Presentation of Childhood Inverted Papilloma with an Antrochoanal Polyp

Case Report
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Abstract
Inverted papilloma is a benign tumor that is characterized by invagination of nasal and paranasal sinus mucosa epithelium into the stroma. It often originates from the lateral nasal wall and is mostly seen among males between 50 and 60 years old. We presented a 14-year-old boy who applied with the complaint of single-sided nasal obstruction and was operated on due to the diagnosis of an antrochoanal polyp. Postoperative pathology revealed the presence of inverted papilloma. It is suggested that inverted papilloma should be considered in childhood nasal lesions.

Key Words: Antrochoanal polyp, inverted papilloma, childhood

Introduction
Inverted papilloma is one of the most frequently seen benign lesions that originate from nasal and paranasal sinus mucosa epithelium stroma (from the Schneiderian membrane). Its incidence rate is 0.5–1.5 per 100,000 people in a year. It often increases at the age of 50–60 years, and the proportion of males to females is 3/1 (1). In its histology, the epithelium, inverted into connective tissue, is observed, and it is generally an exophytic and polypoid lesion. Different from inflammatory polyps, it is pinky and has a vascular appearance macroscopically, and it does not have goblet cells, koilocytes, or eosinophils in its structure microscopically (2). Aggressive surgery treatment is suggested by some authors when the risk of malignancy is considered. Antrochoanal polyps are solid benign lesions displaying unilateral localization, not presenting with allergic predisposition, originating from the maxillary sinus mucosa, and extending into the choana first and then the nasopharynx. It is commonly seen among children. It is treated with surgical excision covering the tissue from which it originates (3). In our case, the postoperative pathology result of the pediatric patient who had been operated on due to the pre-diagnosis of antrochoanal polyp was found to be consistent with inverted papilloma. Based on this case, the diagnosis, treatment procedure, and literature were interpreted for this disease, which is rare in childhood.

Case Report
A 14-year-old boy was admitted to our clinic with the complaint of nasal obstruction. The patient, with no additional pathology in his background and family history, underwent an endoscopic examination, and a polypoid lesion, localized in the left nasal cavity and extending from the left middle meatus into the nasal passage, was found. Paranasal computed tomography revealed a soft tissue density mass originating from the left maxillary sinus ostium and spreading into the nasal passage, in consistency with antrochoanal polyp (Figure 1). For the patient with a pre-diagnosis of antrochoanal polyp, the lesion was totally excised by administering functional endoscopic sinus surgery under general anesthesia and the left Caldwell Luc method (Figure 2). The patient was discharged from the hospital without any complication on the 2nd postoperative day. The postoperative pathology result was evaluated as inverted papilloma. No local recurrence was observed at the postoperative endoscopic follow-up sessions. Paranasal computed tomography performed in the 3rd postoperative month did not reveal a bone defect at the level of the left maxillary sinus hiatus secondary to the operation or the finding of recurrence (Figure 3). Before preparing this case report, written informed consent was obtained from the patient’s parents.

Discussion
Inverted papilloma is a benign lesion that originates from nasal and paranasal sinus mucosa epithelium stroma, but its etiopathogenesis is not known exactly. Allergy, chronic rhinosinusitis, air pollution, smoking and smoke exposure, and HPV (HPV 6-11-16-18) infection are accused of inverting papilloma (2, 4). Since inverted papilloma displays unilateral localization in the nasal cavity, a differential diagnosis with other space-occupying masses should be performed for children. These masses include antrochoanal polyps, encephalocele, hemangiomas, mucocle, and squamous papilloma (2).
In contemporary literature, the number of published case reports on inverted papilloma in childhood and/or adolescence is lower than 20. It was first reported by Eavey (5), who made a classification for childhood and adolescence according to 3 patients at the ages of 6, 12, and 20 years. The disease recurred in 2 patients and was treated with surgical operation. Moreover, Özcan et al. (4) reported that a 9-year-old pediatric patient was treated due to recurrent inverted papilloma.

In comparison to inverted papilloma cases in adulthood, the clinical behaviors of inverted papilloma cases in childhood cannot be predicted because of an insufficient number of cases. Since the basal membrane in these lesions generally protects its intact structure, they do not display surrounding invasion, like malignant lesions. They can lead to remodeling and destruction in the paranasal region, depending on compression and mass effect. A malignant tumor (squamous cell carcinoma) can occur as a separate lesion within inverted papilloma or a postresection metachronous lesion at a rate of 10% (6). For confirmation of the diagnosis and identification of the appropriate treatment method, preoperative and postoperative tissue samples should be taken from the lesion (2). Since our patient was a child and there were no risk factors for malignancy, tissue punch biopsy was not performed for confirming the diagnosis before planning the surgery. Frequent follow-up sessions were planned, because the result of postoperative pathology revealed the presence of inverted papilloma.

Of the imaging methods, paranasal computed tomography, performed in the axial and coronal plane, is the most valuable method due to its common use and the data obtained from it. It is useful, for it provides information about the spread of disease and anatomic features to the patient. Magnetic resonance imaging is beneficial for the differentiation of mucus and polyp and for the differential diagnosis of formations, such as infection, tumor, mucocele, and encephalocele (2). In the case report presented, the patient was evaluated with preoperative computed tomography, and data on anatomic structures were obtained apart from endoscopic images.

Considering the risk for malignancy, aggressive surgical treatment practices have changed in time. Open medial maxillectomy technique or mass excision technique has been replaced by endoscopic sinus surgery. Endoscopic medial maxillectomy, sublabial incision technique for reaching the lateral and anteroinferior walls of the maxillary bone (Caldwell Luc), external frontoethmosphenoidectomy, and removal of invaded sites of the bone with diamond burr in the case of malignancy have been described (2). Özcan et al. (4) reported that the case in their study was treated by sublabial incision following endoscopic transnasal resection.

Recently, Buiret et al. (7) conducted a study with 385 adult patients, and they have suggested that radiotherapy should be administered after primer surgery treatment in the cases with inverted papilloma displaying degeneration or in the presence of proven coexistent squamous cell carcinoma. Since the long-term effects of radiotherapy in childhood are not known, it is not recommended as a treatment method.

Regular endoscopic examination is suggested due to the possibility of the disease's recurrence in 1–24 years and potential malignant transformation. In the case of endoscopic suspicion, imaging methods should be employed (2). Eavey (5) reported a 20-year-old female patient having inverted papilloma with co-existent low-grade squamous cell carcinoma. In the case report.
presented here, no recurrent and residual lesion was found in endoscopic examinations performed during the post-operative 18-month follow-up. In the 3rd post-operative month, anatomic structures and the presence of recurrent-residual mass were investigated by paranasal computed tomography.

In order to perform surgical treatment perfectly in light of the data obtained through pre-operative comprehensive endoscopic examination and imaging methods, biopsy can be required to confirm the final diagnosis, especially for unilateral nasal cavity lesions.

**Conclusion**

In the differential diagnosis, it should be kept in mind that patients, even in childhood, having antrochoanal polyp, which is frequently seen during otorhinolaryngologic practice, may have inverted papilloma.

**Informed Consent:** Written informed consent was obtained from patients who participated in this case.

**Peer-review:** Externally peer-reviewed.


**Conflict of Interest:** No conflict of interest was declared by the authors.

**Financial Disclosure:** The authors declared that this case has received no financial support.

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