CASE REPORT

Rhinology, 39, 176–179, 2001

# Post-adenoidectomy inflammatory pseudotumor*

Usamah Hadi[1], Mohamed El-Bitar[1], Ghazi Zaatari[2]

[1] Department of Otolaryngology-Head & Neck Surgery, American University of Beirut Medical Center, New York, USA
[2] Department of Pathology & Laboratory Medicine, American University of Beirut Medical Center, New York, USA

SUMMARY

*Problem: Inflammatory pseudotumor is a rare pathology in the head and neck area. Multiple post-adenoidectomy complications have been described in the literature without alluding to such an entity.*
*Method: A case report of an inflammatory pseudotumor following an adenoidectomy.*
*Main Result: Pseudotumor of the nasopharynx should be added to the list of possible complications of adenoidectomy.*
*Conclusion: Inflammatory pseudotumor of the nasopharynx is a rare complication that confronts the otolaryngologist and the pathologist with a diagnostic challenge. Surgical excision remains the best therapeutic option.*

*Key words: post-adenoidectomy, pseudotumor, inflammatory, pediatric, nasopharynx*

## INTRODUCTION

The nasopharynx is an anatomical location that harbors a number of masses, both benign and malignant. Hypertrophied adenoids are considered the leading cause of benign masses of the nasopharynx. Respiratory distress and airway obstruction accompany patients with hypertrophied adenoids, at times leading to obstructive sleep apnea, particularly in children who are less than 3 years old.

Adenoidectomy is a commonly performed procedure in children. Generally considered a safe surgery, yet potential complications can emerge and lead to devastating consequences. Multiple post-adenoidectomy complications have been described in the literature. Some are minor and easily controlled while others are major and require skill to diagnose and manage. Post-adenoidectomy masses presents a real challenge to the otolaryngologist head and neck surgeon. The differential diagnosis of such a condition depends on the clinical, radiological and pathological parameters.

We report a child who suffered from a post-adenoidectomy inflammatory pseudotumor of the nasopharynx. To the best of our knowledge, such a complication has not been reported before. In this report we review the clinical course, the pathological features and treatment for this unusual and rare complication. A review of inflammatory pseudotumor in the head and neck region is included.

## CASE REPORT

A 4-year old girl underwent an adenoidectomy 4 months prior to her presentation. One month later she reported the recurrence of her obstructive nasal symptoms. The parents reported the history of a transient upper respiratory tract infection. There was no history of epistaxis, neurological or endocrine abnormalities.

The physical examination revealed a white mucopurulent discharge filling both nasal cavities. A yellow shiny mass was seen protruding from the nasopharynx into the superior part of the oropharynx. She received a course of antibiotics, systemic corticosteroids with topical decongestants, but showed no improvement.

Computed tomographic (CT) scan showed a non-enhancing, broad-based 2x2cm mass arising from the posterosuperior wall of the nasopharynx and impinging on the posterior choanae (Figure 1). There was no invasion of adjacent structures or communication with the central nervous system or paranasal sinuses. The clinical picture was not suggestive of any systemic illness. Consequently, no bacteriologic or serologic studies were performed.

Under general anesthesia, the patient underwent direct nasopharyngoscopy using a 4mm (zero and 30 degrees) Hopkin's telescope. A yellow soft multi-lobulated movable mass was

* Received for publication: February 9, 2001; accepted: May 28, 2001.

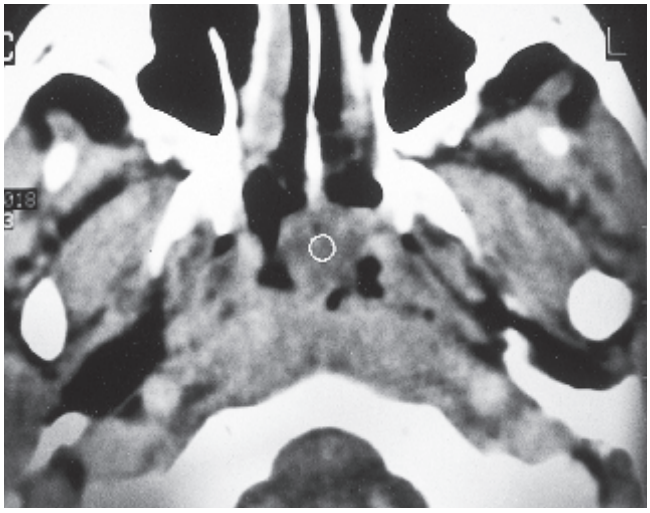

Figure 1. Computed tomographic (CT) scan with contrast, of the nasopharynx showing a non-enhancing soft tissue filling the nasopharynx (circle) and obstructing the choanae.

noted; it was attached to the postero-superior wall of the roof of the nasopharynx by a broad base. There was no erosion of the nasopharynx, vomer, or lateral nasal walls. It completely blocked the choanae. The tumor was excised under direct vision using the adenotome.

The excised mass measured 2 cms and was multi-lobulated, non-compressible, soft yellow with smooth shiny surface. Histologic examination showed the lesion to be composed of elongated spindle shaped cells (Figure 2). These appeared uniform, showed variable compactness, and demonstrated rare mitoses. The surrounding stroma was edematous, showed a rich capillary network, and was heavily infiltrated by a mixture of small lymphocytes, neutrophils and plasma cells. No adenoidal tissue or microorganisms were seen. These findings were consistent with the diagnosis of inflammatory pseudotumor. The patient's postoperative course was smooth and uneventful. A one-year follow up showed no evidence of recurrence.

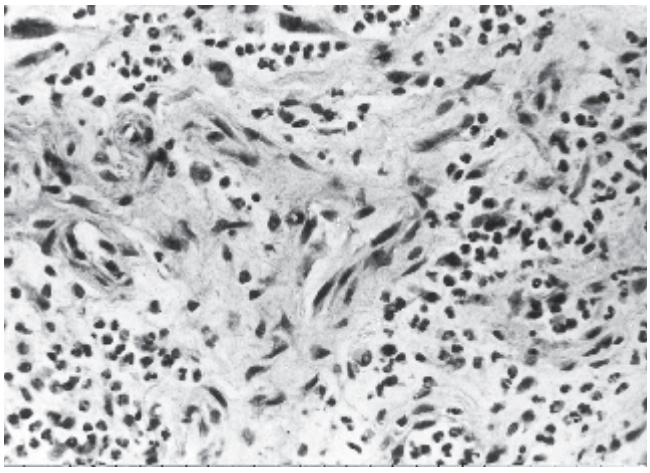

Figure 2. Photomicrograph of the lesion showing spindle shaped cells vascularized stroma and abundant intermixed neutrophils. (hematoxylin & eosin 400x)

DISCUSSION

Multiple post-adenoidectomy complications have been described in the literature.

Among the common and major complaints are significant palate or tongue edema during or after the procedure. This may encroach on the airway and lead to obstruction especially in children 3 years of age or less. Risk factors contributing to airway obstruction include obstructive sleep apnea, neuromuscular disorders, obesity and craniofacial anomalies (Wiatrak et al., 1991; Arnold and Allphin, 1993; Price et al., 1993; Rothschild et al., 1994; Soultan et al., 1999). Sore throat, vomiting, otalgia, dehydration and fever are also frequent complaints. Postoperative hemorrhage usually responds to local measures such as cautery or packing. However, it can be life threatening requiring prompt identification and control of the bleeding vessel. Unanticipated bleeding might reflect an underlying hematological abnormality such as von Willbrand disease (Isaacson and Parke, 1996; DeDiego et al., 1999).

Transient velopharyngeal insufficiency may follow removal of large adenoid tissues. This resolves spontaneously in patients with normal palatal musculature. However, Gates et al. (1992) reported that in patients with abnormal palate anatomy or function, the insufficiency will persist unless corrected by a pharyngeal flap. Perkins et al. (2000) highlighted the presence of a 22q11 deletion in such patients.

Nasopharyngeal stenosis, as reported by Haller et al. (1999) and Conley et al. (1997), is a dreadful complication that requires local flaps and free mucosal graft for successful repair. Nasotracheal and/or nasopharyngeal tubes tend to increase the risk for stenosis.

Other infrequent and rare post-adenoidectomy complications have been described in the literature. These include mandible condylar fracture (Randall and Hoffer, 1998), atlanto-axial subluxation and cervical osteomyelitis (Baker et al., 1996), bacterial meningitis (Isaacson and Parke, 1996), necrotizing fascitis (Feinerman et al., 1999) and torticolis (Bedi et al., 1999).

Mortality rates are low especially with the availability of better anesthetic agents and adequate postoperative monitoring. In spite of this, the risks of aspiration and pulmonary edema remain a threat to the child's well being (Motamed et al., 1999).

Inflammatory pseudotumor is a rare pathology in the head and neck area. In children, it has been typically described in the lungs; it arises de novo and carries a benign course. However, extrapulmonic forms have been rarely reported. Coffin et al. (1995) reviewed 84 extrapulmonary lesions, with only 12 occurring in the head and neck region. Of these, one involved the skin of the face, one involved the meninges and one arose in the orbit. The remaining 9 cases affected the larynx, trachea, oropharynx and the nasopharynx. In the two cases where the nasopharynx was involved, there was no information about their clinical course. The emergence of inflammatory pseudotumor in the nasopharynx has not been well documented. Famous et al. (1992) described a patient with such a lesion arising in the retropharyngeal space. Short of this, there is no mention in the

literature of a post-adenoidectomy inflammatory pseudotumor arising in the nasopharynx. Initially, inflammatory pseudotumors have been reported in the urinary bladder and genitourinary tract after instrumentation (Enzinger and Weiss, 1998). The pathophysiology for the appearance of such a process remains unclear. It has been proposed that instrumental manipulation acts as a stimulant or irritant that eventually produces a dramatic tissue response leading to the formation of an exuberant inflammatory reaction with secondary tissue proliferation. Proppe et al. (1984) stressed that operative injury to human tissues would result in an aberrant healing process at the site of mucosal incision forming spindle cell nodules 5 weeks to 3 months post injury. These lesions appeared shortly after vaginal hysterectomy. This was also described following an episiotomy. Transurethral resection of the prostate gland or urinary bladder leads to similar pathologies. These abnormalities were noted at the surgical site. Proppe et al. (1984) alluded to similar lesions in the larynx following endoscopic excision of a "pyogenic granuloma", and another occuring in the gingiva following extraction of a tooth.

Additional cases of inflammatory pseudotumor have been described in the literature that involved the nasal cavity (Huang, 1993) and the nasal septum (Fischer et al., 2000). Its occurrence in the paranasal sinuses is rare and can mimic malignant tumors as described by Som et al. (1994). Single case reports describe inflammatory pseudotumors at diverse sites such as the tonsils (Newman and Shinn, 1995), submandibular region (Invi et al., 1993), nasopharynx with retropharyngeal space inflammation that resulted in sequential abducen nerve palsies (Famous et al., 1992), and in association with the common carotid artery (Kim et al., 1999).

Although inflammatory pseudotumor appears clinically as a rapidly growing lesion, histologically it has three basic patterns, none of which is associated with malignant microscopic features (Coffin et al., 1995). Despite the benign histologic appearance, their biologic behavior can be that of a locally aggressive nature mimicking a malignant tumor. CT and MRI findings of inflammatory pseudotumor of the head and neck are nonspecific and may suggest invasive carcinomas or granulomatous disease (De Vuysere et al., 1999). In one study by Som et al. (1994), patients with maxillary sinus inflammatory pseudotumor had a soft tissue mass with a mild enhancement on the CT scan and an intermediate signal intensity on both T1-T2 weighted MR images.

Histologically, post-adenoidectomy inflammatory pseudotumor of the nasopharynx is similar to that of other organs. The proportion of spindle cells, lymphocytes, plasma cells and variation in pattern of presentation is highly variable (Coffin et al., 1995). In highly cellular lesions, the histology mimics soft tissue tumors and the differential diagnosis microscopically includes fibroma, fibrosarcoma, lymphoepithelioma, rhabdomyosarcoma, plasmacytoma, spindle cell sarcomas, lymphoma, other pseudosarcomas like fibromatosis, myofibroblastoma, fibrous histiocytoma and inflammatory malignant fibrous histiocytoma (Coffin et al., 1995).

Clinically and radiographically, the appearance of a nasopharyngeal mass post-adenoidectomy presents a real challenge to the otolaryngologist. Entities such as recurrence of adenoid tissue, antrochoanal polyp, angiofibroma, encephalocele, chordoma, teratoma, dermoid cyst, craniopharyngioma should be excluded in the differential diagnosis.

Treatment consists of complete surgical excision, which is usually curative. Recurrence of inflammatory pseudotumor reflects either an incomplete excision related to its site of location and proximity to vital structures or due to a tendency toward aggressive biologic behavior. Establishing the correct diagnosis is vital in order to avoid unnecessary extensive and mutilating surgery (Coffin et al., 1995) or other modalities of treatment.

## CONCLUSION

In conclusion, we have described an unusual and rare complication of adenoidecotmy. Both clinically and histologically it confronts the otolaryngologist with a diagnostic challenge. Inflammatory pseudotumor should be added to the list of possible complications of adenoidectomy. From similar lesions detected at other anatomic sites conservative complete surgical excision seems to be the best therapeutic option.

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Usamah Hadi M.D., F.A.C.S
Department of Otolaryngology,
Head and Neck Surgery
C/O American University of Beirut,
850, 3rd Ave, 18th Floor
New York, NY 10022-6297
USA

Tel: +1-961-1-350000, Ext: 5470,
Fax: +1-961-1-7444464,
E-mail: hb03@aub.edu.lb