International Journal of Pediatric Otorhinolaryngology (2008) 72, 279–283

International Journal of Pediatric Otorhinolaryngology

www.elsevier.com/locate/ijporl

CASE REPORT

Childhood granular cell tumors: Two case reports

N. Leboulanger [a,*], I. Rouillon [a], J.F. Papon [a], P. Josset [b], G. Roger [a], E.N. Garabédian [a]

[a] *ENT and Cervico-Facial Department, Armand-Trousseau Children's Hospital, faculté Paris VI, Paris, France*
[b] *Histopatholoy Department, Armand-Trousseau Children's Hospital, faculté Paris VI, Paris, France*

Received 2 August 2007; accepted 23 October 2007
Available online 20 December 2007

KEYWORDS
Granular cell tumor;
Children;
Larynx;
Trachea;
Multifocal

Summary Granular cell tumors (GCT), also known as Abrikossoff tumors, are rare tumors found largely in children, with few reports of laryngo-tracheal involvement. Two childhood cases of laryngo-tracheal GCT are reported here, of which one case had multifocal lesions. The histopathological features and the therapeutic management of GCT are also discussed.

1. Introduction

GCT, or Abrikossoff tumors (eponymously named after the first physician to have identified this entity in 1926), are rare tumors and are usually benign. They can occur at any site, but are frequently localized to the head and neck, in particular, with involvement of the tongue. Lesions occurring in the upper airways and in the tracheo-bronchial tree are unusual. A 1999 review of the literature [1] identified 32 cases of GCT involving the trachea, of which only 5 cases were in children. Since then, one other case has been reported [2]. Pediatric cases with laryngeal lesions are slightly more frequent, with approximately 20 patients in the literature [3]. In general, solitary lesions are present, but GCTs may also be multifocal.

To the best of our knowledge, only four cases of multifocal GCT with tracheal lesions have been reported in children [4,5].

This article presents two cases of childhood laryngo-tracheal GCT, one of which had multifocal lesions. The histopathological features and the therapeutic management of GCT are also discussed.

2. Case reports

2.1. Case no. 1

In September 2000, a 12-year-old girl was seen with progressive dysphonia and a hoarse voice. The dysphonia had developed very gradually over a period of 1 year in a child with a previously normal voice. In addition, the patient was obese and under the care of a dietician.

Indirect laryngoscopy showed a sessile pedunculated tumor on the entire left vocal cord, sparing the anterior commissure, the arytenoid, the ven-

* Corresponding author. Tel.: +33 144736923; fax: +33 144736108.
E-mail address: nicolas.leboulanger@trs.aphp.fr (N. Leboulanger).

0165-5876/$ – see front matter
doi:10.1016/j.ijporl.2007.10.021

tricular band, and the ventricle. The mobility of the arytenoid was normal. The remainder of the clinical examination was normal, in particular, examination of the buccal cavity, the nasal fossae and the skin.

Direct laryngoscopy and bronchoscopy under general anesthesia showed that the left vocal cord lesion was associated with a spherical lesion measuring approximately 1.5 cm in diameter in the right retro-cricoid area and two soft and sessile lesions on the right aspect of the cervical trachea. These two lesions were not seen at the time of consultation. The tracheal lesions were approximately 0.5 cm in diameter, whitish grey in color and were not significantly diminishing the internal caliber of the trachea. The main bronchi as well as the esophagus appeared healthy.

A cervical CT scan did not show any lymphadenopathy and confirmed the integrity of the paraglottic space, the cricoid cartilage and the paratracheal tissues.

Multiple biopsies were taken. Histological examination revealed a normal mucosa; however, below this were poorly defined aggregates of rounded polygonal cells with eosinophilic and granular cytoplasm and a zone of pseudoepitheliomatous hypertrophy. There were no mitotic figures and fibrous connective tissue surrounded the entire lesion. Immunostaining showed strong expression of S-100 protein. The final histopathological diagnosis was multifocal GCT.

Endoscopic management was the preferred option. A CO_2 laser was used to resect the tracheal and laryngeal lesions. Wide excision margins were employed for the larynx and histopathological examination confirmed that these were free of disease.

Endoscopic follow-up at 2 weeks post-intervention showed a moderate degree of persistent edema of the left vocal cord. The dysphonia was much better, although the voice had not completely normalized at this point in time. Endoscopic examination at 2 months was normal and the child was lost to follow-up.

2.2. Case no. 2

In November 2006, a 12-year-old boy presented as an emergency with a laryngeal lesion. He gave a 1 month history of progressive and significant dysphonia, with inspiratory dyspnea of moderate severity over a few days, which was variable over time.

Nasal fiberoptic examination in consultation showed a rounded sessile lesion of the right laryngeal vestibule, measuring about 1 cm in diameter, which prevented visualization of the vocal cords. There was marked reduction of the caliber of the laryngeal passage, although this was well tolerated by the patient. The variability of the dyspnea was attributable to the sessile and mobile nature of the lesion, which traversed the plane of the cords with each respiratory cycle and produced a degree of obstruction dependent on its exact position. The rest of the clinical examination was normal. There were no palpable cervical masses.

An emergency laryngeal ultrasound revealed a bilobed laryngeal mass, arising partly from the right ventricular band and extending into the lumen of the larynx. For completeness, a subhyoid cystic structure was noted, suggestive of a thyroglossal duct cyst, which had not been previously detected and which could not be found clinically.

The following day, an endoscopy was performed under general anesthesia with direct laryngoscopy. In view of the respiratory compromise caused by the lesion, it was decided to relieve the obstruction immediately (Photos 1—3). The patient was intubated using a small sized stiff intubation tube, which was inserted after laryngeal exposure and displacement of the lesion towards the posterior part of the subglottis. Resection of the pedunculated tumor on the right ventricular band was performed with a CO_2 laser. The tumor showed a minor degree of deeper extension, presumably corresponding to the ultrasound findings. The vocal cords and the adjacent tracheo-bronchial tree were normal. Macroscopically, the resection was complete. Histopathological examination revealed a normal mucosa; however, below this were monotonous round cells with eosinophilic and granular cytoplasm, indicating a GCT. The excision margins were free of disease.

On awakening, there was a spectacular improvement in the dysphonia and dyspnea. A follow-up fiberoptic examination at 1 week showed a scarred, healed ventricular band and normal mobility of both

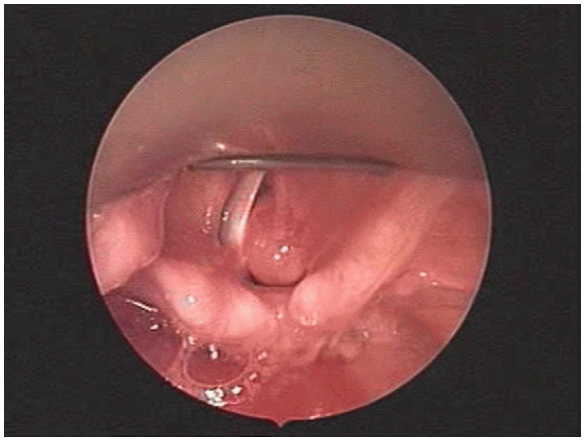

Photo 1 Lesion of the right laryngeal vestibule on inspiration.