CASE STUDY

Subglottis Inflammatory Pseudotumour in a 3-Year-old Child

Seudotumor inflamatorio subglótico en un niño de 3 años

Javier Cervera Escario, a, * Sara Sirvent Cerdá, b Saturnino Santos Santos, a Adolfo Sequeiros González c

a Servicio de Otorrinolaringología Pedíátrica, Hospital Universitario Niño Jesús, Madrid, Spain
b Servicio de Radiodiagnóstico, Hospital Universitario Niño Jesús, Madrid, Spain
c Servicio de Neumología, Hospital Universitario Niño Jesús, Madrid, Spain

Received 10 January 2017; accepted 30 April 2017

We present the case of a 3-year-old child who from the age of 10 months had suffered from recurrent bronchitis. The child was referred to the pneumology department of our hospital due to increased difficulty in breathing. An endoscopy of the airway was performed, under general anaesthesia, with 0° optics (Fig. 1A), from which a soft tumour was appreciated in the subglottis region. No infiltrative aspect was appreciated, it was rosy in colour, palpable, and occupied 75% of the airway calibre. In the imaging study using computed tomography (CT) an occupation of the airway in the posterior area of the subglottis was observed. The study was completed with magnetic resonance (MRI) (Fig. 1B), which reported: "non specific soft tissue mass, located in the posterior wall of the subglottis, compatible with a primary diagnostic option of subglottic angiomata".

Differential diagnosis was made of this lesion. Its location, the endoscopic imaging and radiology pointed to subglottic angioma whilst the patient’s age, gender and the fact there were no locations on the skin or mediastinum or liver pointed against it.

Treatment with propranolol was initiated in accordance with the described protocol a although there was not a total concordance with an initial diagnosis of hemangioma.

A month after the start of treatment, the patient continued with breathing difficulties and a further MRI was performed, which reported: "non specific soft tissue mass, located in the posterior wall of the subglottis, compatible with a primary diagnostic option of subglottic angioma, with the same dimensions as in the previous MRI".

In the light of this situation laryngeal microsurgery was performed with micropincers resecting the mass, which was of soft, somewhat fibrous consistency. There was no significant bleeding. It was impossible to remove the tumour completely due to the lack of anatomical references as no dissection plane enabled the tumour limits to be observed.
months obstruction mixed CT of and eosinophils. Figure 176 Treatment inflammatory Discussion (and active polymorphonuclear 4 mm and 0° optics, in which the image of a tumour may be observed. It occupies 75% of the subglottic lumen, with posterior implant base of 8.9 mm cranial-caudal extension, soft and palpable. (B) MRI with STIR sequence in sagittal plane shows a small mass of soft posterior subglottic parts (thin arrow) and in the axial plane after intravenous administration of paramagnetic contrast corresponding to a mass which is intensely enhanced (thick arrow) and occupies almost all the lumen of the airway.

![Image](http://www.elsevier.es/)

Figure 1 Pathological anatomy of the inflammatory pseudotumor: mixed infiltration composed of mature lymphocytes, plasmatic and occasionally polymorphonuclear cells. The mature T lymphocytes predominate (CD3+) over the B lymphocytes (CD20+).

Treatment with steroids was administered intraoperatively and postoperatively for 10 days. The patient evolved favourably with complete disappearance of the respiratory obstruction symptoms. Check-ups performed reported normal and as no respiratory symptoms appeared in subsequent months it was decided not to perform further endoscopies of the airway under general anaesthesia. The postoperative CT scan showed an increase in subglottis lumen from 2 to 4 mm.

The pathological report reported: sub epithelial oedema, mixed inflammatory infiltration composed of lymphocytes, polymorphonuclear neutrophil leukocytes and occasional eosinophils. Immunohistochemical study reported: positive cells for lymphoid markers (CD45, CD3 and CD20), and negative cells for muscle markers (Myo-D1 and actin) (Fig. 2). Histopathological diagnosis: "inflammatory pseudo tumour".

Discussion

Inflammatory pseudotumours are rare lesions, from the mesenchyma. They are not malignant, are well circumscribed, not encapsulated, and present infiltration of inflammatory cells which look like malignant tumours. These pseudo tumours are found most frequently in the lungs and in the eye socket although they have been described as being located in almost all parts of the body. Very few cases are been reported in paediatric airways, and particularly in the larynx which is referred to in 11 cases in the literature. The treatment of choice is complete endoscopic resection with micropincers, and with the use of the KTP laser. Treatments with corticoid steroids have also been described. Due to the young age of our patient, treatment with corticoid steroids was not recommended long term since there could be major side effects in a child.

It may be confused with other more common diseases which have a highly similar endoscopic appearance and show similar results in radiologic imaging.

Differential diagnosis of inflammatory pseudotumor includes both benign and malignant tumours. The benign ones are: sarcoidosis, tuberculosis, amyloidosis, perichondritis, Wegener’s granulomatosis, fibromatosis and hemangiomas. The malignant ones are: rhabdomyosarcomas, leiomyosarcomas, fusiform cell tumours and melanomas. These are exceptional in the paediatric population.
Subglottis Inflammatory Pseudotumour in a 3-Year-old Child

Conclusion

When a tumour occupies the lumen of the airway in a child, differential diagnosis must include all possible pathologies. It is true that subglottic hemangioma was initially the most viable option, albeit doubtful, due to discordances in gender and age. For this reason a non aggressive medical treatment was sought, without successful outcome. It is important to always take into consideration other diseases which, although rare, may be the cause of this type of tumours.

Conflict of Interests

The authors have no conflict of interests to declare.

References