Regional spread of recurrent respiratory papillomatosis to bilateral cervical lymph nodes
Hien T. Tierney, MD; Harrison W. Lin, MD; Eric H. Holbrook, MD; Ramon A. Franco MD; Ben Z. Pilch, MD, PhD; Derrick T. Lin, MD
Dept. of Otolaryngology, Division of Head and Neck Surgery, Massachusetts Eye and Ear Infirmary, Boston, MA
Dept. of Pathology, Massachusetts General Hospital, Boston, MA

OBJECTIVES:
- To present a rare case of regional spread of benign recurrent respiratory papillomatosis (RPR) to bilateral cervical lymph nodes

INTRODUCTION

Recurrent respiratory papillomatosis (RPR) is a chronic disease that affects both pediatric and adult populations. It is caused by human papillomavirus (HPV) types 6 and 11 and is characterized by a benign proliferation of squamous epithelium along the upper aerodigestive tract. The disease is classified into juvenile versus adult onset and has a clinical course that may be benign or aggressive. Benign disease is usually limited to the larynx and is characterized by few or no recurrences. Aggressive disease is characterized by epithelial atypia and multiple recurrences with disease spread to other sites within the upper aerodigestive tract. The glottic larynx is considered the most common site affected, however, RPR may involve any site along the aerodigestive tract including the oral cavity and soft palate. Although there is a risk of malignant transformation in RPR, it is considered a benign disease. Similarly, papillomas found within the nasal cavity and, by extension, the paranasal sinuses are considered viral-related papillomas where the benign squamous proliferation involves the Schneiderian epithelium of the nasal cavity. As with RPR, Schneiderian papillomas are benign tumors with a predilection for local recurrence. Schneiderian papillomas are divided into fungiform, inverted and cylindrical types based on the specific architectural patterns seen histologically. As with RPR, Schneiderian papilloma is a benign condition and is not known to metastasize in the absence of malignant transformation.

We present a case of benign recurrent papillomatosis involving the upper aerodigestive tract with regional metastases to bilateral cervical lymph nodes. A review of the literature reveals this case to be only the third reported case of benign papillomatous metastasis to the neck.

CASE REPORT

A 47-year-old man presented to our clinic with a longstanding history of adult-onset recurrent respiratory papillomatosis involving his nasal cavity, nasopharynx, oral cavity, oropharynx and tongue. He had previously undergone multiple excisions of his lesions with pathology consistent with Schneiderian papilloma-type lesions with no evidence of malignancy. He presented with a new 3 x 2 cm right level II neck mass which had been present over the last few months. The mass was asymptomatic but had slightly increased in size over the previous few months. The patient was taken to the operating room where he underwent direct laryngoscopy with excision of the right neck mass.

Intraoperatively, he was noted to have typical-appearing papillomas along his palate and posterior oropharyngeal wall without extension into the larynx. The right neck mass was noted to be well-encapsulated with a soft, yellow cheese-like material within it. Pathology results of the right neck mass were consistent with a lymph node containing cystadenomatous foci and epithelium resembling exophytic or fungiform Schneiderian papilloma. There was no evidence of carcinoma.

Polymerase chain reaction studies of the lymph node revealed the presence of HPV type 11. Of note, four years prior, the patient had undergone a lymph node excision on the contralateral neck with pathology consistent with benign papillomatosis.

DISCUSSION

While both respiratory and Schneiderian papillomatosis are considered benign conditions, they are characterized by high rates of recurrence and less frequently, by malignant transformation. Our report highlights a case of regional metastases of cytologically benign papillomatosis to bilateral cervical lymph nodes in a patient with recurrent sinonasal, oropharyngeal, laryngeal and oral cavity papillomas. In 1973, Schoub et al described a case of inverted-type Schneiderian papilloma arising in the left ethmoid sinus which subsequently metastasized to the left neck. Excision of three lymph nodes in this case demonstrated well-differentiated inverted papilloma with no histologic features to suggest malignant transformation. Likewise, Fechner and Sessions in 1977 described a patient with inverted papilloma of the lacrimal sac who presented with cervical metastases containing inverted papilloma. In this case, the patient underwent medial maxillectomy and excisional biopsy of the neck metastases and again, benign inverted papilloma was seen in both specimens. In their discussion, Fechner and colleagues hypothesize that the neck metastases may have represented surgical seeding of inverted papilloma during previous biopsy procedures or that they were, in fact, independent inverted papillomas arising in cervical branchial cleft cysts. The latter hypothesis arose from the fact that branchial cleft cysts contain both a ciliated and squamous epithelium which could, in theory, have given rise to benign inverted papilloma. The authors conclude, however, that both these cases represented “benign metastasis” of papilloma to cervical lymph nodes where lymphatic transport of benign neoplastic tissues led to complete destruction of the lymph node architecture. In the absence of histologic evidence of carcinoma, these authors conclude that cervical extension of inverted papilloma does not represent true malignancy.

Our patient presented with multiply recurrent upper aerodigestive tract papillomatosis and a new level II neck mass. After excisional biopsy and careful histopathologic examination of the neck mass, a diagnosis of benign papilloma was made. The neck mass consisted of a large lymph node with multiple epithelial cysts of various sizes lined predominantly by respiratory-type ciliated columnar epithelium (Figure 2A). The cysts contained focal epithelial invaginations and papillae. Amorphous proteaceous material admixed with degenerating neutrophils was present in cyst lumens. This portion of the lesion, which predominated, was reminiscent of a Warthin’s tumor (papillary cystadenoma lymphomatosum), except that the epithelium was respiratory rather than oncocytic.

In focal areas, the cysts contained papillomatous epithelium closely resembling Schneiderian papilloma (Figure 2C). The epithelium contained foci suggestive of HPV viropathic effect, i.e., focal multinucleated epithelial cells, delay in maturation at the surface, disturbance of nuclear polarity and focal perinuclear cytoplasmic clearing.

CONCLUSIONS:
- A rare case of benign papillomatosis spreading to cervical lymph nodes is described. These findings likely represent “benign metastases” of papilloma via lymphatics to cervical lymph nodes and should not be regarded as malignancy. Regional metastases of papillomas should be considered in any patient with recurrent papillomatosis and a new-onset neck mass.

REFERENCES


FIGURE 1: Clinical exam findings

(A) View of soft palate respiratory papillomas through the rigid zero-degree endoscope (T-tongue). (B) Sinonasal papillomas abutting the middle turbinate (MT) and nasal septum (S).

FIGURE 2: Histopathology

(A) At low magnification, the most recently excised lymph node and the contralateral lymph node excised four years prior (B) are seen to contain multiple cysts of various sizes.

(C) A papillomatous focus has features of Schneiderian papilloma which was also seen on the patient’s previous lymph node excision (D).

(E) At higher magnification, the papillomatous epithelium is histologically benign transitional type with ciliated columnar cells as is commonly seen in Schneiderian papillomas.

(F) High magnification shows features typical of HPV viropathic effect, i.e., focal multinucleated epithelial cells, delay in maturation at the surface, disturbance of nuclear polarity and focal perinuclear cytoplasmic clearing.