Inflammatory pseudotumor of the subglottis


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Summary. A rare case of an idiopathic inflammatory pseudotumor of the larynx is described. Management consisted of midline vertical thyrotomy with anterior cricoid splitting, excision of the subglottic tumor and temporary stenting of the lumen with a siliconized tube. While these lesions are benign, the therapeutic consequences of misdiagnosis can be serious. Effective treatment consists of surgical excision, which results in a permanent cure.

Key words: Inflammatory pseudotumor – Larynx – Surgical excision

Introduction

The inflammatory pseudotumor is an unusual benign lesion which was first described in the lung by Brunn [2] in 1939. The description “pseudotumor” reflects the clinical and radiological presentation of these space-occupying inflammatory lesions as true neoplastic processes, especially when they occur in the lung.

Inflammatory pseudotumors most commonly occur in the lungs, although fewer than 50 cases have been reported in the literature [11, 17, 19]. Other sites reported have been the intratracheal mucosa [21], the urinary bladder [16], the pterygomaxillary space [7] and the maxillary sinus [20].

Only two cases of inflammatory pseudotumor of the larynx have been reported previously [6, 22]. The purpose of this paper is to present a third case which was also situated in the subglottis. The clinical history, histological findings and therapy are elucidated.

Case report

A 56-year-old Caucasian woman was referred to the ENT Department of the University Hospital, Nijmegen, for evaluation and treatment of a subglottic laryngeal tumor. Three weeks before admission, she underwent a tracheotomy elsewhere as treatment for progressive stridor with airway obstruction. Concomitant direct laryngoscopy revealed a subglottic tumor apparently projecting from the posterior wall of the larynx and obstructing the lumen.

Fig. 1. Artist’s drawing of the clinical presentation of the subglottic mass lesion

The provisional histopathological diagnosis of the biopsy specimen was “infected leiomyoma”.

Past medical history revealed slight hoarseness of the voice and slowly progressive dyspnea on exertion which had started a few years previously. Regular check-ups by a chest physician were had but without defining a cause for the dyspnea. On admission indirect laryngoscopy revealed slightly impaired movement of both vocal cords on phonation and a circular subglottic mass which reduced the lumen (Fig. 1). Chest X-ray was normal but a soft tissue density was apparent in the subglottic area. Computed tomography confirmed the soft tissue swelling, which extended from below the vocal cords to just above the trachea.

Routine laboratory investigations revealed no abnormalities, although prior to the tracheotomy elsewhere, erythrocyte sedimentation rate was found to be elevated and total globulins were increased on electrophoresis, suggesting chronic inflammatory disease.

Direct laryngoscopy and tracheoscopy after admission confirmed the existence and extent of the subglottic soft tissue mass. New biopsy specimens were taken and were now found histologically to be chronic granulating inflammation. However, antibiotic and steroid treatment did not result in any regression of the tumor and the diagnosis of the first biopsy was revised to be compatible with an inflammatory pseudotumor.

The patient subsequently underwent a median vertical thyrotomy with splitting of the cricoid arch anteriorly. Tumor was found to extend mainly on the posterior wall and on the lateral side of the subglottic area. The lesion was then excised together with a substantial portion of the posterior part of the cricoid cartilage, which was apparent to be involved. The subglottic lumen was kept patent with a conforming siliconized (Berkostent) tube having an outer diameter of 12 mm and 100 mm length (Lubrelastic Medical Appliances, Hazerswoude, The Netherlands).
Fig. 2a, b. Paraffin section of the initial subglottic biopsy, showing a moderately cellular process of plump spindle cells and a patchy mononuclear infiltrate. H & E staining; a × 25, b × 100

The tube was changed twice and removed permanently after 5 months. The airway had been protected with a tracheotomy tube which was removed 2 weeks later. The patient has been disease-free for 4 years and only slight hoarseness has remained. The definitive pathological diagnosis after laryngeal surgery was inflammatory pseudotumor.

**Histopathology**

The successive biopsies showed consistent features of a moderately cellular process consisting of loosely arranged plump spindle cells which penetrated deeply into the subepithelial connective tissue layer (Fig. 2). Nuclear pleomorphism and mitotic activity were minimal. A conspicuous patchy mononuclear inflammatory infiltrate was present. After immunohistochemical staining of paraffin sections, the spindle cells were seen to be partly positive for actin and sporadically positive for desmin, findings compatible with myofibroblastic differentiation. Electron microscopic results were also compatible with myofibroblasts, consistent with an “inflammatory pseudotumor” (Fig. 3). The overlying mucosa was formed by normal cylindrical and partly squamous epithelium. The cartilage appeared to be unaffected.

**Discussion**

A review of the literature revealed only two histologically confirmed cases of inflammatory pseudotumor of the larynx. The first tumor occurred in an 11-year-old girl who presented clinically with airway obstruction [6]. The pseudotumor extended from just inferior to the posterior glottic commissure to the posterior and lateral walls of the trachea. The lesion was excised surgically