Bilateral Cavernous Sinus Involvement by Perineural Spread of Inverted Papilloma

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INTRODUCTION

- Inverted papilloma generally is a benign tumor of respiratory epithelium that involves the nose and paranasal sinuses, but can extend into the orbit, nasolacrimal duct, pterygoid fossa, and anterior cranial fossa.
- We describe the first reported case of a hybrid inverted papilloma with benign and malignant elements that extended by retrograde perineural spread to produce bilateral cavernous sinus invasion with further perineural spread along the third division of the trigeminal nerve into the masticator space and along the skull base.

CASE REPORT

A 53 year old woman first presented to the Mount Sinai Medical Center with a history of undergoing a right medial maxillectomy for an inverted papilloma nine years previously at another hospital. She remained asymptomatic, until 2001, when epistaxis reappeared and a recurred tumor was suspected. The lesion involved the right maxillary sinus and extended transnasally through the nasal septum into the left maxillary sinus. She underwent resection of the tumor through a a degloving procedure. The specimen showed the microscopic presence of diffuse high grade dysplasia and she underwent a course of radiation therapy to the maxillary and ethmoid sinuses and nasal cavity.

The patient continued to experience recurrences and underwent six procedures over the next five years for recurred tumor on the residual septum, hard and soft palate, and left lateral nasal wall. These entailed endoscopic resections as well as a palatectomy, in 2005, and a left lateral rhinotomy, in 2006. All specimens showed benign inverted papilloma with mild to moderate dysplasia.

Multiple small recurrences continued to be detected on follow up endoscopic surveillance, with CT scans showing only post operative changes. However, in February 2007, she complained of left mid-facial numbness and a MRI revealed an enlarged and enhancing intraorbital mass, which extended into the pterygopalatine fossa and then to the maxillary nerve to where it entered the foramen rotundum. The resected nerve revealed benign inverted papilloma with mild dysplasia but with extensive perineural extension. Over the next month, there evolved a complete left cavernous sinus syndrome with ptosis and a fixed proptotic and chemotic eye. At this time, a small focus of tumor remained in the left nasopalatine and on the soft palate, which on biopsy revealed inverted papilloma without dysplasia.

In the period of May to July 2007, she underwent re-radiation and three cycles of cisplatin and one of cetuximab with some temporary improvement of her ophthalmologic complaints, which soon returned. An MRI in September 2007 showed increasing fullness in the left cavernous sinus where it extended to Meckel’s cave and then into the third division of the trigeminal nerve. A full body PET-CT scan revealed hypermetabolic lesions (SUV range K-18) in the nasopharynx, oropharynx, and bilateral maxillary alveolar ridges. A biopsy revealed inverted papilloma without dysplasia. An MRI taken in December 2007 revealed further extension on the left side into the masticator space. By March 2008, there was enlargement and enhancement of the right maxillary nerve and right cavernous sinus. In July 2008, the patient was again began on cetuximab. However, by December 2008, MRI revealed involvement of the right pterygopalatine fossa, the right cavernous sinus, and both mandibular nerves as well as increased involvement of V1, the sphen and the right temporal lobe. Disease extending along the left stylohyoid muscle invaded the left facial paralysis, with the patient also experiencing increasing trismus and slowly progressive proptosis of the right eye.

DISCUSSION

- Inverted papilloma is a benign sinonasal lesion arising principally on the lateral nasal wall, with extension into the paranasal sinuses. Malignant transformation to carcinoma is estimated to occur in 7-10 percent of the cases, which may be synchronous or metachronous.
- Surgical management is based on the tumor extent and may require an endoscopic and/or external procedure for complete extirpation.
- Radiation therapy in the treatment of inverted papilloma may be beneficial for a small group of patients with aggressive tumors that are multiply recurrent, surgically unresectable, and are associated with malignant transformation.
- Our patient developed multiple recurrences following a full course of regional radiotherapy, and only transient improvement after a second course.
- In all series, tumor extension beyond the sinonasal complex into the anterior cranial fossa, pterygoid fossa, retrobulbar region, and nasopharynx. Tumors may also invade intracranially, although this is a rare occurrence in the absence of malignancy.
- Perineural spread of head and neck cancer is a well-described complication of squamous cell carcinoma, lymphoma, and carcinoid tumor. However, it has never been described in inverted papilloma.
- Perineural extension most commonly involves the branches of cranial nerve (CN) V. With the mandibular division, spread is to the cavernous sinus from tumors of the nasopharynx, masticator space, and parapharyngeal space. Along the maxillary division, tumors extend from the palate, maxilla, nose and nasopharynx, and with the ophthalmic division, spread is from the orbit, forehead, and frontal sinus.
- In this patient case, we believe the tumor spread along the maxillary division and invaded the left cavernous sinus from the right orbit into the right cavernous sinus. Further retrograde perineural extension was to the Gasserian ganglion in Meckel’s cave and laterally along the mandibular nerve.
- This presentation is the first reported instance of this form of extension of inverted papilloma in the literature.

CONCLUSIONS

- Inverted papilloma, despite being a benign tumor, is locally invasive, has a propensity to recur, and can undergo malignant transformation.
- Management is by wide surgical excision and close follow-up.
- The disease history in the present case was over 19 years.
- In the case described, treatment with multiple endoscopic and external resections in addition to adjuvant radiotherapy and chemotherapy, was unsuccessful in arresting its progression with multiple recurrences and perineural spread into both cavernous sinuses.
- Perineural spread and recurrences at multiple separate sites developed with microscopically benign disease.

REFERENCES


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