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Pleomorphic adenoma of the nasal cavity in a 21 – year – old patient – case report

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Abstract

Introduction and aim: Adenoma pleomorphum (multiform adenoma) is the most popular bening tumor, that originates from the salivary glands. Neoplasm develops mainly from large salivary glands and its occurrence in the nasal cavity is rare and falls mainly in the 3-6 decade of life. Only 1% of all pleomorphic adenomas develop in the nasal and sinus cavities. Despite being a benign tumor, malignant transformation can occur in 2-10% of cases.

Description of the case: We report a case of a 21-year-old female ethnically Polish patient with nasal multiform adenoma with initial diagnosis, which indicated a polyp. The final diagnosis was based on the histopathological examination of the resected lesion. The tumor was removed by an endoscopic method with no tissue margins, which may increase the risk of recurrence.

Conclusions: Our case demonstrates that despite pleomorphic adenomas typically occurs in the 3rd to 6th decade of life, clinical symptoms are nondistinctive with a polyp and nasal cavity is a very rare location there should be always be increased vigilance leading to choosing best diagnostic measures to form right diagnosis.

Key words: multiform adenoma, nasal cavity, polyp, young age, case report

Introduction:

A pleomorphic adenoma is a benign tumor, also known as a mixed tumor. These tumors grow slowly, typically encapsulated, with diameters up to 5 cm. However, if neglected, they can grow to much larger sizes. They have a round shape and a smooth surface. Composed of epithelial and myoepithelial components dispersed in a matrix, they can take the form of a mucinous, cartilage-like, or bony mixture. The tumor is well-differentiated, with epithelial and myoepithelial cells forming ducts, clusters, and irregular tubes [1,10].

Pleomorphic adenomas are among the most common tumors of the salivary glands, constituting over half of the tumors in the parotid gland. They occur relatively infrequently in the submandibular gland and even less so in the minor salivary glands [1]. Cases have also been reported in the mucous membrane of the oral cavity, throat, nose, larynx, trachea, sinuses, and lacrimal gland. Instances of pleomorphic adenomas occurring outside the salivary glands are considered atypical [2,3,8]. In the upper respiratory tract, the nasal cavity is the most privileged site for their occurrence [8]. They typically appear in the mucous membrane of the nasal septum, even though mucous glands are mainly located in the lateral wall of the nose [4-7]. Histologically, pleomorphic adenomas in the nasal cavity differ from those in large salivary glands, characterized by higher cellularity and a smaller amount of matrix [4]. The tumor is non-encapsulated, and there are no visible signs of necrosis. Microscopically, three main elements are observed: epithelial, myoepithelial, and mesenchymal, characteristic of a mixed tumor. The epithelial component consists of columnar and cuboidal cells, which, along with epithelial-muscular cells, form tubular, tubular-like, and solid field structures..

A mixed tumor located in the nasal cavity is initially asymptomatic, but it can later lead to progressive nasal obstruction, bleeding, pain, the presence of mucous or mucopurulent discharge, and as the tumor grows, deformities in the external nose and disturbances in smell [2-4].

The diagnosis requires performing computer tomography, magnetic resonance imaging, and histopathological examination of the tumor. Confirming its presence indicates the need for surgical removal [14]. Pleomorphic adenomas may recur, especially if they are excised without a margin of healthy tissue, but the recurrence rate is often low. A mixed tumor may

undergo malignant transformation, and the risk increases over time. Cancer arising from a pleomorphic adenoma is referred to as carcinoma ex pleomorphic adenoma or malignant mixed tumor [1,9]. A pleomorphic adenoma located in the nasal septum has a higher risk of malignant transformation compared to tumors in other nasal cavity locations. Carcinoma ex pleomorphic adenoma has a greater propensity for invasion and most commonly metastasizes to the bones [11,12,13].

A 21-year-old female student, of Polish origin, was admitted to the Otolaryngology Clinic based on a scheduled referral, with a diagnosis of a polyp in the left nasal cavity. ICD-10 code: J33.0. In the interview conducted by the attending physician, the patient complains of nasal obstruction for approximately 6 months. Previously, she experienced nosebleeds. The patient did not report any pain or disturbances in her sense of smell. According to the interview, the patient is not allergic to any medications and does not have any chronic illnesses. Anterior rhinoscopy revealed that the internal nasal passage is filled with a mass consistent with a polyp, while the ear canal and throat showed no abnormalities. No signs of meningeal symptoms or visual disturbances were observed.

Palpation did not reveal enlargement of the cervical lymph nodes. Apart from the deviations mentioned in the otolaryngological examination, no other abnormalities from the norm were found. Blood laboratory test results indicate microcytic anemia: hemoglobin 10.7 g/dl (12-16), hematocrit 34.2% (37-47%), MCV 77 fl (80-94 fl), MCH 24.1 pg (27-31 pg). Results of other laboratory tests were within normal limits. No anomalies were detected in the chest X-ray. The procedure was performed using an endoscopic method under general intratracheal anesthesia, employing the following medications: fentanyl, propofol, rocuronium, and bridion, as well as a gas mixture consisting of oxygen, nitrous oxide, and sevoflurane. The patient was intubated with a 6.5 mm tube through the mouth. The anesthesia duration was 15 minutes, and the surgery lasted 5 minutes. Both the anesthesia and the surgical procedure proceeded without complications. During the surgery, a tumor measuring 3x2.5x1.5 cm was completely removed according to macroscopic assessment. The material was labeled and sent to the pathology laboratory. The tumor obstructing the entire left nasal cavity was removed, attached in the posterior-inferior part of the nasal septum.An anterior left-sided nasal packing was applied. After the procedure, the patient received pain relievers: paracetamol, pyralgin,

tramadol, and locally sulfarinol and rinopantein. The hospitalization, operative procedures, and intraoperative course proceeded without complications. The patient was discharged the day after the surgery in good general condition, with a referral for follow-up at the Otolaryngology Clinic. There were no complaints, and her overall condition was good. Further observation did not reveal any local recurrence. Based on the histological examination of the sent material, it was demonstrated that the tumor was removed without margins of the surrounding tissue, and a diagnosis of pleomorphic adenoma was established, originating from the minor glands of the nasal mucosa.

The cells are well-differentiated, and no atypia, nuclear polymorphism, or increased mitotic activity was observed, suggesting a benign nature of the lesion. The structures are dispersed in a matrix, which is a product of myoepithelial cells. The stroma in the histological image predominates over the epithelial component and consists of a heterogeneous mixture of connective tissue with collagen, mucoid tissue, and mucoid-cartilaginous tissue. Cartilaginous islands with scattered epithelial and myoepithelial cells in the matrix are also visible (Figure 1,2,3).

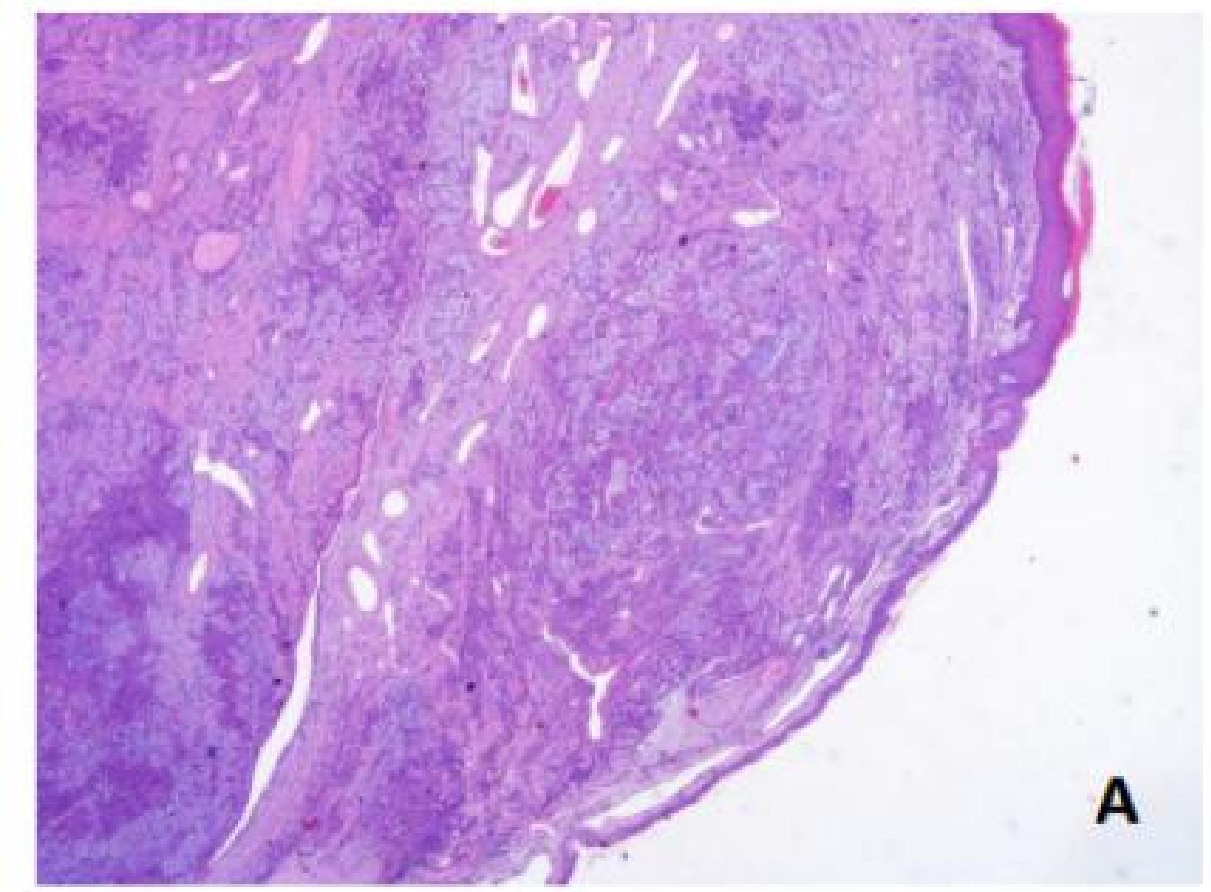

Figure 1

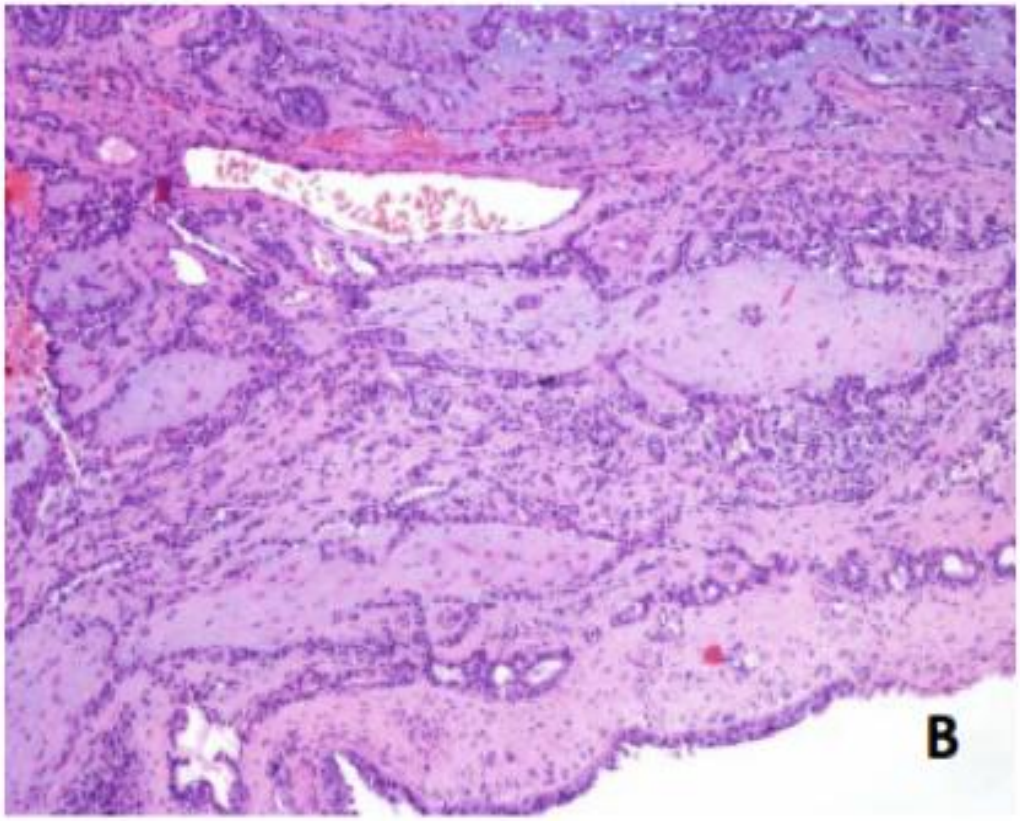

Figure 2

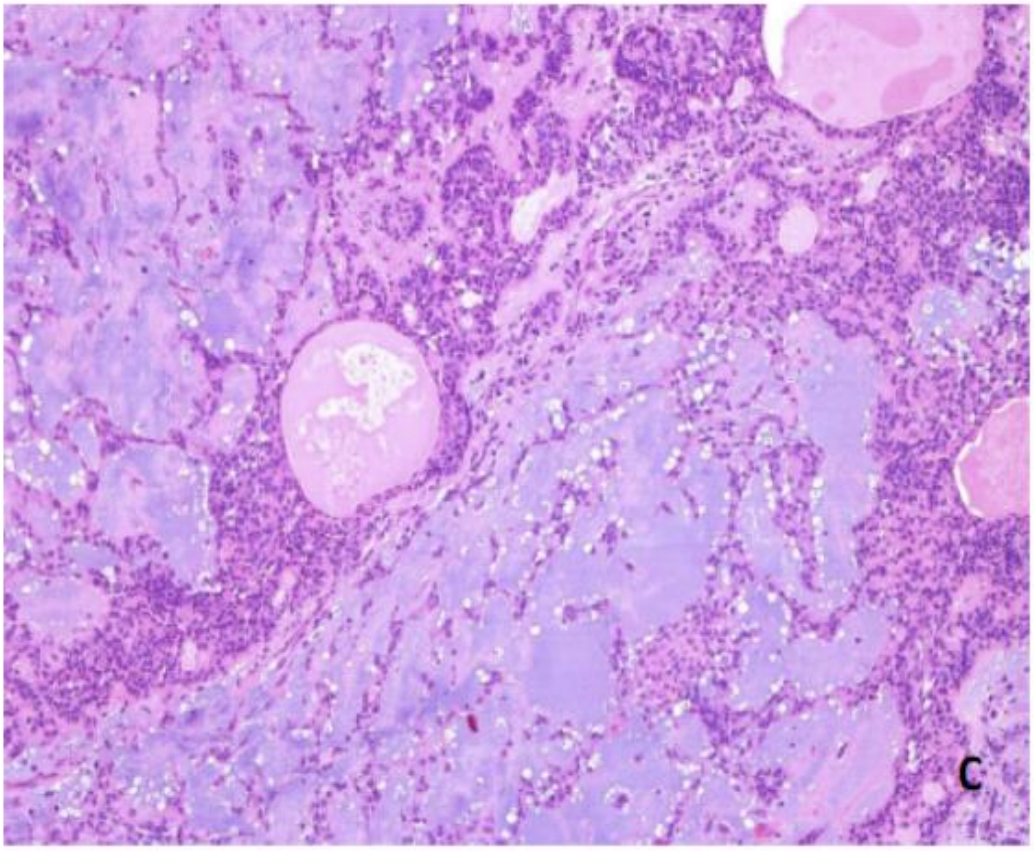

Figure 3

Fig. 1, 2,3 Microscopic view of pleomorphic adenoma originating from minor glands of nasal mucous membrane: A-HE, 20x; B - HE, 100x; C - HE, 100x. Microscopic slide faciliated by Department of Pathomorphology, Medical College of The University of Rzeszów

Discussion:

Pleomorphic adenoma (adenoma pleomorphum) is the most common benign tumor of the salivary glands [23]. It consists of epithelial and mesenchymal tissue [20]. It is predominantly found in major salivary glands, with over 80% of cases developing in the parotid glands, less frequently in the submandibular and sublingual glands [23]. In small salivary glands, pleomorphic adenoma occurs rarely [20]. The most common location is the nasal cavity, followed by the sinuses and nasopharynx, constituting about 1% of all pleomorphic adenomas [17, 21].

Literature also describes the occurrence of the tumor arising from small salivary glands in the oral cavity, hard palate, larynx, trachea, and lacrimal glands [23]. They most commonly develop in the 3rd to 6th decades of life with a predisposition in women [17, 21]. In the literature, the first documented case of pleomorphic adenoma in the nasal cavity occurred in 1929 [17]. Despite the majority of minor mucous and serous glands being located in the lateral wall of the nasal cavity, 80% of pleomorphic adenomas develop from the mucosa of the nasal septum [17, 21]. Several theories have been proposed regarding the origin of the tumor in the nasal cavity. One suggests that the adenoma may arise from the accessory organ. Another theory proposes that it may result from the migration of embryonic epithelial cells of ectodermal origin into the nasal septum. Meanwhile, Evans and colleagues suggested the origin of pleomorphic adenoma from major salivary glands [18].

Clinical symptoms of pleomorphic adenoma of minor salivary glands are often nonspecific and depend on the tumor's location [18, 19]. Typically, the tumor presents as a slow-growing, painless mass that may persist for many years without causing any symptoms [19, 22]. In the case of pleomorphic adenoma in the nasal cavity, the main symptoms include progressive nasal obstruction and intermittent nosebleeds [22]. Other symptoms may include olfactory disturbances, voice changes, sleep apnea, and mucopurulent rhinorrhea [17, 21, 23]. When the tumor reaches significant sizes, it can lead to external nasal deformation, swelling, and pain [22]. In our patient, we observed nasal obstruction and bleeding. Rhinoscopic examination revealed a polypoid, pink-gray mass with a soft consistency and smooth surface, typically not bleeding profusely during removal [22, 23].

A tumor filling the nasal cavity needs to be differentiated from other diseases of the nose and paranasal sinuses such as nasal polyps, angiofibroma, inverted papilloma, squamous cell carcinoma, adenocarcinoma, mucoepidermoid carcinoma, adenocystic carcinoma, and melanoma [22]. Due to the unusual site of tumor development and nonspecific symptoms, the diagnosis of pleomorphic adenoma in the nasal cavity is often challenging in many cases [18, 23]. In addition to a thorough otolaryngological examination, diagnostic procedures for pleomorphic adenoma in the nasal cavity include nasal endoscopy, computed tomography (CT), magnetic resonance imaging (MRI), cytology, and histopathology [19, 23]. Imaging studies are valuable in determining the precise location and size of the tumor [19]. CT is essential before surgical procedures to identify the tumor's origin and plan the surgical intervention. When the CT image is ambiguous or suggests a malignant change, an MRI

should be performed. MRI helps rule out involvement of surrounding structures and facilitates differentiation from other conditions that may occur within the nasal cavity and paranasal sinuses [22]. Fine-needle aspiration biopsy can be helpful in reaching a definitive diagnosis. Unfortunately, the histological diversity of pleomorphic adenoma can lead to misdiagnosis and ineffective treatment.

	Gruczolak wielopostaciowy dużych gruczołów ślinowych	Gruczolak wielopostaciowy małych gruczołów ślinowych
Komórki nabłonkowe	Śladowe ilości	Liczne
Komórki mioepitelialne	Liczne	Śladowe ilości
Pseudokapsułki	Obecne	Brak

Only complete tumor resection and histopathological examination of the entire specimen provide a conclusive diagnosis [23]. In the histopathological image, pleomorphic adenomas of minor salivary glands stand out for having a more cellular component than stromal compared to pleomorphic adenomas of major salivary glands [21].

Tab. 1 Comparison of histopathological features of pleomorphic adenomas of minor and major salivary glands [20].

Pleomorphic adenoma of the nasal mucosa has low radiation sensitivity and a weak response to chemotherapy. Therefore, the treatment of choice is surgical resection with clear margins [17, 20]. The surgical procedure may involve endoscopic surgery, lateral rhinotomy, face-lifting, or a combination of open and endoscopic techniques, depending on the size and location of the tumor, as well as the experience of the treating center [21].

Although pleomorphic adenoma of the nasal cavity is a benign tumor, it undergoes malignant transformation in 2.4% - 6% of cases, with a predisposition in women [20, 22]. Local recurrence of the disease has been noted in 7.5%–8.8% of cases [20]. Factors that increase the risk of recurrence include the absence of surgical resection margins, the presence of pseudopods, capsule penetration, and tumor rupture [19, 20]. Therefore, it is important to use the appropriate surgical technique with maintaining an adequate margin without violating the continuity of the outer capsule [19, 20, 21]. Continuous patient observation is

recommended since the majority of recurrences occur within the first 18 months after surgical treatment [21, 22].

In the case we have described, the patient's young age posed a significant challenge in reaching a correct diagnosis. Polyps are the most common nodular changes in the nasal cavity in young adults and are typically diagnosed based on laryngological examination. Due to the similarity in rhinoscopic examination to pleomorphic adenoma, there is a risk of misdiagnosis, leading to the choosing of a different, less effective treatment method [15, 16]. Therefore, precise differentiation between polyps and other nodular changes in the nasal cavity becomes particularly important to ensure appropriate treatment and optimal therapeutic outcomes for the patient.

Summary

Multiple papilloma of the nose, though rare, poses challenges for both doctors and patients alike. Its atypical occurrence at a young age, especially before the third decade of life, raises questions regarding risk factors and pathogenesis. Also concerning is the lack of sufficient scientific data regarding this condition in young individuals. With technological advancements, the diagnosis of such tumors becomes increasingly precise, but still requires a diversified approach.

In the case of our patient, clinical symptoms could suggest both nasal polyps and multiple papilloma. This highlights the importance of accurate diagnosis, particularly in the context of potential recurrences and surgical treatment. It is worth emphasizing that differentiation between these two conditions can be difficult, and misdiagnoses may lead to inappropriate treatment.

Modern diagnostic methods such as computed tomography, magnetic resonance imaging, and nasal endoscopy are extremely helpful but also costly and time-consuming. Therefore, continuous improvement of clinical skills among physicians and access to modern medical technologies are crucial aspects.

After establishing the final diagnosis of multiple papilloma, another challenge arose: surgical treatment. Although tumor resection without a margin of healthy tissue is a standard

procedure, there is still a risk of recurrence. Therefore, the patient should be under constant observation, requiring collaboration among multiple specialists, including otolaryngologists, pathologists, and radiologists.

In conclusion, cases of multiple papilloma of the nose in young individuals are rare and present challenges for physicians. They require a complex diagnostic and treatment process, as well as ongoing patient care. Further scientific research and clinical experience are necessary to better understand this rare disease entity and improve treatment effectiveness.

Disclosures

Author's contribution

Conceptualization: M.P. Methodology: S.Z. Software: S.Z. Validation: T.B. Formal analysis: Katarzyna M. Investigation: T.B. Resources: A.B. Data curation: A.B. Writing original draft preparation: M.P. S.Z. T.B. A.B. Katarzyna M. V.C. Karolina M. Writing review and editing: M.P. Katarzyna M. V.C. Karolina M. Visualization: V.C. Supervision: Katarzyna M. M.P. Project administration: M.P.

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