Subglottic inverted papilloma: Case report and literature review

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Abstract
Inverted papilloma is a benign tumor derived from the Schneiderian epithelium. It usually originates in the nasal cavity or paranasal sinuses. We report a rare case of an atypically located subglottic inverted papilloma diagnosed in a 4-year-old boy. A literature review revealed that this tumor has never been reported in the subglottic region or in a patient as young as 4 years old.

1. Introduction

Sinonasal papilloma is a rare, benign neoplasm, which originates from the Schneiderian membrane in the nasal cavity. The first case of sinonasal papilloma was described by Ward in 1854. These papillomas are referred to as “inverted” because they grow endophytically toward the underlying stroma. This pattern of growth was first described by Ringertz in 1938. Inverted papillomas (IPs) constitute only 0.5–4% of all nasal tumors.1 They typically affect men between 40 and 70 years of age, the male to female ratio is 4:1, and these tumors are rare during childhood and adolescence. The papilloma is usually located on the lateral nasal wall, especially in the middle meatus. Inverted papillomas are characterized by frequent recurrence and a potential for malignancy.2 Although it is rare for IP to originate outside the sinonasal cavity, but it was reported in the literature in unusual sites. We report a unique case of an atypically located inverted papilloma in an unusual age group.

2. Case presentation

A 4-year-old boy complaining of biphasic stridor and shortness of breath was presented to the emergency department. His parents had noticed a slowly progressive noisy breathing starting at the age of 2 years. This was initially mild, and it gradually increased in severity, until it became significant one month prior to his presentation. There was no dysphonia or aspiration, and he had no prior history of endotracheal intubation. Physical examination showed that his temperature was 36.5 °C, but he was tachypneic, stridulous and had suprasternal retraction. The child was admitted to the pediatric intensive care unit. His condition did not require intubation, and he was kept on humidified oxygen mask. Radiological studies
in form of chest X-ray was irrelevant. Blood work and ABG were normal. As the distress persisted, the patient was transferred to the operating room, where bronchoscopy revealed an isolated, pedunculated mass arising from the anterior subglottic wall. It was occupying about 50% of the lumen. The mass was excised completely and sent for histopathological evaluation. The final diagnosis was inverted papilloma (Fig. 1a and b). The patient had an uneventful postoperative course, showing marked improvement in breathing. A 6-months follow-up showed no recurrence.

3. Discussion

Inverted papilloma is a true tumor arising from the Schneiderian mucosa. Nasal papillomas can be classified according to their pattern of growth as endophytic (inverted papilloma), exophytic (fungiform papillomas), or cylindrical cell papillomas. IPs are primarily located in the nasal cavity or paranasal sinuses. The most characteristic features of this tumor are its tendency to recur, its destructive capacity, and its propensity for association with malignancy.3 The typical histopathological picture is that of a multilayered epithelium inverted into the underlying edematous stroma. Squamous cell epithelium, transitional cell epithelium and cylindrical cell epithelium are all frequently found, and the basement membrane is usually intact. The stroma is edematous and highly vascularized, and there is often an inflammatory cell infiltration. The etiology is uncertain, but a viral etiology has been suggested by many authors. Many subtypes of human papilloma virus (HPV) have been isolated from the surgical specimens. Papovavirus has also been detected in IP specimens.4

IP typically involves the lateral nasal wall and middle turbinate and at least one paranasal sinus. The most commonly involved sinuses are the maxillary and ethmoid, followed by sphenoid and frontal sinuses. These tumors have a high recurrence rate, reported to be up to 75%. This could be a result of either multicentricity or incomplete excision during the first treatment session. The incidence of the development of squamous cell carcinoma within the IP is 5–15%.5 IP is diagnosed by a combination of history, examination, and radiological studies, but a definitive diagnosis requires biopsy examination. CT scan is the radiological modality of choice for the diagnosis of IP. Typical radiological findings are unilateral opacification of the maxillary or ethmoid sinus, with nasal cavity involvement. The treatment is total excision, performed either by lateral rhinotomy via a midfacial degloving approach, or recently, via an endoscopic approach.

Lowen et al. (1995) reported cases of IP treated surgically, followed by postoperative radiotherapy if the IP was associated with malignancy, or when it was not completely excised. Postoperatively, a watchful follow-up is highly recommended due to the frequency of recurrence and the association of this tumor with malignancy.

Hathiram et al. (1990) reported an IP in a 50-year-old man who unusually presented with dyspnea and dysphagia due to a huge lesion extending from the right nasal cavity to the oropharynx and the laryngopharynx. Guillemaud et al. (2009) reported nine patients (out of 77 patients included in the study) with IP originating in the sphenoid sinus. Furthermore, Cho et al. (2008) described a 15-year-old boy with an isolated sphenoid IP. Kosugi et al. (2008) also reported an isolated IP in the sphenoid sinus, which did not involve the sphenoid sinus, in a 40-year-old man. In 2007, Iwata et al. reported a relatively unusually located IP, in a 74-year-old man. This patient had a pulmonary (right lower lobe) IP, which was associated with an elevated carcinoembryonic antigen (CEA) level and squamous cell carcinoma-associated antigen. In 2009, Raemdonck et al. reported the case of a 24-year-old woman who presented with a right lacrimal fossa mass. Her pathology report following resection of the whole lacrimal sac revealed the presence of an inverted papilloma (IP) with a sharp transition between the papilloma and the normal lacrimal duct epithelium.

There are few reports in the literature on IPs in the temporal bone and the middle ear. In 2010, Kainuma et al. described a case of a 65-year-old man with inverted papilloma of the middle ear, which was surgically resected by radical tympanomastoidectomy. The patient underwent postoperative radiation therapy.

In conclusion, although rare, diagnosis of an inverted papilloma can be considered in the differential diagnosis of a subglottic benign lesions.

References