Rare and unusual benign tumors of the sinonasal tract and pharynx: Case series and literature review

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Received December 8, 2021; Accepted February 25, 2022

DOI: 10.3892/etm.2022.11263

Abstract. There are a number of benign tumors of the nose and pharynx that are seldomly reported in literature but that can sometimes prove difficult to treat and extremely important for differential diagnosis. The present study presents cases of rare benign tumors localized in the pharynx, nasal and sinus cavities, as well as reviews of literature and historical references for each type of tumor. Unilateral nasal hemangioma in a 72-year-old male which, although not a rare pathology, raised problems due to auto-resection of the tumor. The surgeon was able to pull it out with ease without bleeding; it is possible that the mass would have eventually fallen out. Pilomatrixoma is a relatively uncommon ectodermal benign tumor of the skin derived from hair matrix cells. Surgical resection is curative but recurrence is possible (≤5% risk). The presented case is of a 26-year-old female with a pilomatrixoma of the left cheek who, for aesthetic reasons, refused a classical external surgical approach. Trans-oral resection was performed, which proved feasible but laborious and prone to recurrence. Inverted Schneiderian papilloma is a rare benign tumor of the nasal and sinus cavities with increased potential for invasion, recurrence and malignant transformation compared with other types of papilloma and other benign tumors of the area. The tumor represents 0.5-4.0% of all nasal tumors and has been described under different names, such as villiform cancer and cylindrical/transitional papilloma. The present study reports a rare case of bilateral papilloma in a 68-year-old male. He presented with bilateral evolving nasal obstruction and hyposmia. Following surgery, the patient was treated by a multidisciplinary team and followed by a respiratory rehabilitation program.

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Key words: case report, nasal cavity, benign tumor, cavernous hemangioma, epistaxis, surgical treatment, benign pilomatrixoma, inverted papilloma, trans-oral approach, ghost cells, social insertion, differential diagnostic

Introduction

The anatomic area represented by the nose, paranasal sinuses and pharynx is frequently the site of appearance for tumors (benign or malign) of various histopathologic origin. Some of these tumors are, however, rare and unusual and require special attention in diagnostic and therapeutic management. All cases presented are of uncommon benign tumors of the nose, sinus or pharynx associated with a high risk of changing from benign to malignant and of unclear etiology. Environmental, and clinical risk factors cannot always be ruled out and the clinician must always consider that genetic predisposition is commonly augmented and complicated by environmental factors (1).

Hemangiomas are benign tumors that originate from vascular endothelial proliferation. They are relatively common in the head and neck (>50%) but rare in the nasal cavity and paranasal sinuses and can originate from vessels in numerous types of tissue, such as the skin, mucosae, bone, muscle and glands (2). The nasal cavity is occasionally the site of appearance (2); hemangiomas represent ~20% of all benign tumors of the nasal cavity and of these, 65% are located on the septum, 18% on the lateral wall and 16% in the vestibule (3). They typically arise from soft tissue (skin, mucosa, vessels) and although they may cause bony changes or destruction, rarely arise from bone (4). Although rare, this tumor must be considered in the differential diagnosis of intra-nasal bleeding mass (bleeding polyps of the septum, angio-fibromatous polyp) (5).

Capillary hemangiomas are more frequently observed than the cavernous type. The capillary type is more frequently associated with the nasal septum site whereas the cavernous

type appears more frequently on the lateral wall of the nasal cavity (5).

Pilomatrixoma, also termed calcifying epithelioma of Malherbe or Epithelioma calcificans Malherbe, is a relatively uncommon benign tumor of the skin derived from the hair matrix cells. In 1880, when it was first described by Malherbe and Chenantais (6) it was believed to arise from sebaceous glands (7) but in 1961, Forbis and Helwig (8) discovered its origin in hair matrix cells and proposed the term pilomatrixoma to avoid a connotation of malignancy. Similarly, to the other two rare tumors in the present article, the tumor commonly (but not exclusively) occurs in children as a hard subcutaneous nodule or cyst with unremarkable overlying epidermis 0.5-3.0 cm in size, with the largest reported case at 24 cm (9); it is typically located on the scalp, face and upper extremities. Excluding lymph nodes, it is the second most excised superficial mass in children after epidermoid cysts (10). However, pilomatrixomas can be easily misdiagnosed and/or missed in differential diagnosis (6). Clinical findings will aid in an accurate diagnosis. Surgical removal is curative but incomplete excision can lead to recurrence, although rare. Malignancy has been rarely reported (6). The tumor is also relatively frequent in dogs; Kerry Blue and soft-coated Wheaton Terriers, standard poodles and Old English sheepdogs exhibit increased susceptibility (11).

Inverted Schneiderian papilloma has been considered the best term to describe the tumor properties of inversion, location, and distinctiveness of character (12). The first to describe it was Ward in 1854 but Billroth was credited with describing the first true papilloma of the nasal cavity and called it villiform cancer (13). Other names such as fungiform papilloma, cylindrical or transitional papilloma have also been used in literature. Papillomas are rare benign tumors originating from the Schneiderian respiratory membrane and can be classified into three distinctive types: Exophytic, oncocytic and inverted papilloma (14-16). The tumors are locally aggressive (bone destruction), have a high recurrence rate if partially removed and exhibit a tendency for malignant transformation into squamous cell carcinoma (likelihood, ≤20%) (17). The age of the patients may vary from 10 to 87 but the majority of cases present at 50-70 years with a male:female ratio of 3.3:1 (18). The etiology remains controversial but factors such as human papillomavirus infection, chronic inflammation, allergy, occupational pollution (sulfur, tobacco) are considered key (15,16,19). The recommended treatment includes complete surgical resection and life-long follow-up for potential recurrence (16,20-23).

The present study aimed to present the authors' experience in treating this type of pathology as well as reporting on curious turn of events that these tumors can take (e.g., auto-resection of hematoma) with the hope that it proves useful to other ENT-HNS professionals.

Materials and methods

The present study was approved by Research Ethics Committee of the Faculty of Medicine, Titu Maiorescu University (approval no. 6/21.09.2021; Bucharest, Romania). All patients provided written informed consent and approved the publication of their data.

CT evaluation. All tumors were assessed by clinical, imagistic and histopathological examination at the Ilfov County Clinical University Hospital, Bucharest, Romania, between May 2015 and August 2019. The patients underwent complete ear, nose and throat (ENT) examination, complete with endoscopy (where possible). In two of the presented cases, the hemangioma and inverted Schneiderian papilloma underwent classic image evaluation via enhanced computed tomography of the sinus and nasal cavity. The standard protocol was applied: Patient in supine position, scout perpendicular to the hard palate, tube voltage and tube current 125 kV and 80-160 mAs with scan from the hard palate to above the end of the frontal sinus. Scan direction was caudocranial (slice thickness, 0.625-1.000 mm) to obtain axial and coronal images (24).

Ultrasonography evaluation. The pilomatrixoma was assessed by an experienced ENT specialist with ultrasonography competence using an Acuson 128XP scanner (Siemens Medical Solutions) equipped with a 7- to 12-MHz linear array transducer. The longitudinal and transverse scans of the mass were obtained with gray scale and power Doppler ultrasonography. Then two experienced radiologists evaluated the printed images and gave the same description regarding tumor size, shape, margin, echo texture, echogenicity, presence, shape, and amount of calcification, presence of a hypoechoic rim and Doppler flow pattern. Size was defined as the largest tumor diameter. Tumor echo texture was described as homogeneous or heterogeneous, and echogenicity was described as hyperechoic if tumor echogenicity was higher than that of muscle.

Histopathological evaluation. All cases were histologically diagnosed by two independent pathologists, under light microscope at 20, 100 and 200x magnifications, using hematoxylin-eosin staining. Fixation was achieved with 10% formaldehyde buffered solution at room temperature for 24 h. The slicing of wax-embedded material (resected tumor) was performed by ultramicrotome at 3 μm. Staining was performed according to the basic protocol: Dewaxing (xylene 3-5 min); Dehydration (ethanol at 100, 100 and 95% for 2 min each, water wash for 2 min); hematoxylin (pre-prepared solution) staining at room temperature (3-6 min depending on sample size followed by water wash for 1 min); differentiation (mild acid for 1 min and water wash for 1 min); bluing 1 min followed by water wash 1 min, 95% ethanol 1 min and water wash 1 min; eosin, 45 sec, dehydration in ascending alcohol (95, 100 and 100% for 1 min each); clearing (xylene 2-4 min) and cover-slipping (Canada Balsam). No immunohistochemical studies were performed.

Literature review. Literature review was performed using search engines, such as Web of Science, PubMed, NCBI, Wiley Online Library, Sage Journals, Science Direct, Scopus and MEDLINE, using the following key words: Cavernous nasal hemangioma, pilomatrixoma, inverted Schneiderian papilloma, self-resection, trans-oral approach, vascularization, differential diagnosis. Cases that presented pediatric and congenital pathology as well as localization other than the nasal cavity were not included. Inclusion criteria were histological match, same localization, same surgical technique and unusual tumor development.

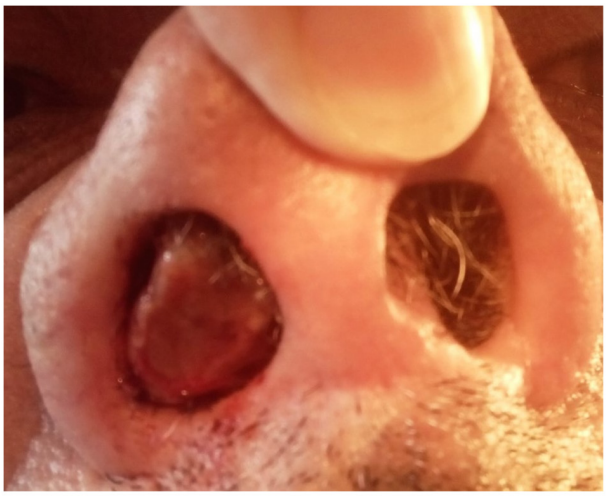

Figure 1. Anterior rhinoscopy (macroscopic appearance of hemangioma). Patient presented with protruding red mass which bled easily when handled.

Case report

Case 1. A 72-year-old male patient presented in May 2015 in the ENT-HNS Department of the Ilfov County Clinical University Hospital, Bucharest, Romania, by referral from the Internal Medicine Department, with a right nasal tumor. The patient experienced intermittent right-sided minimal epistaxis for several months in conjunction with progressive nasal obstruction. The patient presented when the nasal obstruction became total (during the last 2 months prior to admission) and when a tumor protruded out of the right nostril (Fig. 1). A nasal endoscopy examination was not possible since the tumor obstructed the right narina and was clearly visible from the outside. The mass was pinkish-red, necrotic, hard and bled easily when handled. The site of origin in the nasal cavity was not clear. Enhanced axial computed tomography showed a homogeneous, well-circumscribed enhancing mass ~32x17x28 mm in size that filled the anterior part of the right nasal cavity, extending from the lateral wall. The mass had a vascular pedicle on the lateral side and contacted the anterior pole of the inferior turbinate without modifying its osseous structure. The cartilaginous septum was slightly deviated to the left but not perforated. The subcutaneous tissue of the lateral nasal wall was also unaffected. The nasal process of the maxillary bone was close to the mass but unaffected (Fig. 2).

As endoscopic surgical approach was not possible, lateral rhinotomy was planned but as the surgeon started handling the tumor, it came out *en bloc*, much like a polyp, without requiring force and without any bleeding. No incision or resection from the surrounding tissue was necessary. The planned procedure was therefore not performed as the tumor fell out (auto-resection). Since no bleeding was present, the site of implantation of the pedicle was not readily apparent but was hypothesized to be the inferior turbinate. The surgical specimen consisted of a yellow-pinkish, hard, cartilaginous-like lesion; when dissected, it presented a purple-red, clot-like interior (Fig. 3).

Histopathological examination showed large blood-filled spaces lined with flattened endothelium and vessels of different shapes and sizes with areas of oedema and hemosiderinic pigment (Fig. 4). The tumor was a cavernous hemangioma with no sign of malignancy. The patient followed an uneventful post-operative course and was discharged within 3 days. Follow-up at 30 days, 3 months and 1 year revealed no sign of recurrence or residual disease.

Following discharge, the patient was referred to Pulmonary Rehabilitation Clinic of the Iași Clinical Rehabilitation Hospital, where he underwent pulmonary function tests for inclusion in the respiratory recovery program. The patient performed the respiratory rehabilitation program for 2 weeks in the hospital under strict supervision of the rehabilitation team before continuing permanently at home. During the program, the patient performed breathing exercises, coughing, expectoration and exercises to train the muscles of the chest, abdomen, neck, head, limbs and respiratory system. The benefits of the respiratory rehabilitation program were

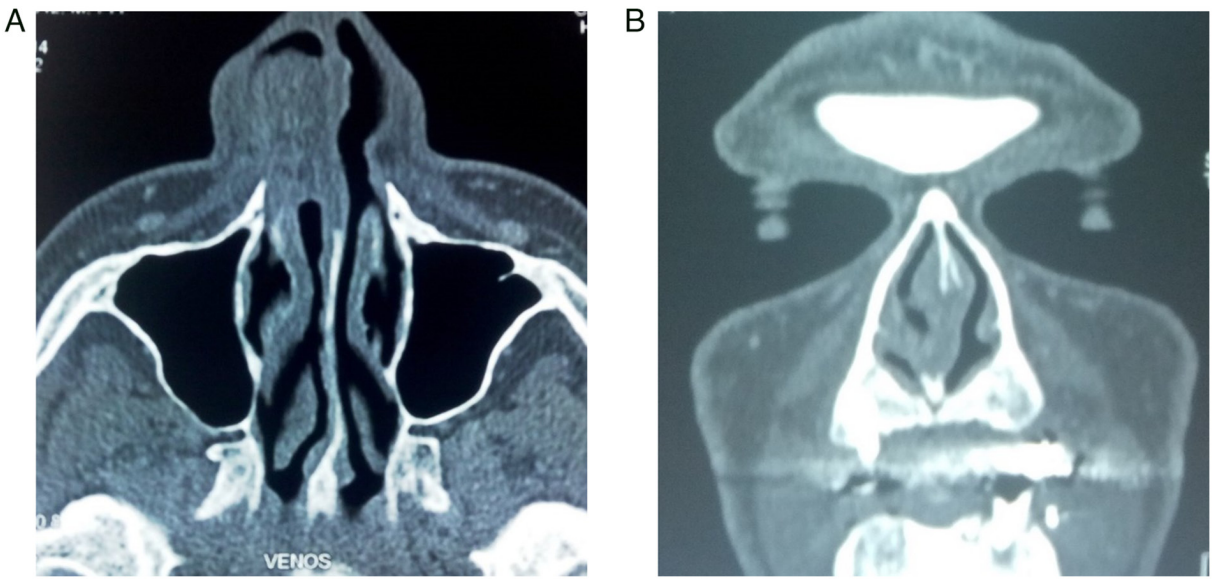

Figure 2. Enhanced CT scan. Well-circumscribed enhancing mass totally obstructed the nasal fossa. (A) axial and (B) coronal section.

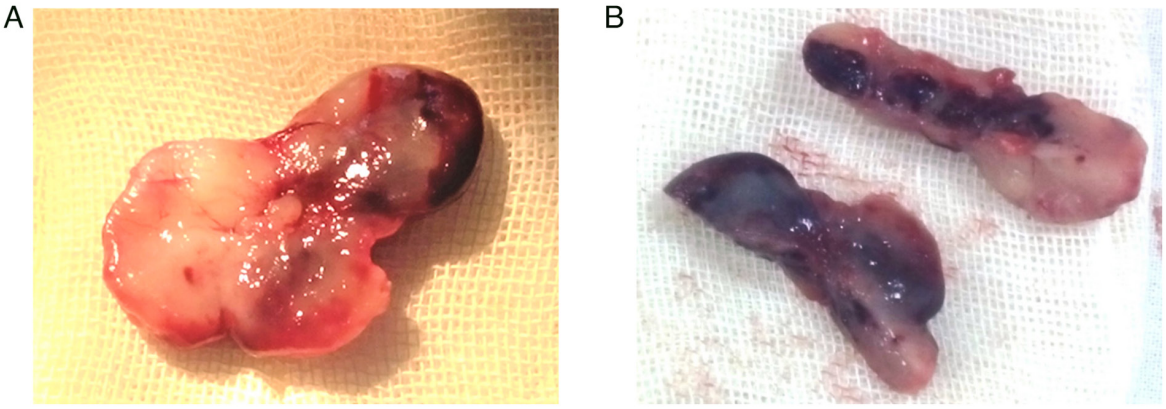

Figure 3. Macroscopic findings of excised cavernous hemangioma. (A) Whole yellow-pinkish, hard, cartilaginous-like tumor. (B) Dissected tumor exhibited a purple-red, clotted interior.

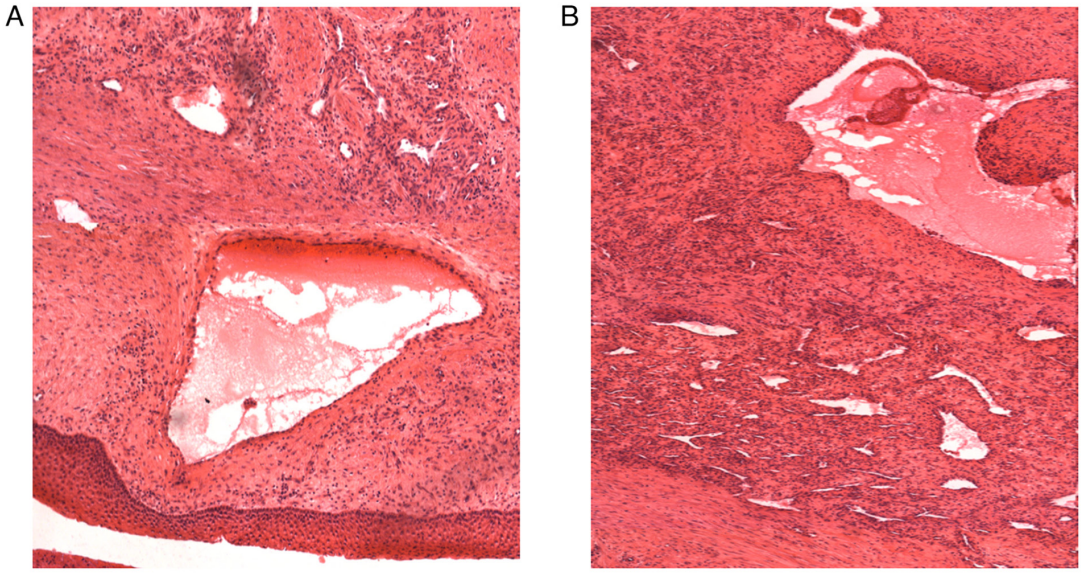

Figure 4. Microscopic findings of excised cavernous hemangioma (Hematoxylin-eosin staining; magnification, x20). (A and B) Cavernous blood-filled structures, lined by a single layer of endothelial cells.

assessed at 3, 6 and 12 months and consisted of increased exercise capacity evidenced by the 6-min walk and oximetry test, decreased symptoms, including anxiety, with rapid return to the social environment. The patient was advised to continue the permanent respiratory rehabilitation program at home, according to an established protocol, with periodic evaluation every 3 months (25).

Case 2. A 26-year-old female patient presented in June 2017 in the ENT-HNS Department of the Ilfov County Clinical University Hospital, Bucharest, Romania, with a hard subcutaneous nodule of the left cheek. The tumor was superficial, mobile over the underlying area, ~1 cm in diameter and exhibited no associated tenderness. It had been present for several years with no progression in size. The ultrasound examination described a superficial, 10.7 mm hypoechoic tumor of the prezygomatic area. Its margins were well defined and it presented no vascular signaling upon Doppler examination (Fig. 5). The patient refused classical external incision to avoid facial scarring. Therefore, a trans-oral approach was performed, which proved to be laborious since the tumor was superficially located. Locating and resecting a

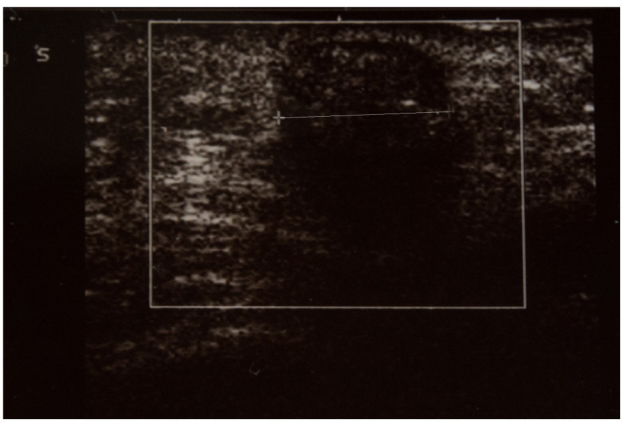

Figure 5. Ultrasound examination of pilomatrixoma revealed an oval-shaped, subcutaneous, well-defined, hypoechoic tumor ~10 mm in diameter.

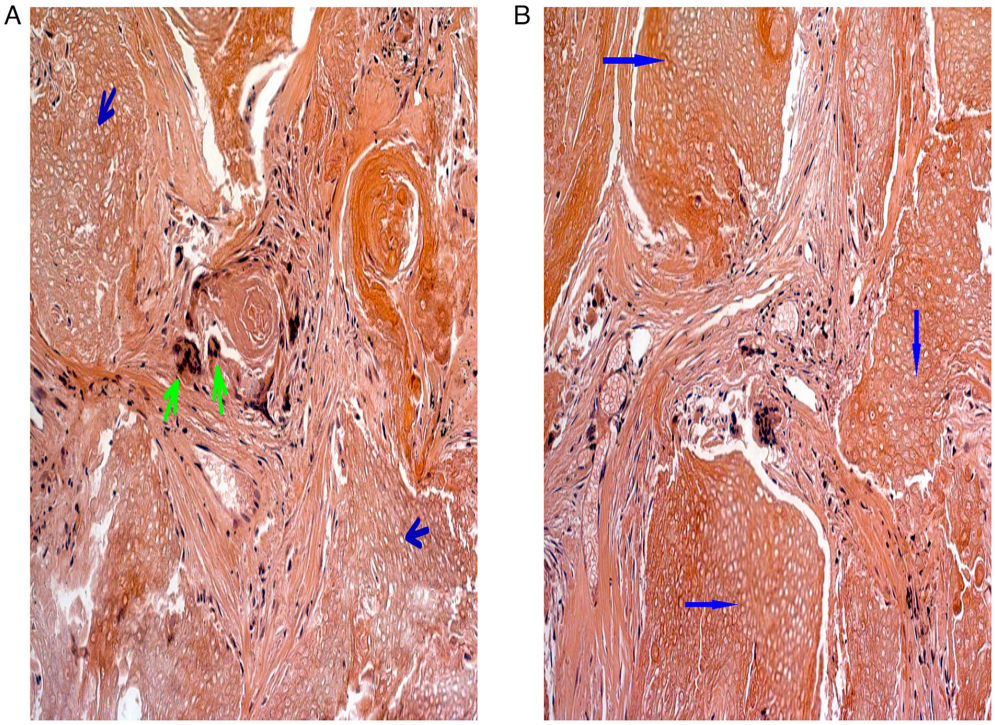

Figure 6. Microscopic findings of pilomatrixoma. (A) Giant (green arrows) and shadow cells (blue arrows) with naked nuclei and abundant cytoplasm. (B) Basaloid cells became larger islands of shadow cells (blue arrows). Hematoxylin-eosin staining; magnification, x20.

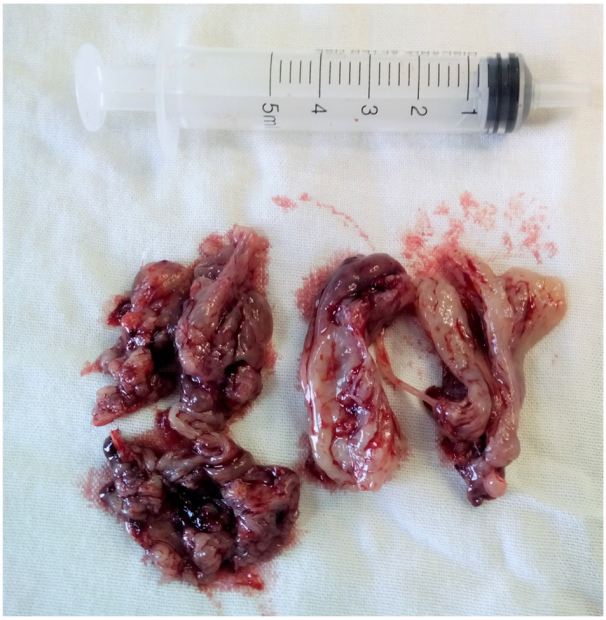

Figure 7. Macroscopic image of excised inverted Schneiderian papilloma (large, reddish yellow polypoid masses with firm, cartilage-like consistency and irregular surface).

mobile tumor trans-orally via mucosa incision is more difficult than via tegument incision and the potential for bleeding increases. Nevertheless, the resection was successful and an oval shaped, hard tumor covered by a well-defined connective tissue capsule and filled with reddish calcification deposits was excised. The patient recovery was uneventful and there were no signs of recurrence at 6 month and 1-year post-operative follow-up. There was also no scarring of the cheek tegument.

The histological examination with hematoxylin-eosin staining showed the typical pilomatrixoma characteristics of shadow or ghost cells with a central unstained area representing the shadow of a lost nucleus. Basaloid cells with an elongated basophilic nucleus and scant cytoplasm at the periphery of epithelial islands were also present, along with calcium deposits and foreign body reaction (giant cells; Fig. 6).

Case 3. A 68-year-old male patient presented in August 2018 in the ENT-HNS Department of the Ilfov County Clinical University Hospital, Bucharest, Romania, with bilateral evolving nasal obstruction, bilateral purulent blood-tinged rhinorrhea, hyposmia and fluctuating headaches and facial pressure. Numerous types of medication, such as antibiotics, anti-inflammatory agents and nasal decongestant, achieved no improvement. The patient was evaluated first at Internal Medicine Clinic of the Ilfov County Clinical University Hospital, Bucharest, Romania, for headaches. The differential diagnosis included arterial hypertension and sleep apnea syndrome. The evaluation protocol included complete blood work, cranio-facial CT and complete cardiological, neurological and ENT examination with endoscopic examination of the nasal fossa. The endoscopic examination revealed large pink-yellowish masses that almost totally obstructed the nasal airways bilaterally. This was confirmed by imaging. The masses were excised under endoscopic control with excision into healthy tissue. Post-operatively, nasal breathing was possible almost immediately after removing the nasal packing, which was kept in place for 48 h, and recovery was uneventful. The patient was advised to attend ENT examinations every

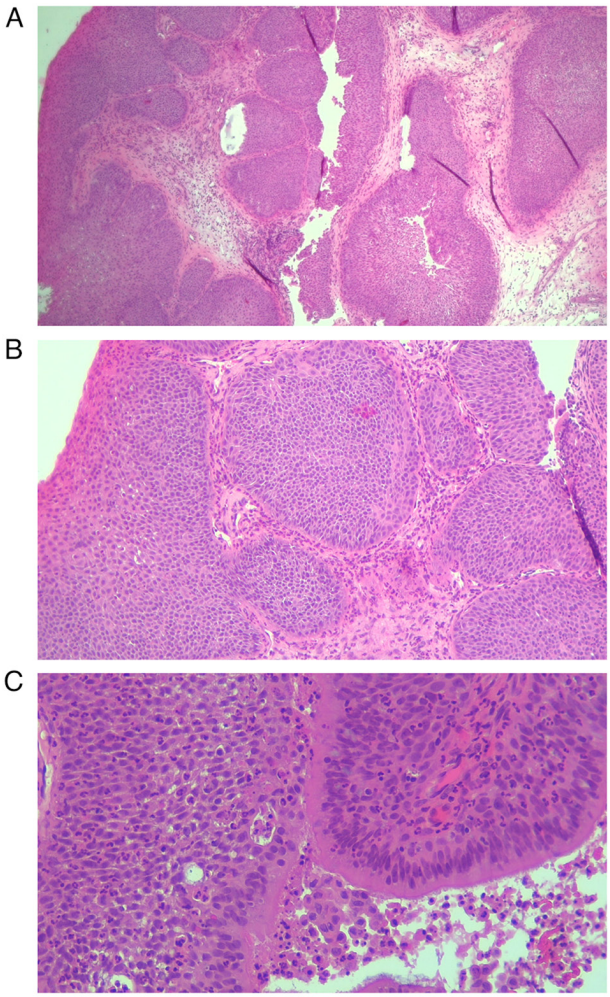

Figure 8. Microscopic images of inverted Schneiderian papilloma (papillary proliferation within the mucosa with connective vascular axis covered by squamous epithelium). Hematoxylin-eosin staining was observed at (A) x40, (B) x100 and (C) x200 magnification.

month for the 2 years and has been symptom-free for the past 5 years. The macroscopic aspect of the polypoid mass was reddish yellow with firm, cartilage-like consistency and irregular surface. The masses were large (≤5 cm in length; Fig. 7). The histopathological examination revealed papillary proliferation within the mucosa with connective vascular axis covered by squamous epithelium. These findings suggested inverted Schneiderian papilloma (Fig. 8).

The patient followed the respiratory rehabilitation program in the same rehabilitation clinic as Case 1, for 2 weeks in the hospital and then permanently at home with evaluation every 3 months and exhibited improvements in quality of life, exercise capacity, decreased of symptoms and fast social reintegration (25,26).

Discussion

Case 1. Hemangiomas of the nasal cavity are rare, benign vascular tumors. The exact etiology is unclear but it has been hypothesized that hemangiomas are a type of tumor since they are destructive and have blood vessels that exhibit tumor-like aspects (27). Alternatively, hemangioma has been described not as a tumor but as a hamartoma or congenital anomaly that may be caused by opening of blood vessels previously closed at birth (28). The proliferation of local blood vessels and increased regional hydrostatic pressure caused by repeated local stimulation affect the occurrence of hemangioma (2).

The histological sub-typing of hemangioma classifies them according to histological appearance as capillary, cavernous, mixed or hypertrophic (5).

Capillary hemangioma, also known as lobular capillary hemangioma or pyogenic granuloma, is the most common type of hemangioma and typically arises from the anterior cartilaginous nasal septum (5). Capillary hemangioma is composed of capillary sized vessels lined with flattened epithelium separated by collagen stroma (2).

Cavernous hemangioma is rare; it occurs on the osseous septum or lateral nasal wall and is composed of large endothelium-lined vascular spaces (2). Cavernous hemangioma of the nose and paranasal sinus is uncommon; most cases arise, as in the present case, from the inferior turbinate. Other structures that support this type of growth are vomer, lamina perpendicularis ossi ethmoidalis and maxillary sinus (5). The incidence of cavernous hemangioma is the same for men and women and the mean age of presentation is 40 years (29). The mixed type of hemangioma exhibits proliferation of endothelium-lined, thin-walled blood vessels of different sizes (2).

The present case included all the aforementioned characteristics of this hemangioma: Unilateral, red or purple, not painful, slowly growing hemorrhagic mass. It also produced progressive nasal obstruction and epistaxis. The tumor was necrotic, which eventually led to auto-resection due to decreased blood supply to the pedicle. To the best of our knowledge, auto-resecting tumor, nasal hemangioma or otherwise, has not been previously reported. The term auto-resection was used to explain an uncommon pathogenic mechanism, which would profit from further study regarding tumoral blood supply. Since no surgical action was taken to resect the hemangioma, the term auto-resection was considered to best describe the unexpected outcome, which was of novel clinical significance and it represent a better solution for an otherwise clear surgical indication. The problem resides in the rarity and unpredictability of such a development. There were no clear signs that the tumor would auto-resect or that vascularization of the area was afflicted in any way. The advantages of auto-resection are that it spares the patient a physically and psychologically traumatic experience, especially in the case of an open approach (lateral rhinotomy or midfacial degloving).

Differential diagnoses of hemangioma include benign (angiofibroma, venous hemangioma, hemangioendothelioma, angiomatous glomus tumor, lymphangioma) and malignant tumors (hemangiopericytoma, hemangiosarcoma, squamous cell carcinoma, adenocarcinoma, metastatic malignancy) of the nasal cavity. The definitive diagnosis is given by histological confirmation.

The tumor typically obstructs the nasal cavity, which makes endoscopic examination impossible, as in the present case. Therefore, CT scan is key in planning the course of treatment. CT scan typically reveals anatomical location and extent of the tumor. The underlying bone is usually normal but may be deformed by adjacent long-term pressure from the

expanding mass (30,31). The present case, like most reports of nasal hemangioma, involved a mass that bled easily when touched. Thus, although biopsy provides key information, the risk of bleeding is high and must be considered. Since the case presented an unusual solution (auto-resection) that involved circulation to the area, angiography and/or magnetic resonance angiography are recommended to diagnose potential vascularization problems and predict auto-resection. Features such as poor vascularization of the mucosa or turbinate area or vascular malformations of the sino-nasal region may suggest eventual auto-resection. However, this hypothesis requires further study.

The treatment of choice for nasal hemagioma is surgical excision. In extensive tumors, the treatment of choice is complete excision with preoperative embolization (32). Other effective methods for treatment of hemangioma include sclerotherapy, cryotherapy, corticosteroid treatment and resection by YAG-laser (33). The surgical approach (midfacial degloving, lateral rhinotomy, trans-palatal and trans-antral approach and LeFort I osteotomy) depends on location and extent of the tumor. The minimal invasive, trans-nasal endoscopic approach has also been suggested by other studies (29,32) but is not always available due to economic reasons, especially in developing countries. The planned surgical option for the present patient was lateral rhinotomy but this was not required.

Case 2. Pilomatrixomas are of ectodermal origin that arise in the lower dermis from the outer root sheath cell of the hair follicle (6,33) and form a connective tissue capsule. A characteristic diagnostic sign is that the tumor slides freely over the underlying area; this has been described as the 'tent sign' as the irregular surface of the mass can be felt by stretching the skin over the tumor (34). There is no associated lymphadenopathy. The skin of the cheek and periorbital area are the most common locations. A blue discoloration of the overlying tegument has sometimes been reported (6). There may also be a history of regional trauma (≤9%) prior to developing the tumor (4,6). The significance of this is yet unknown.

Histologically, pilomatrixomas consist of anucleate squamous (called ghost or shadow cells) with a central unstained area representing a shadow of a lost nucleus, benign viable squamous and foreign body giant cells. These neoplasms exhibit characteristic transition of cells (6). The lining of the cyst consists of basaloid cells with a round or elongated basophilic nucleus and scant cytoplasm at the periphery of epithelial islands (35) that mature into eosinophilic anucleated squamous cells. Calcium deposits and foreign body reaction commonly occur (36) and ossification has been reported (37). Multiple pilomatrixomas have been associated with numerous genetic and non-genetic disorders such as Gardner, Turner and Rubinstein-Taybi syndrome, trisomy 9, Steinert disease, myotonic dystrophy and sarcoidosis (6,37-39).

Pilomatrixoma, which presents as an irregular nodule on the skin, is differentiated from epidermal cysts, which are firm, round and mobile and occur primarily in adolescents and adults, and dermoid cysts, which are firmly attached to underlying tissue. A differential diagnosis should be made with pilomatrix-carcinoma, a rare malign tumor of hair matrix cells and that arises from pilomatrixoma (6). Black *et al* (37) reported that clinical behavior of pilomatrix carcinoma in adults resembles that of basal cell carcinoma in its potential to metastasize. Treatment for the malign tumor is wide local excision (37).

Diagnostic tests and imaging studies are often unnecessary in workup of a superficial, benign skin lesion such as pilomatrixoma (6). However, tests are sometimes performed to exclude diagnosis of malignancy or to determine the depth of a lesion (4). Pilomatrixoma in the parotid or preauricular region may require further imagistic examination and dissection from the parotid gland. Fine-needle aspiration may reveal the presence of ghost and basaloid cells and calcium deposition in the mass, which are diagnostic of pilomatrixoma (40). However, without the presence of ghost cells in the aspirate, the diagnosis may be misleading (40). Ultrasound diagnosis is helpful for diagnosis and easy to perform. Other imagistic methods, such as CT and magnetic resonance imaging, provide detail of the surrounding structure and depth of the lesion but are too expensive to use in an otherwise simple diagnosis (6).

The treatment of choice and standard therapy for benign pilomatrixoma is complete surgical excision. If the overlying tegument is adherent to the tumor, it may also require excision (6). Morales and McGoey (41) advocated incision and curettement for cosmetic preservation in large tumors or for those in exposed areas and found no recurrence. Danielson-Cohen (6) noted 4% recurrence following complete surgical excision. However, in certain cases, such as the one presented, classical external incision of the skin, especially in the cheek, is not an option; the trans-oral approach is feasible but more laborious and prone to recurrence. This risk arises from poorer exposure of the incision, increased mobility and bleeding and therefore higher likelihood of remnant tissue. No recurrence was present in this case at 6 months and 1 year postoperatively which brings into consideration the assumption that both treatment and technique coordinate the doctor to a successful performance and that, occasionally, medical research can require a high degree of theorizing, abstraction and innovation (42).

Case 3. Inverted Schneiderian papillomas are benign tumors of the nasal and sinus area most commonly approached endoscopically (43,44) and have been discussed in literature for over a century (18). Due to their rarity and confusing nomenclature, they remain a topic of controversy (45). The reported incidence of papilloma is 1.7-7.0% (46). They are typically located unilaterally, whereas the present case involved bilateral localization. A similar bilateral case was reported by Neagos *et al* (47) in 2014. The recurrence rate is 0-27% and the endoscopic approach is recommended in cases with limited invasion of the nasal fossa and ethmoid cells (43,44). The decision to resort to open surgery depends on the size, localization and histopathological tumor type (48,49). Cortisone and antibiotic treatment are also paramount for postoperative care (48,49).

The inverted Schneiderian papilloma has a peak incident in individuals aged 50-69 years, as proven by numerous studies (2,12,15,18,47). The retrospective study by Bielamowicz *et al* (50) on 61 cases reported a mean age of 63 years and a male:female ratio of 2:1. There are, however, reports on papilloma in patients aged 5-25 years (51-54). Malignant transformation in recurrent cases, as well as

coexisting inverted papilloma and squamous cell carcinoma, have been documented (55,56).

The present hemangioma case involved a rare benign tumor which appeared to auto-resect due to loss of blood supply. It was hypothesized that the pedicle necrotized over time and cut off blood supply to the mass. This allowed the surgeon to remove it with ease and left no bleeding, but it is possible that the mass would have fallen out.

Pilomatrixoma, also termed calcifying epithelioma of Malherbe, is a relatively uncommon benign tumor of ectodermal origin derived from the hair matrix cells. The most common misdiagnosis is a dermoid cyst. Typical diagnostic methods include clinical examination, ultrasound and histopathology. Surgical removal is curative. The recurrence frequency reported is ~5%. The present patient refused an external approach due to esthetic reasons. Therefore, trans-oral resection was performed. This approach, although possible, is counterintuitive, more laborious and prone to recurrence due to incomplete resection of the pilomatrixoma. There was no recurrence in the present case at 6 months and 1 year post-operatively. Nevertheless, the classical external approach is recommended when possible.

Inverted papillomas have a high recurrence rate and propensity for malignant change. The present case was distinctive due to its rare bilateral nature. The endoscopic approach is the most successful in terms of functional, aesthetic results, short hospitalization period and improving quality of life. For large, invading tumors, open surgery is recommended to minimize recurrence and malignant development risk.

In conclusion, ENT tumors benefit from multidisciplinary approach to diagnosis and treatment and, in addition to ENT specialists, often require attention from general and vascular surgeons, pneumologists and rehabilitation specialists to provide a high quality of life following removal and complete healing.

Acknowledgements

Not applicable.

Funding

No funding was received.

Availability of data and materials

All data generated or analyzed during this study are included in this published article.

Authors' contributions

HM, PAP and AN conceived the study, performed patient selection, care and operations, collected data and edited the manuscript. PAP was responsible for designing, performing and supervising the pulmonary rehabilitation evaluation program. AIM, CM and IS made substantial contributions to acquisition, analysis and interpretation of data. AIM, CM and AN performed data analysis and prepared figures. HM and PAP confirm the authenticity of all the raw data. All authors have read and approved the final manuscript.

Ethics approval and consent to participate

The present study was approved by Research Ethics Committee of the Faculty of Medicine, Titu Maiorescu University (approval no. 6/21.09.2021; Bucharest, Romania). All patients provided informed written consent.

Patient consent for publication

All patients provided informed consent and approved the publication of their data.

Competing interests

The authors declare that they have no competing interests.

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