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Multidimensional Approaches of A Tracheal Papilomatosis Clinical Case

KEYWORDS: tracheal papillomatosis, mechanical obstruction, squamous papilloma

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ABSTRACT *Tracheal papillomatosis is a rare benign tumor in adults, being more common in children, in whose etiology the human papilloma virus infection (HPV) is incriminated. We present the case of a 46-year-old woman, initially treated for asthma, to whom the wheezing, stridor and the inspiratory dyspnea were increasing. Due to the risk of asphyxia, we underwent emergency fibrobronchoscopy which showed tracheal vegetation formations with extension up to approximately 2-3 cm above the tracheal spur which was obstructing the trachea almost totally. The last histopathological examination showed squamous papilloma. The bronchoscopy desobstructive treatment - electroresection and electrocautery - offered a real benefit. It improved the quality of life and the imagistic examinations were in normal limits. There is need to follow this treatment due to the risk of recurrence or malignant transformation. This case report highlights the above pitfalls and looks into the current management of benign tracheal tumours.*

Tracheal papillomatosis is a benign neoplastic condition that involves the trachea, being rare in adults and more common in children, in whose etiology the human papilloma virus infection (HPV) is incriminated. More frequent ethiological agents for tracheal papillomatosis are HPV-type 6 and HPV-type 11; (Shibuya H, Kutomi T, Kujime K& et al. 2008; Harris K &Chalhoub M 2011), but there are also HPV-type 16 and HPV-type 18 with a poorer prognostic. (Valentino J, Brame CB, StudtmannKE & et al. 2002). In the bronchial tree there is a continuous papillomatous growth of the bronchial epithelium in response to the infection with human papilloma virus (HPV). (Shibuya H, Kutomi T, Kujime K& et al. 2008; Harris K &Chalhoub M 2011)

Tracheal papillomatosis is a rare disease with the incidence between 4-12 cases per 1,000,000 person/year. (Bishai D, Kashima H & Shah K 2000). There are two forms of manifestation: juvenile onset and adult onset. During the juvenile onset, with the peak with a mean age onset at 3.8 year, the transmission of HPV can occur after cesarean delivery which is known to be the main mechanism of transmission in juvenile onset or, can be due to a vertical transmission of the virus after vaginal delivery of an infected mother. (Harris K &Chalhoub M 2011; Kosko JR &Derkay CS 1996) Referring to adult onset of tracheal papilomatosis, it affects both sex with a male and female ratio 4:1, with onset between 3-4 decades of life and remains unclear whether sexual contacts or latent viral activated by risk factors such as gastroesophageal reflux, may be the main ways of HPV transmission in adult onset of respiratory papillomatosis. (Harris K &Chalhoub M 2011).

In the natural history of tracheal papilomatosis, some cases could regress spontaneously, but there is the risk to recur even after years of regression. During pregnancy, for example, the lesions are prone to aggravation (Xue Q, Wang H &Wang J 2010). The risk factors for malignant transformation that are incriminated are: smoking, irradiation, cytotoxic drugs, p53 mutation, HPV11 infection, high severity score or highactivity of 2′, 5′-oligoadenylate synthetase (RadyPL, Schnadig VJ, Weiss RL et al 1998; Gerein V, Rastorguev E, Gerein J et al 2004).

The diagnosis and the therapy of this disease is challenging due to the absence of specific clinical manifestations, recurrent nature, complications and also due to the risk of malignant transformation (Xue Q, Wang H &Wang J 2010). The clinical symptoms are hoarseness, wheeze, cough, chronic dyspnea, choking, syncope or voice change. These symptoms may be typically for the obstruction of the upper airways. The stridor may be audible on the auscultation of the chest. The physical exam is not usually helpful in putting the diagnosis of this condition. The chest radiography is usually normal. Because of its nonspecific clinical manifestations, tracheal papillomatosis is easily mistaken for asthma, acute laryngitis, upper respiratory infection or bronchitis, but asthma therapies are inefficient in this case. Presenting non-specific symptoms may lead to delayed or incorrect diagnosis, but the risk of asphyxia due to mechanical obstruction makes the clinician suspect a tracheal tumor.

Endoscopy should be performed as soon as possible in suspected patients, which would enable the diagnosis to be established early and correctly. Fibrobronchoscopy is the main method to put a final diagnosis, completed by a histological exam and HPV DNA by PCR testing. (Harris K &Chalhoub M 2011; Xue Q, Wang H &Wang J 2010)

The main therapy goals of tracheal papillomatosis are curing lesions and preventing recurrence. Surgical removal on endoscope is the fundamental treatment, and the most extensively used approaches in recent years are laser ablation and microdebrider removal. Other available therapies include electrocautery and cryotherapy. (Xue Q, Wang H &Wang J 2010)

Adjuvant medical treatment is needed especially for recurrent cases (respiratory reccurentpapilomatosis - RRP), and refers to antiviral and immunoregulation drugs. Other available adjuvant drugs include anti-reflux drugs, mitomycinC, cyclooxygenase2 inhibitors, retinoids, zinc and indole-3-carbinol, interferon –α.The commonly used antiviral drugs include cidofovir, ribavirin, acyclovir and ganciclovir. (KimberlinDW(2004);Bielecki I, Mniszek J&Cofała M 2009)

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Neodymium-doped yttrium aluminium garnet (Nd:YAG) laser has also been successfully used in RRP. (Janda P, Leunig A, Sroka R & et al 2004)

The research and development of a multivalent HPV vaccine has progressed rapidly in recent years, remaining unclear the mechanisms of action, effectiveness and future clinical utility. (Chesson HW, Forhan SE, Gottlieb SL& et al 2008)

In the light of these data we present a case of a 46-year-old woman who came to our department and who was initially treated for asthma, the wheezing, the stridor and the inspiratory dyspnea were increasing. These symptoms gradually worsened over the past 2 months, by the time she came to our department the symptoms were already acute. She was a non-smoker, known with gastroesophageal reflux and arterial hypertension and had no history of radiotherapy to the trachea. The physical examination was normal, except for the inspiratory and the expiratory stridor. Her routine laboratory tests showed high values of gycemia. The pulmonary function test could not be performed in that moment due to her critical clinical status. The chest X-ray appearance was without modifications (Figure 1), but because of the risk of asphyxia, she underwent emergency fibrobronchoscopy which showed tracheal vegetation formations with extension up to approximately 2-3 cm above the tracheal spur which was obstructing the trachea almost totally. (Figure 2 (A) and 2(B)) The final histopathological examination showed squamous papilloma. The focal epithelium is kept isolated and the respiratory type becomes transitional. Koilocitoza images are also presented. Immunohistochemistry Ki 67 indicates a high mitotic index which requires careful monitoring of the case. Bronchoscopy desobstructive treatment - electroresection and electrocautery - offered a real benefit, a chest computer tomography scan was subsequently performed and it was within normal limits. (Figure 3)

The evolution after two months was good and the quality of life improved; she also underwent fibrobronchoscopy and the trachea was without any lesions (Figure 4).The case requires careful attention because of the risk of recurrence or malignant transformation.

Discussion

Tracheal papillomatosis was reported in children and adults, with the onset during adulthood, more common among men and in the third decade of life (Ogata-Suetsugu S, Izumi M, Takayama K &et al. 2011). Our case was a woman with onset in the fourth decade of life, so with the onset later than the cases described in the literature.

Most papillomas are found in the larynx, just 5% of cases had distal involvement of the trachea, so our case report is rare due to his location in the trachea.

Malignant degeneration into squamous cell carcinoma occurs in 3% - 5% of cases and more often in patients with a history of smoking or radiation therapy (Cook JR, Hill DA, Humphrey PA& et al. 2000). In our case there are no risk factors for malignant transformation. The route of HPV transmission remains unclear. She has a risk factor for latent viral activation which is the gastroesophageal reflux.

In our patient, the success was represented by the removal of the tumor using electroresection and electrocautery, throught fibrobronchoscopy. After 5 years of follow up there are no symptoms or local signs of recurrence, actually there is no need for adjuvant medical therapies. However, a clinical follow-up is necessary to confirm the absence of recurrence or malignant transformation.

The main limit of case management is the absence of polymerase chain reaction (PCR) for the detection HPV type, in order to prove the ethiological agent of tracheal papillomatosis.

This case report highlights the above pitfalls and looks into current management of benign tracheal tumours.

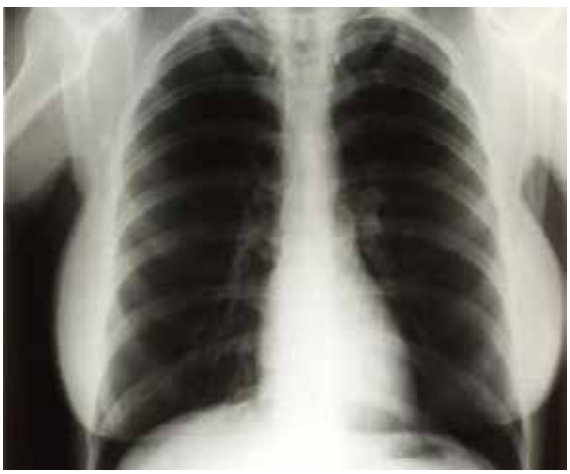

Figure 1. Normal Chest X-ray

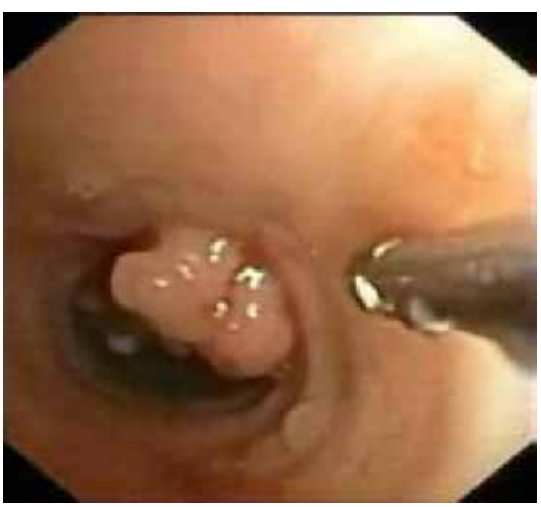

Figure 2 (A)

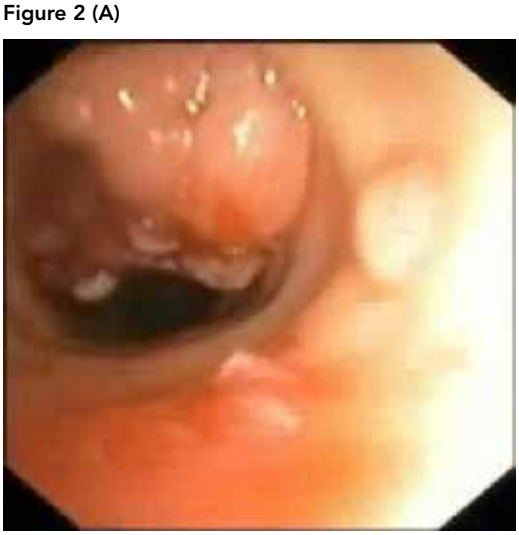

Figure 2 (B)

Figure 2 (A) and 2 (B) Fibrobronchoscopy imaging - tracheal vegetation formations with extension up to approximately 2-3 cm above the tracheal spur which was obstructing the trachea almost totally

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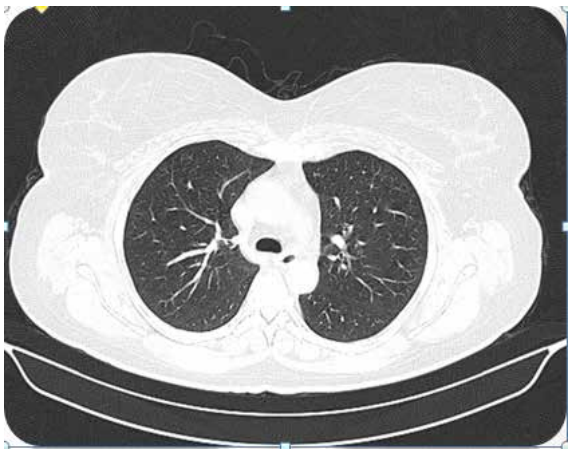

Figure 3 – Normal thoracic CT scan

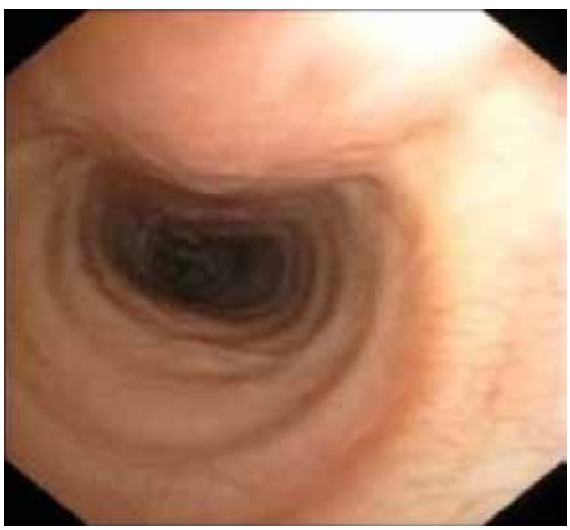

Figure 4 - Fibrobronchoscopy imaging – trachea without any lesions (2 months after electroresection and electro-cautery)

REFERENCE Shibuya H, Kutomi T, Kujime K, Hara K and Hisada T (2008). An Adult Case of Multiple Squamous Papillomas of the Trachea Associated with Human Papilloma Virus Type 6. Inter Med 47: 1535-1538. | Harris K and Chalhoub M (2011). Tracheal papillomatosis: what do we know so far?Chronic Respiratory Disease 8(4) 233–235. | Valentino J, Brame CB, Studtmann KE, and Manaligod JM (2002).Primary tracheal papillomatosis presenting as reactive airway disease.Otolaryngol Head Neck Surg 126:79–80. | Bishai D, Kashima H, Shah K (2000) The cost of juvenile-onset recurrent respiratorypapillomatosis. Arch Otolaryngol Head NeckSurg 126:935–939. | Kosko JR and DerkayCS(1996). Role of cesarean section in prevention of recurrent respiratory papillomatosis—is there one? Int J PediatrOtorhinolaryngol 35(1): 31–38. | Xue Q, Wang H and Wang J (2010). Recurrent respiratory papillomatosis: an overview. Eur J ClinMicrobiol Infect Dis 29:1051–1054. | Rady PL, Schnadig VJ, Weiss RL et al (1998) Malignant transformation of recurrent respiratory papillomatosis associated with integrated human papillomavirus type 11 DNA and mutation of p53. Laryngoscope 108:735–740. | Gerein V, Rastorguev E, Gerein J et al (2004) 2′, 5′-Oligoadenylate synthetase activity analysis and human papilloma virus typingas prognostic factors in patients with recurrent respiratory papillomatosis. J LaryngolOtol 118:750–756. | KimberlinDW(2004) Current status of antiviral therapy for juvenileonset recurrent respiratory papillomatosis. Antiviral Res 63:141–151. | Bielecki I, Mniszek J, Cofała M (2009) Intralesional injection of cidofovir for recurrent respiratory papillomatosis in children.Int J PediatrOtorhinolaryngol 73:681–684. | Janda P, Leunig A, Sroka R, et al (2004). Preliminary report of endolaryngeal and endotracheal laser surgery of juvenile-onset recurrent respiratory papillomatosis by Nd:YAG laser and a new fiber guidance instrument. Otolaryngol Head Neck Surg 131(1): 44–49. | Chesson HW, Forhan SE, Gottlieb SL et al (2008).The potential health and economic benefits of preventing recurrent respiratory papillomatosis through quadrivalent human papillomavirus vaccination. Vaccine 26:4513–4518. | Ogata-Suetsugu S, Izumi M, Takayama K et al. (2011). A Case of Multiple Squamous Cell Papillomas of the Trachea.Ann ThoracCardiovascSurg 17: 212–214. | Cook JR, Hill DA, Humphrey PA, Pfeifer JD, El- Mofty SK.(2000) Squamous cell carcinoma arising inreccurent respiratory papollomatosis with pulmonary involvement: emerging common pattern of clinical features and human papillomavirus serotype association. Mod Pathol13: 914–18. |