associated disease or toxic symptoms, means that the pulmonary cavitated nodules may be due to distal spread from the laryngeal papillomas.

Examinations using CT can show similar findings and are helpful in assessing the degree of upper-airway narrowing. The lesions can be sessile or pedunculated soft-tissue masses, or they can have asymmetrical wall thickening.<sup>1</sup> Furthermore, CT can be used to assess the extent of the pulmonary nodules and any associated complications. The subsequent CT scan of the patient in our case showed atelectasis and infection in addition to multiple lung nodules.

Recurrent respiratory papillomatosis sometimes regresses spontaneously — particularly the limited juvenile-onset type.<sup>3</sup> If there is pulmonary involvement, however, the disease often runs a protracted course that requires multiple endolaryngeal procedures to maintain a sufficient airway.<sup>10</sup> Theories of how distal pulmonary spread occurs include aerial dissemination,<sup>11</sup> direct sequential spread of virus down the airway,<sup>12</sup> and spread from a multicentric origin.<sup>13</sup> The theory of aerial dissemination from a proximal site is more widely accepted than the others and is probably facilitated by endoscopic interventions.<sup>12,14</sup> The patient in our case underwent tracheostomy and repeated endoscopic resection, which might have been associated with the frequent recurrence and distal spread of the disease.

Malignant transformation to squamous cell carcinoma has been reported in both adult and juvenile cases.<sup>10</sup> Therefore, continuous surveillance of patients is necessary. The patient in this case report has already

been followed up in our hospital for 7 years. Recent tissue examination of removed papillomas have shown moderate dysplasia. Continuous follow-up by imaging studies will help to detect any malignant transformation, progression of disease, or complication.

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