Myoepidermal cell carcinoma of the deep lobe of parotid gland 16 years after superficial parotidectomy due to basal cell adenoma of the superficial lobe – case report

Rak mioepidermalnokomórkowy płata głębokiego ślinianki przyusznej 16 lat po parotidektomii powierzchownej z powodu gruczolaka podstawnokomórkowego płata powierzchownego – opis przypadku

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ABSTRACT: The authors present a case of a 65-year-old female who had a superficial parotidectomy due to basal cell adenoma in 2004. In 2020, the remaining portion of the parotid gland with tumor was resected. Postsurgical histopathologic assessment revealed myoepidermal cell carcinoma. The authors also describe the clinical characteristics of the neoplasm and multiple occurrences of different tumors in the same parotid gland.

KEYWORDS: basal cell adenoma, myoepidermal cell carcinoma, parotid gland

STRESZCZENIE: W niniejszej pracy przedstawiono przypadek 65-letniej chorej, u której w 2004 r. wykonano parotidektomię powierzchowną z powodu gruczolaka podstawnokomórkowego, a w 2020 r. usunięto pozostałości ślinianki przyusznej wraz z guzem, rozpoznanym w pooperacyjnym badaniu histopatologicznym jako rak mioepidermalnokomórkowy. Ponadto omówiono charakterystykę kliniczną nowotworów oraz mnogie występowanie różnych guzów w tej samej śliniance.

SŁOWA KLUCZOWE: gruczolak podstawnokomórkowy, rak mioepidermalnokomórkowy, ślinianka przyuszna

INTRODUCTION

The available literature contains thorough descriptions of diagnostic and therapeutic conduct in parotid tumors. Imaging diagnostics (X-ray, CT, MRI) is the necessary means for assessing nodular lesion of the salivary gland; it allows to determine tumor size, location, and relation to the facial nerve. The diagnostic value of fine needle biopsy remains controversial. In contrast, superficial parotidectomy which represents the basic surgical intervention in benign tumors of the parotid gland is currently not a topic of discussion. Difficulties in reoperating the parotid gland after previous parotidectomy due to adhesions and in locating the trunk and branches of the facial nerve are another problem.

In fact, the literature contains reports of synchronous tumors in the same salivary gland, although such lesions are rare. However, we did not find a case report of two tumors in the same parotid gland (in the case of a second tumor in the preserved deep lobe of parotid gland) several years apart.

CASE REPORT

Female patient B.W. aged 65 was admitted to the clinic on May 18, 2020 with a tumor of the right parotid gland. Comorbidities included multiple sclerosis. In November 2004, the patient was diagnosed with a tumor of the right parotid gland. Ultrasound examination revealed: “a 31 x 17 x 17 mm foci in the middle and lower portion of the parotid gland (...) The lesion with clear borders has patterns of calcifications (...) The remaining salivary glands show no pathologies”. On BAC: “the image indicates a glandular...
A round lesion of 24 x 23 mm with numerous calcifications at the center, which may correspond to adenoma – the lesion is adjacent to the mandibular branch” (A. Ruciński MD PhD). Diagnostics was extended to include CT scan (March 2020), which revealed: “A tumor of approx. 2 x 2.5 cm in the upper pole of the right salivary gland between the mandibular branch and the mastoid, below the external auditory canal; heterogeneous, solid, with strong contrast-enhancement patterns after contrast and numerous calcifications in the center. The tumor’s external borders are difficult to trace segmentally. Tumor nature is unclear, possible proliferation. Several lymph nodes, up to about 5 mm, visible on the tumor periphery. Also, ambiguous lymph nodes within all nodal groups on the right side of the neck, with stronger post-contrast enhancement than the others. They may be metastatic. On the side of the left neck, small nodes, difficult to interpret (...) On neurological examination: The tumor of the right parotid gland raises the suspicion of an aggressive lesion. Ambiguous nodal changes on the neck, bilaterally” (E. Krakus MD PhD).

The patient had right-sided facial palsy which lasted for several weeks in the postoperative period. It was profound at first, but gradually subsided until it healed completely.

After the procedure, the patient underwent periodic laryngological and systematic neurological follow-up (MRI of the head every year due to multiple sclerosis) which revealed no tumor recurrence.

In February 2020, subsequent follow-up MRI of the head revealed: “A 21 x 18 mm lesion in the right parotid gland covered only in a single sequence – to be assessed in ultrasound – lymph node/ focal lesion” (K. Skrobisz MD PhD). On ultrasound in March 2020: “the upper residual pole of right parotid gland has a dorsal round lesion of 24 x 23 mm with numerous calcifications at the center, which may correspond to adenoma – the lesion is adjacent to the mandibular branch” (A. Ruciński MD PhD). Diagnostics was extended to include CT scan (March 2020), which revealed: “A tumor of approx. 2 x 2.5 cm in the upper pole of the right salivary gland between the mandibular branch and the mastoid, below the external auditory canal; heterogeneous, solid, with strong contrast-enhancement patterns after contrast and numerous calcifications in the center. The tumor’s external borders are difficult to trace segmentally. Tumor nature is unclear, possible proliferation. Several lymph nodes, up to about 5 mm, visible on the tumor periphery. Also, ambiguous lymph nodes within all nodal groups on the right side of the neck, with stronger post-contrast enhancement than the others. They may be metastatic. On the side of the left neck, small nodes, difficult to interpret (...) On neurological examination: The tumor of the right parotid gland raises the suspicion of an aggressive lesion. Ambiguous nodal changes on the neck, bilaterally” (E. Krakus MD PhD).

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examinations and clinical picture, the patient was admitted to the clinic and qualified for surgery. Physical examination revealed no other abnormalities apart from the scar after the previous parotidectomy. Function of the facial nerve was normal on both sides for all branches. On May 19, 2020, the patient underwent total parotidectomy. The remnants of the superficial lobe and deep lobe of the right parotid gland were excised together with the tumor in the parapharyngeal space. Preauricular incision was made. The ‘pointer’ of the cartilage of the external auditory canal and the posterior belly of the biventer muscle were found, but the attempt to reach the stylomastoid foramen and find the facial nerve trunk within the submandibular triangle was impossible due to very strong adhesions that prevented safe, gentle preparing for the pace. Therefore, the mastoid was opened with a drill, revealing the facial nerve trunk. It was exposed to the outlet within the stylomastoid membrane. From there, the trunk and branches of the facial nerve were dissected and separated from the tumor directly deep under the nerve, penetrating into the parapharyngeal space. Group II and III lymph nodes were also excised. On postoperative histopathological examination: Myoepithelial cell carcinoma (Carcinoma myoepitheliale) of the right parotid gland. Size change to 1.6 cm with an irregular outline, and a focal shallow infiltration of the surrounding tissues and single blockages in the adjacent lymph vessels” (dr. R. Lenckowski).

The patient was treated with postoperative radical radiotherapy at a dose of 60 Gy in 30 fractions in the period 17.07.–25.08.2020. After the treatment, the patient was found with gradually subsiding deep facial nerve paralysis in all branches (grade V on the House-Brackmann scale), which subsided completely after about 3 months.

Follow-up MRI on December 1, 2020: “Signs of scarring on tumor bed. No other pathological tissue masses or areas of diffusion restriction – radiologically without recurrence. The tumor bed is reconstructed by adipose tissue at the site of the right parotid gland” (A. Gajdowski MD PhD).

**DISCUSSION**

Basal cell adenoma was first described by Kleinasser and Klein in 1967. It is a rare tumor, most often located in the parotid gland. Basal cell adenomas constitute 1–2% of all salivary gland tumors of epithelial origin. There are six histopathological types of basal cell adenoma: solid, tubular, trabecular, membranous, cribriform, and myoepithelial-derived stroma rich [1]. González-García R. et al. [2] report that adenoma is characterized by multifocal hyperplasia and a more frequent tendency to relapse than in the case of polymorphic adenoma.
Myoepithelial cell carcinoma is a rare salivary gland tumor. The clinical course is still not known due to the small number of reports describing long-term follow-up [3]. Goode et al. [4] examined a large number of 234 patients and found that 75% had been cured, 9% had relapsed, 5% had metastases but survived after additional surgery, and 25% had died. The authors mentioned poor prognostic factors like patient’s age, tumor size, histological structure (a large number of division figures and a low number of cystic components), nerve infiltration, the presence of necrosis and anaplasia. The primary treatment for myoepidermal cell carcinoma of the parotid gland is excision of the entire salivary gland. On the other hand, Zenga et al. [5] assessed the effectiveness of postoperative radiotherapy in patients with myoepidermal carcinoma of the parotid gland. Out of 19 cases with a small surgical margin (less than 2 mm), 15 were only followed-up, and 4 underwent supplementary irradiation. The authors found no difference in the prognosis of 5-year survival in both groups.

**Fig. 4.** (A-B) Facial nerve function in the presented case 6 months after total parotidectomy. Normal function of the facial nerve in all twigs.

**Fig. 5.** (A-C) Follow-up MRI in the presented case 5 months after total parotidectomy. The right parotid bed is visibly remodeled by adipose tissue.
Descriptions of synchronous tumors can be found in both Polish and international literature [6–9]. The Polish literature also mentions a case similar to that presented by Bień et al. [8]. Nonetheless, we did not find a case report that would describe histopathologically different tumors of the salivary glands several years apart in the same parotid gland.

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