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 bronchoscopie 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.

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 endoscopy 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 recurrcntpapillomatosis - RRP), and refers to antiviral and immunoregulation drugs. Other available adjuvant drugs include anti-reflux drugs, mitomycinC, cyclooxygenase2 inhibitors, retinoids, zinc and in-dole-3-carbinol, interferon –α. The commonly used antiviral drugs include cidofovir, ribavirin, acyclovir and ganciclovir. (KimberlinDW(2004); Bielecki I, Mniszek J &Coafała M 2009)
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, though 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.
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