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CASE STUDY

Laryngeal stenosis as late manifestation of rhinoscleroma. Case report

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KEYWORDS
Rhinoscleroma;
Subglottal stenosis;
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Abstract
Rhinoscleroma is a chronic, progressive, granulomatous disease resulting from infection with Klebsiella rhinoscleromatis. It is generally uncommon, but endemic in certain regions of the world. It typically affects the nasal cavity, but also other parts of the respiratory system, such as the laryngotracheal region, which may be life-threatening if compromised. We present a 46-year-old male patient with a history of rhinoscleroma diagnosed and treated 15 years ago, with subglottal stenosis as a late sequela of this disease.

PALABRAS CLAVE
Rinoescleroma;
Estenosis subglótica;
Klebsiella rinoescleromatis

Estenosis subglótica como manifestación tardía de un rinoescleroma. Presentación de un caso

Resumen
El rinoescleroma es una entidad crónica, progresiva y de aspecto granulomatoso, resultado de la infección por *Klebsiella rinoescleromatis*. Es en general infrecuente, pero endémica en ciertas regiones del mundo. Afecta típicamente a la cavidad nasal, pero también a otras partes del sistema respiratorio, como la región laringotraqueal, cuyo compromiso puede representar un riesgo para la vida. Presentamos el caso de un paciente de sexo masculino de 46 años de edad, con el antecedente de rinoescleroma diagnosticado y tratado hace 15 años, que presenta estenosis subglótica como secuela tardía de esta enfermedad.

Introduction

Rhinoscleroma is a chronic granulomatous disease of the upper airway. It is the result of infection by *Klebsiella rinoescleromatis*, a gram-negative, capsulated bacterium.[1] This infection is an uncommon condition, but endemic in certain regions of the world such as Central and South

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America, Central and Eastern Europe, Africa and Asia.[2] It appears as a chronic, progressive entity with granulomatous aspect, which typically causes rhinoscleroma, but can also affect other parts of the respiratory system.[3] Laryngeal and tracheal affectations require special attention, as they can pose a risk to the patient's life. The presence of dysphonia, dyspnoea, stridor and cough suggest laryngotracheal involvement, with the subglottic region being the most affected subsite. In these cases, only endoscopic examination will reveal the lesion. In the literature, the incidence of laryngeal involvement in rhinoscleroma varies between 12.5% and 63.6% of cases.[2-5]

Case study

We present the case of a 46-year-old male patient from an endemic region, with a history of rhinoscleroma, who was histologically diagnosed 15 years before, who had immigrated to our area in that period, with no evidence of nasal relapses since then. He consulted due to dyspnoea and dysphonia of long evolution. He was examined through nasofibroscopy, which revealed synechiae of the vocal folds and subglottic stenosis with respiratory lumen of 5 mm in phonation (Figure 1), thus being, strictly speaking, a glottic-subglottic stenosis. Inhaled steroids were indicated and a direct laryngoscopy was performed; synechiae were sectioned and a Gore-Tex® keel was placed in the right anterior third region. Biopsy material was taken from the subglottic region, the delayed pathology report of which showed the presence of a chronic rhinoscleromatous granulomatous process at the laryngeal level. One week after surgery, the patient was in good condition under antibiotic treatment with levofloxacin. Telelaryngoscopy showed separate vocal folds with normal mobility, without dysphonia and a well positioned keel (Figure 2). At the time of writing of this report, there was no evidence of recurrence, and the patient was in very good condition. The patient presented no other risk factors for laryngeal stenosis.

Discussion

Laryngeal sequelae of infection by *K. rinoescleromatis* have been reported in approximately 12.5% to 63.6% of cases.[2-5] They can represent a risk to life, given the obstructive affectation of the airway. Our case presented synechiae of the vocal folds and subglottic stenosis. The management of these disorders seeks, regardless of the cause, to restore the laryngeal anatomy and function. The type of surgery depends on the individual case. This patient underwent

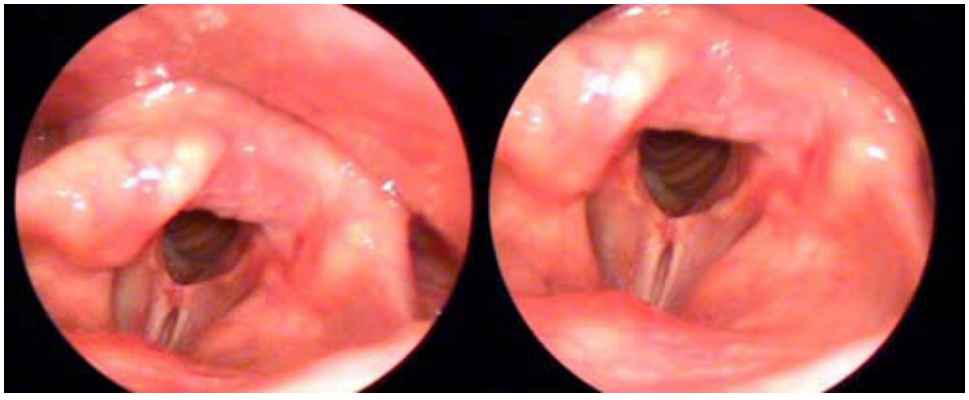

Figure 1 Fibroscopic evaluation prior to surgical intervention.

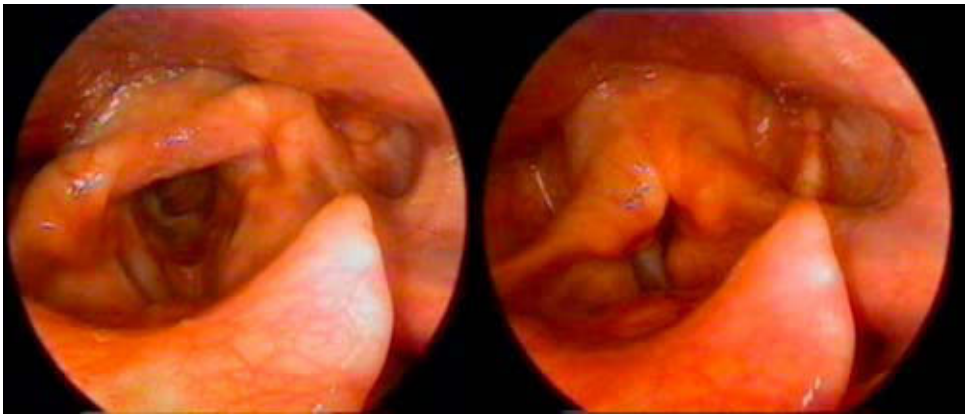

Figure 2 Postoperative fibroscopic visualization. The correctly positioned Gore-Tex® keel can be observed in the subglottic region.

section of synechiae and placement of a Gore-Tex® keel, which yielded satisfactory results. Gore-Tex® corresponds to a microporous fluoropolymer with great flexibility of vibration; it is easy to place and has a reasonable cost. It is also a very easily-handled prosthesis, which does not have to be modelled previously and which is completely biocompatible, so its use is recommended.[6]

Surgical methods to resolve this pathology can be grouped into two general categories. The endoscopic method, which includes traditional dilation and laser technique[7] and, secondly, open surgery, which still remains the best method for handling severe laryngotracheal stenosis, especially in cases requiring tracheostomy.[8] With this case, we seek to reflect on a disease that requires a high level of suspicion and that should currently be borne in mind given the increase in immigration from endemic regions.

Conflict of interests

The authors declare no conflict of interests.

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