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Case report/Kazuistyka

An isolated laryngeal sarcoidosis in a child threatening to the airway – A case report

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ABSTRACT

Sarcoidosis is a chronic granulomatous disease of an unknown etiology. Involvement of head and neck area with sarcoidosis is rare whereas isolated laryngeal involvement is even more exceptional and rare particularly in children. Laryngeal involvement with sarcoidosis is a challenging situation not only to diagnose but also to treat. Laryngeal sarcoidosis may be seen as an isolated lesion or as an associated lesion of systemic sarcoidosis. Laryngeal involvement may cause dysphonia, dysphagia and dyspnea. Fatal airway complication like stridor can occur. We present a rare case of an isolated laryngeal sarcoidosis in an 8-year-old male child involving the epiglottis and aryepiglottic folds with compromising the supraglottic airway leading to stridor.

Introduction

Sarcoidosis is a chronic granulomatous disease affecting the multiple organs in the body of an unknown etiology. It is a multi-system disease that can involve any organs and is an uncommon among pediatric patients [1]. Common sites of body involved are lungs, hilar and mediastinal lymph nodes, eyes, skin, liver, bone and the nervous system. Dermatologic involvement of sarcoidosis was first described by Jonathan Hutchinson in 1875 and the histopathology was described by Boeck in 1899 and later on the disease called as Boeck's disease whereas the laryngeal sarcoidosis with confirmed histopathology report was first described by Poe in 1940 [2]. The diagnosis of sarcoidosis is based on clinical, radiological and histological examination. The laryngeal involvement is rare, occurs in 0.5–8.3% of patients with known sarcoidosis [3]. In the sarcoidosis, the symptoms are usually mild even in case of extensive involvement. This tends to progress slowly and may passes through relapse and remission. But in laryngeal involvement is marked by narrow airway where mild involvement can manifest life-threatening situation.

Case report

An 8-year-old male child presented to the outpatient Department of Otorhinolaryngology for breathing difficulty

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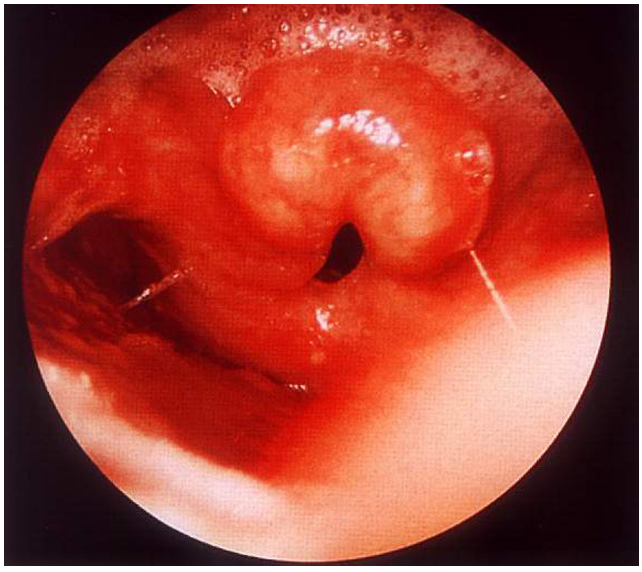

Fig. 1 – Flexible nasopharyngolaryngoscopy showing diffuse swelling of epiglottis and aryepiglottic fold

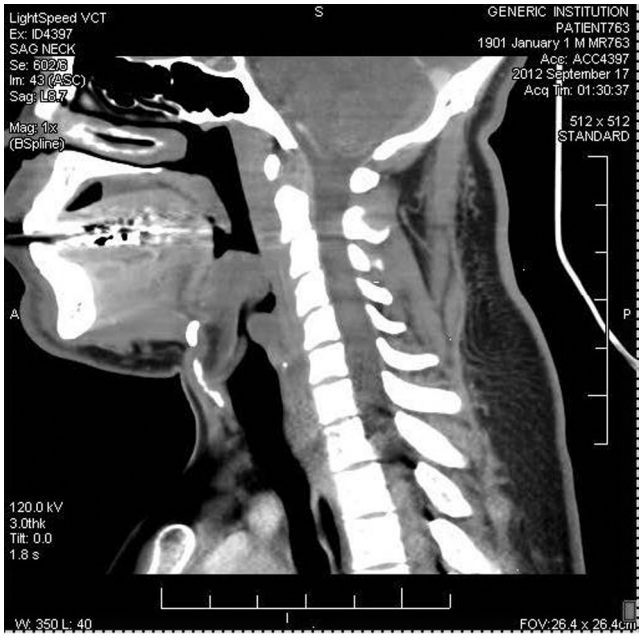

Fig. 2 – CT Scan picture showing a large supraglottic mass obstructing the supraglottic airway

since 3 days. He had history of dysphagia, cough and mild breathlessness since 3 months. The child had taken antibiotics from local physicians for presumed upper respiratory tract infections but without any successful outcome. He had no loss of appetite or loss of weight. Flexible laryngoscopy revealed diffuse edema of supraglottis including epiglottis, aryepiglottic folds and arytenoids cartilages with normal glottis (Fig. 1). Chest X-ray showed no evidence of pulmonary disease or hilar lymphadenopathy. Computed tomography (CT) scan of the neck showed a mass arising from the epiglottis and aryepiglottic folds measuring 3.5 cm × 2.5 cm (Fig. 2). The patient undergone direct laryngoscopy and tissue taken from the epiglottis for biopsy. Histopathological examination showed granulomatous inflammation with epitheloid granulomas, occasional multinucleated giant cells and without any evidence of caseous necrosis (Fig. 3). A diagnosis of sarcoidosis was made. Laboratory investigations revealed the values: ESR (35.0 mm/h), C reactive protein (13.5 mg/l), angiotensin converting enzyme (ACE) (24.2 IU/l) and negative for antinuclear antibody (ANA) and anti-DNA. Because of the focality of sarcoidosis at the supraglosttis, patient was undergone two episodes of office procedure for intralesional injection of triamcinolone. Along with this, we also started systemic corticosteroids. After 6 months of treatment, child was

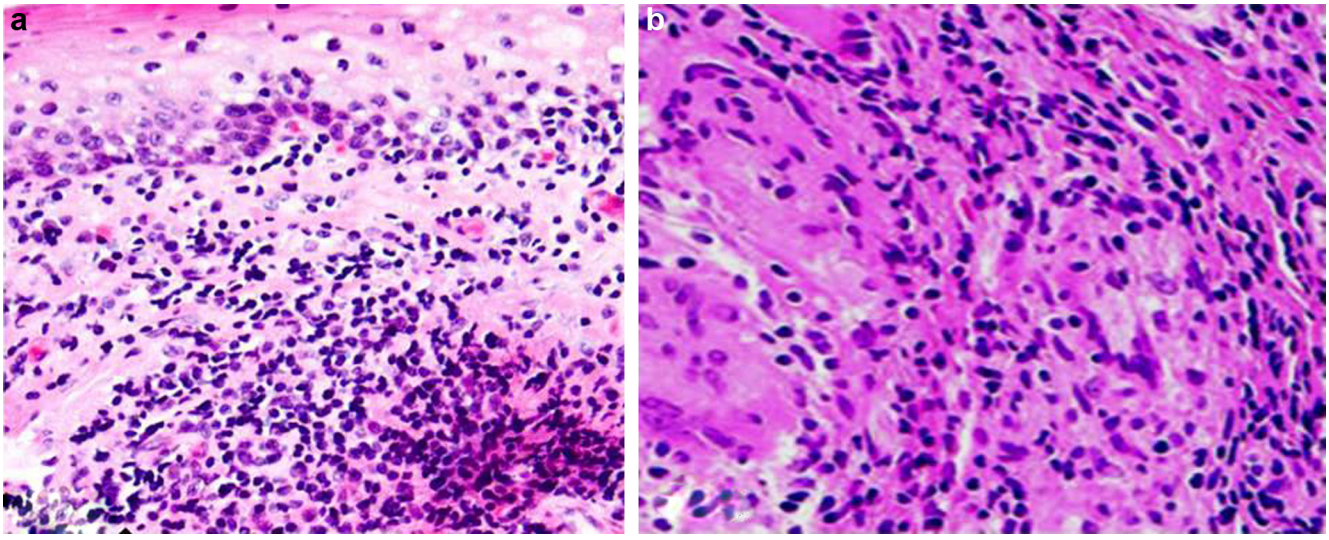

Fig. 3 – Microphotograph showing hematoxylin–eosin stained noncaseating granulomas with giant cells and surrounding epitheloid macrophages typical of sarcoidosis (Original magnification 200× [a], 600× [b])