

Giant pleomorphic adenoma of the nasal cavities and paranasal sinuses

Olbrzymi gruczolak wielopostaciowy nosa i zatok przynosowych

Paweł Sroka, Sławomir Okła

Clinic of Otolaryngology, Head and Neck Surgery, Świętokrzyskie Center of Oncology

Article history: Received: 11.10.2017 Accepted: 27.06.2018 Published: 30.09.2018

ABSTRACT:

Pleomorphic adenoma is the most common benign tumor of the salivary glands. Pleomorphic adenoma occurs in the sinonasal cavities only in 1% of cases, which is an expression of this tissue's ectopic aspect. The histopathological image of ectopic pleomorphic adenoma resembles those that originate from the minor salivary glands. They have increased cellular and reduced stromal component in relation to adenomas of the major salivary glands. Malignant transformation of pleomorphic adenoma can occur in 2-10% of cases. This work presents a case of giant pleomorphic adenoma of the nasal cavities and paranasal sinuses with extensive destruction of the nose, paranasal sinuses and eye sockets in a 37-year-old patient, treated by endoscopic surgical resection.

KEYWORDS:

nasal and paranasal sinus cancer, ectopic pleomorphic adenoma, endoscopic surgery of the nose and paranasal sinus

STRESZCZENIE:

Gruczolak wielopostaciowy jest najczęstszym nowotworem niezłośliwym, wywodzącym się z gruczołów ślinowych. Około 1% gruczolaków wielopostaciowych rozwija się w jamach nosa i zatok, co jest wyrazem ektopii tej tkanki. Obraz histopatologiczny ektopicznych gruczolaków wielopostaciowych przypomina te, które wywodzą się z małych gruczołów ślinowych. Charakteryzują się one wzmożoną komórkową i zmniejszoną stromalną w stosunku do gruczolaków dużych gruczołów ślinowych. Transformacja złośliwa ektopicznych gruczolaków wielopostaciowych występuje w 2–10% przypadków. Przedstawiono przypadek olbrzymiego gruczolaka wielopostaciowego nosa i zatok przynosowych z rozległą destrukcją nosa, ścian zatok przynosowych i oczodołów u 37-letniego pacjenta, leczony drogą endoskopowej resekcji chirurgicznej.

SŁOWA KLUCZOWE:

guzy nosa i zatok przynosowych, ektopiczny gruczolak wielopostaciowy, chirurgia endoskopowa nosa i zatok przynosowych

INTRODUCTION:

Pathological masses in the nasal cavities and paranasal sinuses are found in 1-4% of the population [13]. They constitute a very diverse group of diseases with a possible both inflammatory and cancerous etiology; it should be remembered that they may co-exist. They may go unnoticed for months and years due to symptoms similar to those of chronic infection. Most of them are non-cancerous lesions whose share is most often between 68% and 80%, although in some reports it is estimated at about 60% and even below 60% [8,13]. Data regarding benign tumors

indicate that they account for 13-23% of pathological masses within the nasal cavities and paranasal sinuses, however the range found in the literature is wide: 5 to 31%, while the least numerous group is malignant tumors 8-17%. [8,13]. Occurrence of salivary gland tumors within the nasal cavities and the sinuses constitutes this tissue's ectopic aspect.

Salivary gland tumors constitute less than 1% of all tumors and comprise about 3-6% of head and neck cancers. [10,18,20]. These tumors develop in large (parotid, submandibular and sublingual salivary glands) as well as in the minor salivary glands, which

include small clusters of glandular tissue in the upper gastrointestinal tract. Tumors of the minor salivary glands constitute 10-15% of all tumors of the salivary glands [18]. Pleomorphic adenoma is the most common tumor of both the major and minor salivary glands, constituting about 65% of all salivary gland tumors [2,19], and about 40% of tumors of the minor salivary glands [19]. It develops most often in the 3rd-6th decade of life, more often in women, but it can occur at any age [1].

Approximately 1% of all pleomorphic adenomas are those that develop within the nasal cavities, sinuses and nasopharynx [16]. Pleomorphic adenoma of the minor salivary glands in the histopathological image stand out with a much larger cellular than stromal component compared to pleomorphic adenoma of the major salivary glands. [6,11]

There are between 450 and 750 small salivary glands [9] within the head and neck, most numerous accumulated in the oral and pharyngeal mucosa, however, they are also found in the ectopic area in the larynx, nasal cavities, and in the paranasal sinuses. It is most commonly reported that tumors of the minor salivary glands constitute 10-15% of all salivary gland tumors [18], however, the spread of literature data reaches 9-25% [14,17]. They are most commonly located on the palate (50%), in particular on the borderline between the soft and hard palate [17], lips (15%), buccal mucosa (12%), tongue and floor of the mouth (5% each) [18].

There is general agreement that the smaller the salivary gland, the more likely it is that the tumor developing therein will be malignant [20]. Benign tumors dominate among tumors of the major salivary glands; malignant tumors constitute 15-32% of parotid gland tumors and 41-45% of submandibular gland tumors. The National Cancer Registry as well as most studies do not distinguish between malignant tumors of sublingual salivary glands, they are usually classified together with tumors of the minor salivary glands of this location [9,20]. The percentage of malignant tumors among all tumors of the minor salivary glands is relatively the largest and reaches 50% [9,14], with quite a large variation depending on the location; about 90% in the floor of the mouth, tongue and the retromolar region [14,18], 40-60% within the palate and lower lip [9,18], least frequently in the upper lip area (approximately 25%) [18].

Pleomorphic adenoma is the most common tumor of both the major and minor salivary glands, constituting about 65% of all tumors of the salivary glands, and about 40% of tumors of the minor salivary glands [19]. It was found that only about 1% of pleomorphic adenomas are those that form in the nasal cavities and nasopharynx. [16]. Despite the fact that the majority of ectopic minor salivary glands in the nasal cavities

are located on the lateral wall of the nasal cavity and turbinate [1], adenomas of the vast majority of nasal cavities (80%) derive from the nasal septum mucosa, below 20% from the lateral side of the nasal cavity [7]; in the case of some large tumors, the original location could not be unambiguously determined [6]. Tumors of the minor salivary glands constitute 4-8% of all cancers of nasal cavities and paranasal sinuses [12]. Pleomorphic adenomas of the minor salivary glands may occur at any age; however, they most often develop in the 3rd-6th decade of life [1]. While it is recognized that adenomas are more common in women [1], in the case of pleomorphic adenomas located in the nasal and sinus cavities, gender predomination was not found [3,6]

Clinical symptoms most frequently reported by patients are unilateral nasal obstruction 71%, nosebleeds 56% [1], the remaining symptoms are impaired sense of smell, rhinorrhea, voice change secondary to nasal obstruction.

CASE REPORT:

A 37-year-old patient reported chronic nasal congestion, anosmia, altered speech (closed nasality) and chronic rhinorrhea. Based on the interview, it was established that the patient had an x-ray of the paranasal sinuses about 2 months earlier, but its technical quality was poor, which probably led to its faulty interpretation; "partial occlusion of the nasal cavity" was found based on the x-ray. There are small wall protrusions of the mucous membrane visible in the medial

regions of the maxillary sinuses. The remaining paranasal sinuses in the picture are properly aerated." Moreover, it was also found that he suffers from psoriasis, gout and moderate mental retardation, and therefore the time of onset of symptoms has not been clearly established. The study found tumor mass, without signs of superficial ulcer filling the left nasal cavity and moving the septum to the right, which resulted in practically complete nasal obstruction. The remaining elements of the ORL test as well as physical examination did not bring any significant elements for further treatment. CT examination was performed first as part of imaging diagnostics, supplemented by MR examination, before the final decision regarding treatment. He was qualified for biopsy under local anesthesia. The following results were obtained: "Tumor fragment with (...) pleomorphic adenoma. Necessary excision of tumor in its entirety with subsequent histological examination." (Department of Pathology of Neoplasia of the Świętokrzyskie Center of Oncology in Kielce) and in CT: "A visible change of TU visible in the nasal cavity, dimensions 68x57mm, with calcifications, filling the entire nasal cavity, front rush (czy to jest ok), both



Fig. 1. Visible tumor encapsulated in greater part of the circumference, liquid-solid with dimensions of approx. 60x73x60mm, filling the entire nasal cavity, frontal sinus, with destruction of bony and cartilage nasal septum structures. Tumor destroys the medial walls and protrudes and models structures of the medial-bottom eye sockets conically to the outside. Congestive-fluid lesions in the scope of frontal, sphenoid and left maxillary sinuses.

maxillary sinuses, with destruction of the medial walls of both eye sockets and infiltration of the anterior-lateral walls of both maxillary sinuses, modeling of medial walls of both eye sockets (without signs of infiltration in the eyeball), destruction of sphenoid sinus walls, filling its entire lumen, and with destruction of the bony and cartilaginous nasal septum." Image diagnostics were supplemented with MR study: "W rzucie lokalizacji wymienionych w bad TK owalny, encapsulated in the greater part of the circumference, a liquid-solid tumor of about 60x73x60mm, causing destruction of bony structures (as in CT study). The tumor shows signs of diffusion limitation in the area of solid zones, the largest on average up to approx. 20mm ... Upwards, the tumor destroys the medial walls and protrudes and models structures of the medial-bottom eye sockets conically to the outside. congestive-fluid lesions in the scope of paranasal sinuses." (Department of Pathology of Neoplasia of the Świętokrzyskie Center of Oncology in Kielce) (Fig. 1).

Patient qualified for surgical treatment - endoscopic tumor resection. dislocation of the nasal septum to the right with its intussusception to the maxillary sinus and rush, lateral dislocation of the medial wall of the left maxillary sinus and left rush with intussusception into the eye socket were found intraoperatively, whereas compressive growth only in posterior part of septum - suspicion of tumor stem (czy to ok) or infiltration, middle nasal concha and ethmoidal bulla can on the left side also suspected of infiltration. Removed in stages, in fragments of tumor including, left anterior ethmoidal cells and the left medial wall of the maxillary sinus together with the inferior and middle nasal concha, the whole nasal septum was removed and frontal walls of the sphenoidal sinuses were removed. Tumor on the periphery with thin but clear capsule. No characteristics suspected of infiltration from ceiling and frontal recess and in rush on the right side. Extensive post-surgical cavity required use of 8 Meroceel dressings and 2 sets of setons made of gauze; posterior tamponade was also inserted using

a Folley catheter. Tamponade was gradually removed in day 3 and 4. After cleaning of the nasal cavity several times (czy to jest ok), the patient was discharged home on the 6th day.

Histopathology showed signs of adenoma pleomorphum, mixed type, in all examined tumor fragments (Department of Pathology of Neoplasia of the Świętokrzyskie Center of Oncology in Kielce, Dr J. Kopczyński). During periodic post-surgical follow-up, no recurrence symptoms were found within 12 months. This is confirmed both by CT examination and endoscopic image: Fig. 2, Fig. 3.

DISCUSSION

The histopathological image of pleomorphic adenoma consists of epithelial and stromal components. There are three main histological subtypes - myxoid (80% stroma), cellular (predominantly myoepithelial cells) and mixed (classical) [19]. Adenomas found in the nasal cavities had a more numerous epithelial/cellular component and less numerous stromal (sometimes very stingy) compared to adenomas of the major salivary glands [6]. Mixed and cell types were found among pleomorphic adenomas of the nasal cavities and sinuses; no stromal type was found [19]. It is a factor considered as the reason for a lower frequency of recurrences around 2.4-10% compared to adenomas of the major salivary glands [3,4,6,7,15,16,19]. The majority of confirmed local recurrences were observed within the first 18 months after surgical treatment [3].

A mixed type of pleomorphic adenoma was found in the obtained postoperative HP study in the presented case.

The frequency of transformation of malignant pleomorphic adenoma ranged from 2% [15] 6% [3] to 10% [4,6,10] and it is believed that it increases with the duration of disease [3]. It

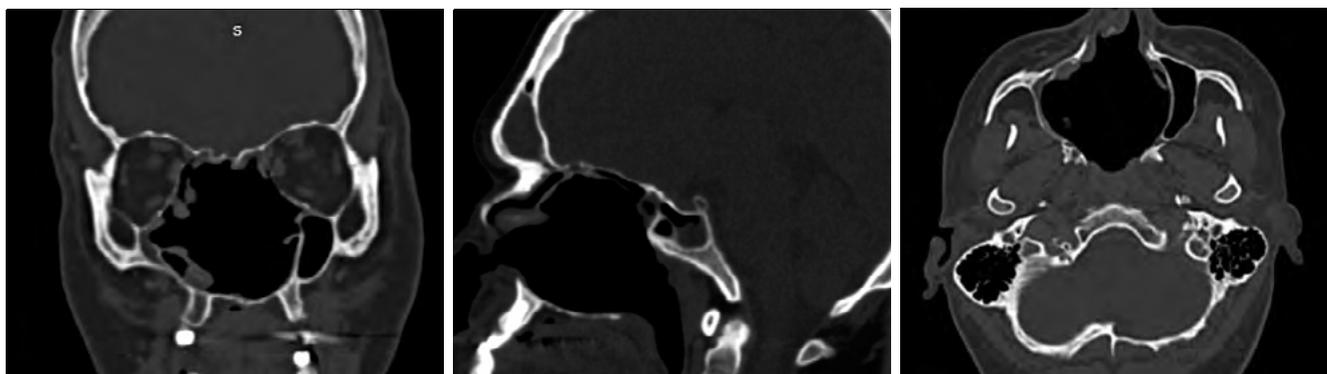


Fig. 2. CT of paranasal sinuses after surgery. Visible extensive tumor loge and signs of modeling of nasal cavity walls, paranasal sinuses, orbits, nasal concha through growth of expansive form of benign tumor.

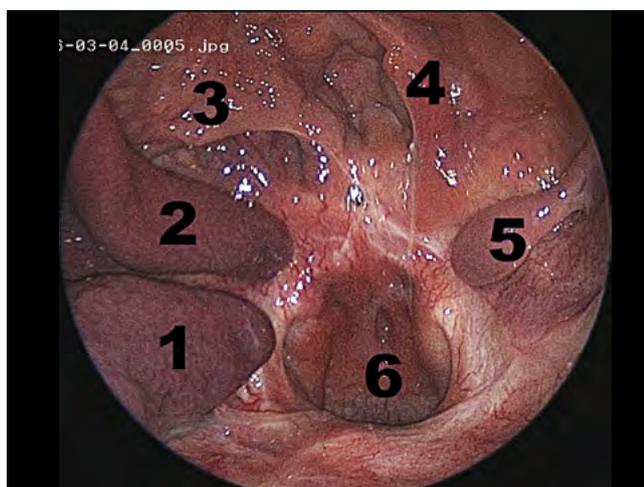


Fig. 3. 1 Right inferior nasal concha; 2 Right middle nasal concha; 3 Right superior nasal concha; 4 Left superior nasal concha; 5 Left middle nasal concha; 6 Nasopharynx

was observed that the mean age of a patient with malignant salivary gland tumor is about 10 years greater than of a patient with benign tumor [5, 10].

Imaging examinations are necessary before operative treatment and prove useful during diagnostics. Computed tomography shows tumor size and location, modeling features or infiltration and destruction of neighboring structures, especially bony structures. Lesions of calcifications within the tumor may be visible. Magnetic resonance imaging of small/mild pleomorphic adenomas shows their good limitation and spherical shape as well as fairly homogeneous signal of low intensity in T1 and high T2 sequences [11]. In the case of large adenomas shown in magnetic resonance imaging, their heterogeneous consistency and polycyclic shape are visible; cystic, fluid or stromal tumor components as well as tumor pouch can be seen as well (low signal intensity in T2 and contrast enhancement T1). [11.16]. Reduction of diffusion (DWI) within solid tumor zones, which is

rather characteristic of malignant tumors, can be visible. This is due to reduction of diffusion of water molecules in extravascular space (mainly extracellular) in relation to increased number of cells, their edema, neo-angiogenesis, necrosis or increase of connective tissue, typical for extensive cell tumors [5]. In the case of tumor localized in the nasal cavities and paranasal sinuses, magnetic resonance imaging allows to assess the existence or absence of infiltration into soft tissue structures (e.g., orbital contents, meninges) which is in contact with the tumor after destruction of the bony wall of the nasal cavity and sinuses. It also allows to distinguish congestive lesions in sinuses obstructed by the tumor due to indenting tumor masses.

Treatment consists in surgical resection while preserving tumor-free margins. Surgical access may include independent endoscopic surgery, lateral rhinotomy, midfacial degloving or a combination of open techniques and endoscopic technique, and should be selected depending on tumor size and location and experience of the treatment center. Surgical treatment of pleomorphic adenomas in their typical location (major salivary glands) consists in resection within normal tissue and technique aimed at removing the tumor without disturbing external continuity of the capsule. This was not possible in the described case; tumor was resected via endoscopic technique in fragments from the center to the perimeter, causing reduction of its mass, and the final stage consisted in removing the capsule, checking for any infiltration into adjacent tissues. The chosen surgical technique is burdened with an increased recurrence risk resulting from tumor fragmentation and the possibility of „implanting” tumor tissue, however, the alternative could only be extensive and maxillo-facial resection which also, due to tumor size and location, did not guarantee normal tissues resection and lack of tumor fragmentation. In addition, risk of recurrence of local pleomorphic adenoma originating from the minor mucous glands is lower as a result of different histological structure - superiority of cellular component over stromal [3,4,6,7,15,16,19].

CONCLUSION

Pleomorphic adenoma is a rare paranasal sinus and nasal cavity cancer, most often derived from the nasal septum. It differs in increased cellular component in the histopathological image in

relation to pleomorphic adenomas of the major salivary glands. Even with large tumors, endoscopic surgical treatment is effective and leaves a good functional and aesthetic effect. Despite the low recurrence rate reported, post-treatment follow up should be continued due to potential risk of local recurrence.

References

- Arora A., Sharma K.: Pleomorphic adenoma of the nasal cavity-an unusual presentation. *Int J Otorhinolaryngol Head Neck Surg.* 2017 Jan; 3 (1) 162-265.
- Al Momen A., Al Khatib A.: Pleomorphic adenoma of the nasal septum: a case report and review of the literature. *Pan Arab J Rhinol* 2016, 06: 36-38.
- Aydm S., Celebi O., Sanh A., Hardal U.: A case of pleomorphic adenoma of the inferior turbinate. *Turk Otolareng Arsivi*, 2010; 48 (3): 120-125.
- Baglam T., Durucu C., Cevik C., Bakir K., Oz A., Kanlikama M.: Giant pleomorphic adenoma of the nasal septum. *Indian J Otolaryngol Head Neck Surg* (October-Dezember 2011) 63 (4): 393-395.
- Cieszanowski A.: Zastosowanie badania rezonansu magnetycznego w onkologii. *Onkol. Prak. Klin.* 2013; 9; 2: 60-69.
- Compagno J., Wong R.: Intranasal mixed tumors (Pleomorphic Adenomas). *A.J.C.P.* August 1977 Vol. 68. No. 2: 213-218.
- Elwany S., Mandour Z., Talaat I.: Pleomorphic adenoma of the nasal septum. *Journal of Otoalryngology Advances.* Vol. 1. Issue 1: 22-26 (www.openaccesspub.org).
- Gupta R., Moupachi S.S., Poorey V.K.: Sinonasal Masses: A Retrospective Analysis. *Indian J Otolaryngol Head Neck Surg* (2013) 65 (1): 52-56.
- Guzzo M., Locati L., Prott F., Gatta G., McGurk M., Licitra L.: Major and minor gland tumors. *Critical Reviews in Oncology/Hematology* 74 (2010) 134-148.
- McKenna R.: Tumors of the Major and Minor Salivary Glands. *CA-A Cancer Journal for Clinicians*, Vol. 34. No. 1. (Jan/Feb 1984): 24-39.
- Motoori K., Takano H., Nakano K., Yamamoto S., Ueda T., Ikeda M.: Pleomorphic Adenoma of the Nasal Septum: MR Features. *AJNR Am J Neuroradiol* 21: 1948-1950 (Nov/Dec 2000).
- Pantavaidya G., Vaidya A., Metgudmath R., Kane S., D'Cruz A.: Minor salivary gland tumors of the sinonasal region: Results of a retrospective analysis with review of literature. *Wiley Online Library* 31 January 2012 (www.wileyonlinelibrary.com) *Head&Neck* 1704-1710.
- Rawat D.S., Chadha V., Grover M.: Clinico-patological Profile and Management of Sino-nasal Masses: A prospective Study. *Indian J Otolaryngol Head Neck Surg* (2013) 65 (Suppl 2): S388-S393.
- Shivakumar T., Sudhir N., Poonam J., Pankaj Ch.: A review of Salivary gland Neoplasms and its management. *Otolaryngology online journal*, Volume 4 Issue 3 2014.
- Składzień J., Stręk P., Oleś K., Wiatr M., Glowacki R., Tomik J.: Pleomorphic adenoma of the nasal cavity. *The American Journal of Case Reports*, 2009; 10: 161-165.
- Wierzchowska M., Bodnar M., Burduk P., Kaźmierczak W., Marszałek A.: Rare benign pleomorphic adenoma of the nose: short study and literature review. *Videosyrgeria Miniinv* 2015; 10 (2): 332-336.
- Wyszyńska-Pawelec G., Gontarz M., Zapala J., Szuta M.: Minor Salivary Gland Tumours of Upper Aerodigestive Tract: A Clinicopathological Study. *Hindawi Publishing Corporation Gastroenterology Research and Practice* Volume 2012, Article ID 780453.
- Vicente O.P., Marques N.A., Aytes L.B., Escoda C.G.: Minor salivary gland tumors: A clinicopathological study of 18 cases. *Med Oral Patol Cir Bucal*. 2008 Sep; 13 (9): E: 582-8.
- Yen-Ling Kuo, Tzong-Yang Tu, Chia-Fan Chang, Wing-Yin Li, Shyue-Yih Chang, An-Suey Shiao.: Extra-major salivary gland pleomorphic adenoma of the head and neck: a 10-year experience and review of the literature. *Eur Arch Otorhinolaryngol* (2011) 268: 1035-1040.
- Ziółkowska M., Bień S., Okła S., Żyłka S.: Charakterystyka epidemiologiczno-kliniczna 705 nowotworów gruczołów ślinowych. *Otolaryngologia Polska* 2013 (67): 154-163.

Word count: 2720 Tables: – Figures: 7 References: 20

Access the article online: DOI: 10.5604/01.3001.0012.1467 Table of content: <https://otorhinolaryngologypl.com/issue/11380>

Corresponding author: dr Paweł Sroka, Clinic of Otolaryngology, Head and Neck Surgery, Świętokrzyskie Center of Oncology, ul. S. Artwińskiego 3, 25-734 Kielce, phone no.: +48 41 36 74 336; e-mail: p.sroka@prokonto.pl

Copyright © 2018 Polish Society of Otorhinolaryngologists Head and Neck Surgeons. Published by Index Copernicus Sp. z o.o. All rights reserved

Competing interests: The authors declare that they have no competing interests.

Cite this article as: Sroka P., Okła S.: Giant pleomorphic adenoma of nasal cavities and paranasal sinuses; *Pol Otorhino Rev* 2018; 7(3): 26-31

