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CASE REPORT

VASCULAR ANOMALY OF THE PAROTID GLAND: CASE REPORT

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Abstract

Introduction. Vascular anomalies of the parotid gland are rare congenital defects and encompass variations in vascular structure and arterial blood supply. Due to the rarity of these defects, the diagnosis and treatment of vascular anomalies of the parotid gland can be complex and require an individualized approach. Treatment of these anomalies may include conservative methods or surgical interventions, depending on the nature of the anomaly and the severity.
Case presentation. A 50-year-old woman with a right parotid gland neoplasm was examined and a surgical intervention was performed. During removal of the mass, no large feeding vessels could be found. The histopathological examination showed the presence of venous vessels, arterioles, dilated ectatic venules lined with flat endothelial lining, lymphatic vessels, and smooth muscle. The immunohistochemical study showed positivity for vascular markers. Based on laboratory studies, a diagnosis of vascular malformation of the parotid salivary gland was established.
Conclusions. Vascular anomalies of the parotid glands in adults should be considered hyperplasias, which can be easily removed without damaging the gland's structure, as they are more commonly located at its periphery.

Résumé

Anomalie vasculaire de la glande parotide: rapport de cas

Introduction. Les anomalies vasculaires de la parotide sont des malformations congénitales rares qui englobent des variations de la structure vasculaire et de la vascularisation artérielle. En raison de leur rareté, le diagnostic et le traitement des anomalies vasculaires de la parotide peuvent être complexes et nécessiter une approche individualisée. Le traitement de ces anomalies peut inclure des méthodes conservatrices ou des interventions chirurgicales, selon la nature et la gravité de l'anomalie.
Présentation du cas. Une femme de 50 ans présentant une tumeur de la parotide droite a été examinée et une intervention chirurgicale a été réalisée. Lors de l'ablation de la masse, aucun gros vaisseau nourricier n'a été détecté. L'examen histopathologique a révélé la présence de vaisseaux veineux, d'artérioles, de veinules ectatiques dilatées bordées d'un revêtement endothélial plat, de vaisseaux lymphatiques et de muscle lisse. L'étude immunohistochimique a montré une positivité aux marqueurs vasculaires. Sur la base d'analyses de laboratoire, un diagnostic de malformation vasculaire de la parotide salivaire a été posé.

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Keywords: parotid gland, neoplasm, surgery, vascular anomalies.

Conclusions. Les anomalies vasculaires des glandes parotides chez l'adulte doivent être considérées comme des hyperplasies, qui peuvent être facilement retirées sans endommager la structure de la glande, car elles sont plus fréquemment localisées à sa périphérie.

Mots-clés: glande parotide, néoplasme, chirurgie, anomalies vasculaires.

Introduction

The parotid gland is the most common site for tumours of the salivary glands, which are composed of epithelial, lymphatic tissues, and vessels. In the literature, all vascular neoplasms are generally classified into a common group – hemangiomas. Hemangiomas are benign tumours of vascular origin characterized by enhanced proliferation and renewal of endothelial cells[1,2]. Hemangiomas can occur anywhere on the body, but 65% of them are located in the head and neck region. They mostly affect the salivary glands, with the parotid gland being the most common site of involvement (81-85%)[1,3]. Hemangiomas in adults account for only 0.4% to 0.6% of all parotid gland tumours[4]. Hemangiomas of the parotid glands are usually encountered in children and account for approximately 50% of parotid gland tumours that develop during the first year of life[3]. Most hemangiomas of the parotid glands develop during the first 6-8 months of infancy, and then involution occurs during the first decade of life[5]. On the contrary, hemangiomas of the parotid glands in adults are quite rare and usually do not regress spontaneously. In infants, hemangiomas typically manifest externally as reddish or purplish skin lesions[3]. In adults, there is asymptomatic swelling of the parotid gland[6,7].

However, vascular anomalies of the parotid gland still occur in adults. Vascular anomalies remain one of the least studied entities encountered in clinical practice. The general term "angioma" is still used for both tumours and vascular malformations[8,9]. Vascular tumours can be benign, locally aggressive or malignant. Vascular malformations are divided into capillary, lymphatic, venous, arteriovenous malformations, or arteriovenous fistulas[10-12]. Vascular anomalies encompass a spectrum of disorders, ranging from simple "birthmarks" to life-threatening tumours.

The diversity of the clinical presentation, the course of this type of vascular tumours and treatment

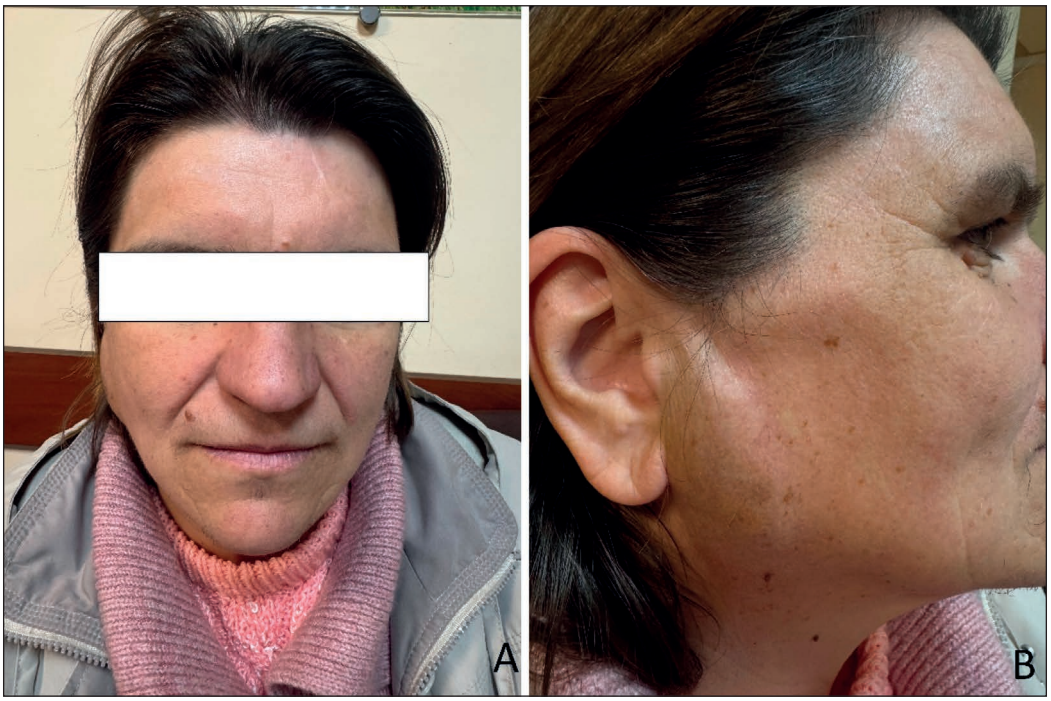


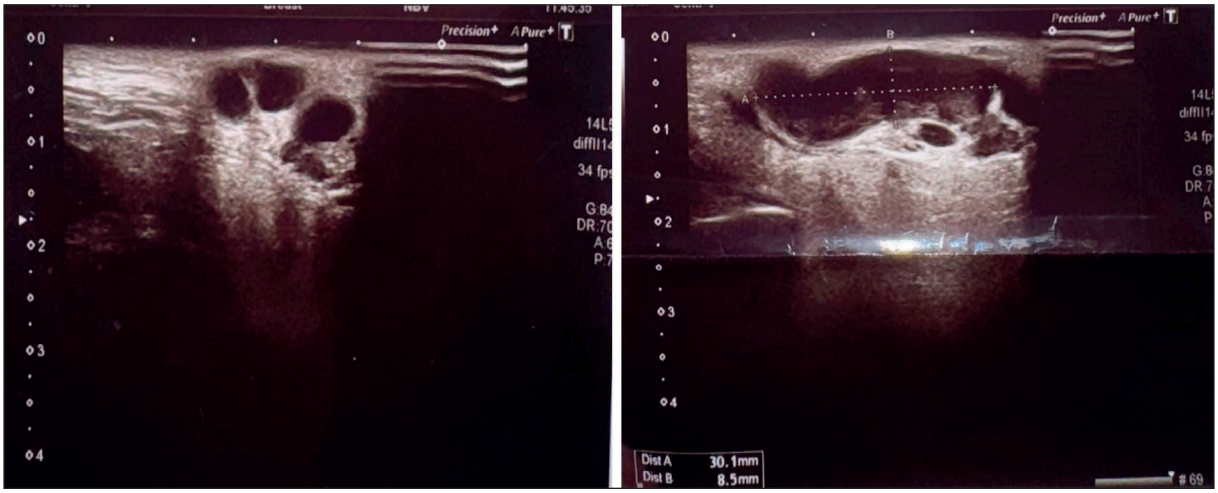
Figure 1. A 50-year-old patient. Preliminary diagnosis: neoplasm of the right parotid gland. a) Right facial asymmetry; b) Swelling in front of the right earlobe.

methods have led to the need to create a new "biological" classification. The International Society for the Study of Vascular Anomalies has introduced a unified classification that divides vascular lesions into vascular malformations (morphogenetic defects) and proliferative vascular lesions (tumours)[13,14]. Furthermore, in 2011, it was suggested to include a separate nosological form – vascular hyperplasia – in the classification of pathological formations of blood vessels (Roginsky, 2011). Therefore, in the diagnosis and treatment planning, it is necessary to differentiate the approach according to these types.

A separate group of vascular anomalies is represented by neoplasms located in the parotid gland, with the need to address the issue of the volume of surgical intervention (parotidectomy, gland resection, excision of the tumour) or conservative treatment (beta-blockers, hormone therapy, etc.)[15-20].

Case presentation

A 50-year-old woman presented to the department of head and neck tumours of the communal non-profit enterprise "Podilskyi Oncology Center" of the Vinnytsya Regional Council, Ukraine, with complaints of facial asymmetry and swelling in front of the right ear lobe, which gradually increased in size. This tumour has been observed for 9 years. During the examination of the patient, a soft, painless, non-fluctuating, well-defined tumour, approximately 3 cm x 4 cm in size, was found in the right parotid area (Figures 1a and 1b). The swelling was compressive and non-pulsatile. There were no signs



a b

Figure 2 a and b. Ultrasound of the right parotid salivary gland.

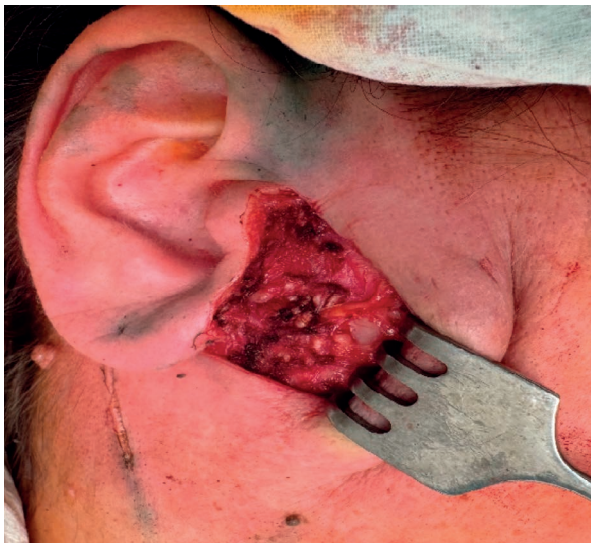


Figure 3. Intraoperative photograph a fter removal of the neoplasm.

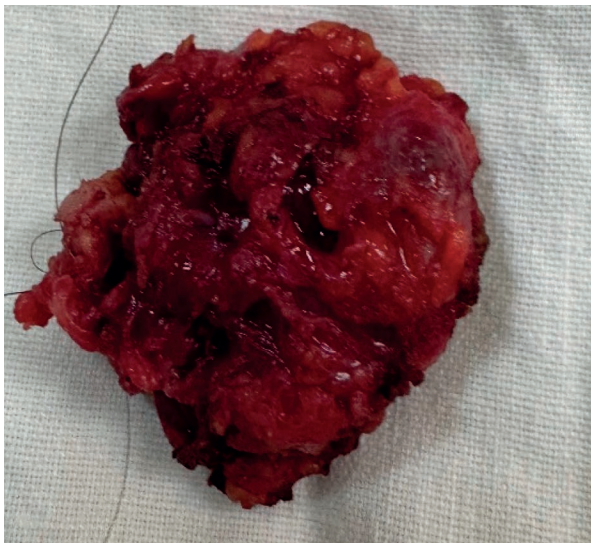


Figure 4. Macroscopic examination of the neoplasm of the right parotid salivary gland.

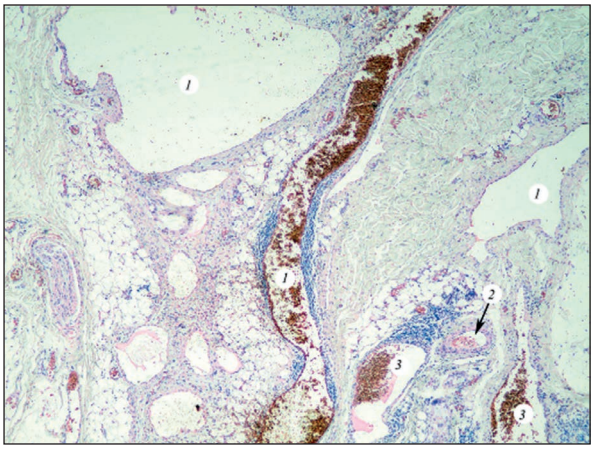


Figure 5. Vascular malformation of the parotid salivary gland. Predominant venous vessels (1) of various calibers, vascular bundle: arteriole (2), accompanying venules (3). Hematoxylin-eosin x40.

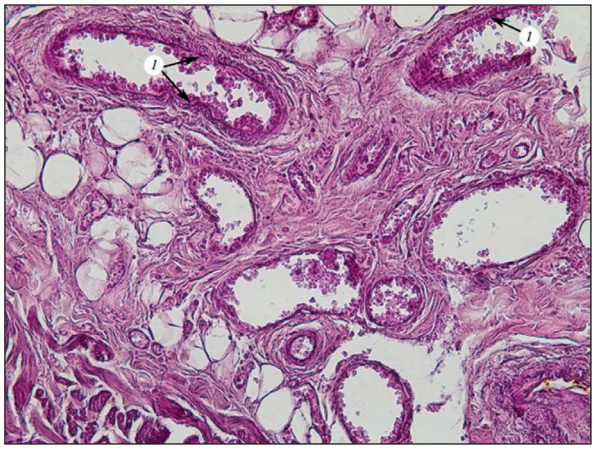


Figure 6. Vascular malformation of the parotid salivary gland. Blood vessels with the presence of an interrupted internal elastic membrane in their wall (1). Resorcinol-fuchsin according to Weigert x200.

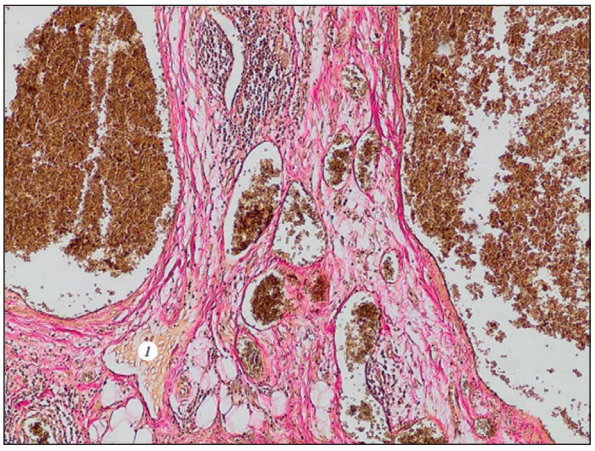


Figure 7. Lymphatic vessel (1) as part of a vascular malformation of the parotid salivary gland. Hematoxylin-eosin x100.

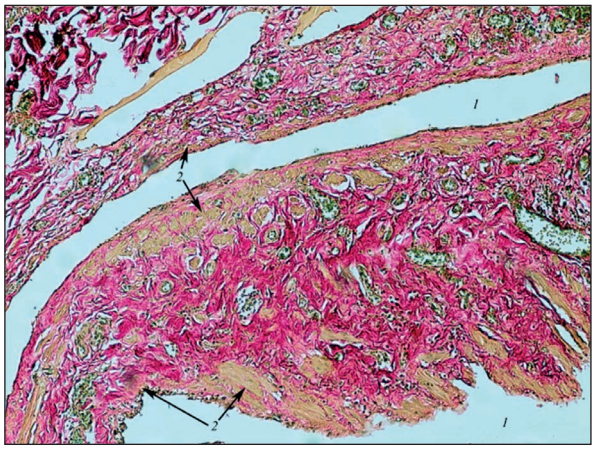


Figure 8. Vascular malformation of the parotid salivary gland. Lumen of uneven width (1), different number of smooth muscle fibers (2) in the wall of venous vessels. Van Gieson x100.

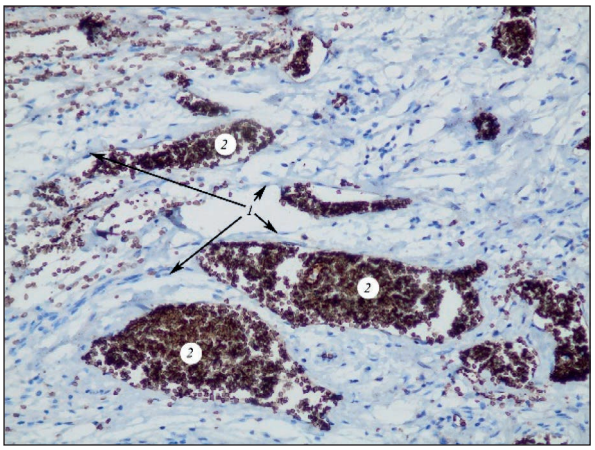


Figure 9. Vascular malformation of the parotid salivary gland. No expression of the GLUT1 marker in the endothelium of tumour vessels (1), positive expression in erythrocytes in the lumen of the vessels x200.

of noise or pulsation above the swelling. The skin over the tumour was unchanged. There was no lymphadenopathy.

During ultrasound examination of the soft tissues in the right parotid gland, hypoechoic areas measuring from 10.6x8.6 mm to 30.1x8.5 mm and anechoic areas measuring from 4.7x4.5 mm to 8.7x6.3 mm were visualized. The contour was smooth and clear. The echogenicity was reduced (Figures 2a and 2b).

During the puncture biopsy, a fluid of brown-red color was obtained. The cytological examination revealed endothelial cells and macrophages.

The patient was scheduled for resection of the right parotid salivary gland. During the surgical intervention, diffuse vascular dilation of the superficial portion of the right parotid gland was observed.

Upon removal, large feeding vessels could not be identified, and blood loss during the surgery was minimal. The neoplasm was successfully removed without compromising the integrity of the parotid gland (Figure 3).

The excision of a newly formed dark red, soft-elastic mass was performed. Macroscopically, it was a lobulated tumour measuring 3x3 cm (Figure 4).

The histopathological examination showed the presence of venous vessels, arterioles, extended ectatic venules lined with flat endothelial lining (Figures 5 and 6), lymphatic vessels and smooth muscles in the wall of venous vessels (Figures 7 and 8). The immunohistochemical examination showed positivity of vascular markers (Fig. 9).

The postoperative evolution of the patient was without complications.

Discussion

The classification of vascular lesions has been the subject of debate for many years. Prior to the 1980s, the vascular lesions were referred to as hemangiomas. In 1982, Mallikin and Hlowatsky first classified the vascular lesions into hemangiomas and vascular malformations based on endothelial characteristics[21]. The terms "hemangioma" and "vascular malformations" have been used interchangeably for the past few years[22].

Vascular malformations of the parotid gland are extremely rare conditions, with approximately 50 cases reported in the literature[9]. In most studies, the incidence in women is higher than in men. In most cases, the anomalies are located only in the superficial area of the parotid gland. Vascular anomalies manifest as painless, slow-growing soft tissue enlargements. Typically, there are no associated symptoms, and patients often seek medical treatment for cosmetic purposes. There are no signs of facial nerve involvement, lymph node enlargement, or skin infiltration. The diagnosis can be made clinically, but since this condition is very rare, a precise imaging visualization is necessary. Most often, only the superficial part is affected, but in some cases, the entire parotid gland may be involved[8,11,22,23].

Ultrasonography can detect vascular anomalies, but in some cases it may be less informative. Therefore, magnetic resonance imaging (MRI) is primarily the method of choice in complex diagnostic cases, and MRI angiography may also be indicated[5].

The main goal of treatment is the rational removal of the neoplasm, restoration and preservation of parotid function, and correction of the cosmetic effect. There are various treatment options available, such as laser therapy, cryotherapy, embolization, and corticosteroid treatment, but since the diagnosis is often unclear in most cases, surgical treatment remains the recommended method of treatment.

Conclusions

Vascular anomalies of the parotid gland that occur in adulthood with a higher likelihood may be classified as vascular hyperplasias, which are benign neoplasms. Vascular anomalies may be located at the periphery of the parotid gland, allowing for organ-preserving surgeries without disrupting the gland's structure.

Author Contributions

A.A.K. was responsible for the clinical diagnostic, diagnostic procedures, treatment decisions, performed the surgery and wrote the manuscript. S.M.S. provided diagnosis, conceptualization, methodology.H.I.K.- original draft preparation, review, editing. All authors have read and agreed to the published version of the manuscript.

Compliance with Ethics Requirements:

"The authors declares no conflict of interest regarding this article.

The authors declares that all the procedures and experiments of this study respect the ethical standards in the Helsinki Declaration of 1975, as revised in 2008(5), as well the national law." "Informed consent was obtained from the patient included in the study."

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