GIANT SUBMANDIBULAR GLAND TUMOR: MUCOEPIDERMOID CARCINOMA WITHIN THE PLEOMORPHIC ADENOMA (MIXED TUMOR)

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Pleomorphic adenoma (mixed tumor) is the most common type of salivary gland tumors, most often involving the parotid glands. The study presented a case of a giant pleomorphic adenoma of the submandibular gland in a 78-year old woman. Due to the initially slow and progressive growth (during a period of 25 years) without treatment, the tumor attained a size of 25x19x16 cm and a weight of 4 kg. The patient underwent surgery. The histopathological examination showed that the tumor was subject to malignant transformation: within the pleomorphic adenoma one observed mucoepidermoid carcinoma lesions of high-degree malignancy. In spite of the good postoperative esthetic and functional effect the patient was diagnosed with distant metastases to the lungs.

Key words: pleomorphic adenoma, mucoepidermoid carcinoma, neoplasms of the submandibular gland

The pleomorphic adenoma is one of the most common tumors of the small and large salivary glands. As one of the few benign tumors it may be subject to malignant transformation (1, 2).

In most cases the mixed tumor develops in the parotid glands (66-96%). Its location in the submandibular salivary glands is 8-fold less frequent (12%). The most rare location includes the sublingual gland, small salivary glands of the mucous membranes of the oral cavity, throat, lips, maxillary sinuses, and bronchi (nearly 3%) (4, 5).

Some of the submandibular gland tumors are considered as malignant. Warning signals of potential tumor malignancy include the following: accelerated growth, immobilization in relation to surrounding tissues and skin, retraction, ulceration of the skin covering the tumor, pain, lymphadenopathy within the local lymph drainage system, hypoglossal nerve (tongue paresis) or lingual nerve paresis (dysesthesia within the tongue), facial nerve paresis, and ichoroid secretion.

Surgical treatment is the basic therapeutic method in case of salivary gland tumors. Primary surgery includes the complete excision of the salivary gland with the tumor. If the tumor infiltrates surrounding tissues mandibular and submandibular lymph nodes resection is required. Surgery of submandibular gland tumors is as follows: simple resection of the gland, submandibular gland and submandibular fossa content resection, as well as gland excision with surrounding structures. The efficacy of the first surgical intervention is decisive.

Postoperative radiotherapy is indicated after the excision of the malignant tumor, which infiltrates the capsule or surrounding tissues. Radiotherapy is also indicated in case of surgical contraindications, lack of patient consent to surgery, and as a palliative therapeutic method (6). In case of III and IV degree clinical stage tumors surgical resection is individual. Considering patients with clinically confirmed lymph node metastases radical surgery of the cervical lymph nodes is recommended with adjuvant radiotherapy.
CASE REPORT

A 78-year old female patient was admitted to the Department of General Surgery, Regional Hospital in Sucha Beskidzka, due to a giant cervical tumor.

Eight years earlier the patient was diagnosed at the Department of Internal Medicine, due to a cervical tumor, which was 8cm in diameter. The patient was diagnosed with follicular adenoma. However, she did not report for elective surgery.

For the first time the patient noticed the appearance of the tumor 25 years ago. During the initial 16 years tumor growth was slow and did not interfere with her everyday activities. The patient got used to its presence. For the next eight years the tumor significantly grew in size, and during recent years, one observed the appearance of a suppurating, bleeding and emitting an unpleasant odor, ulceration on its lateral wall. The tumor began to interfere with the patients’ daily life. The patient lived in poor social conditions with an alcoholic son and lacked proper care.

The patient reported to the hospital, due to general weakness, shortness of breath, and right-sided chest pain caused by the compression of the tumor.

The patient was admitted to the hospital in poor general condition with symptoms of cachexia (20kg weight reduction during the past year). The physical examination showed the presence of a giant submandibular, irregular tumor, 25 cm in diameter with a 10 cm ulceration of the skin infiltrating the face and compressing the chest. Over the lungs there was no vesicular murmur below the right shoulder angle and dullness in the area. The chest X-ray was as follows: presence of a tumor in the right lung, the left lung being without pathology. The ultrasound examination showed the presence of a right pleural effusion. A pleural tap was performed and 1400 ml of a dark-yellow fluid was obtained. Pleural effusion cytology showed no abnormalities. BAC of the submandibular tumor was performed. The histopathological examination revealed the presence of a parotid-like gland malignancy.

The patient was qualified for surgery. During the surgical procedure a right submandibular gland lesion, 25x19x16 cm in size and weighing 4 kg was excised. Postoperative drainage was required. Wound healing per primam without complications was observed. The sutures were removed on the seventh postoperative day.

During hospitalization facial, lingual and hypoglossal nerve disturbances were not observed. Proper wound healing was observed. On the second day after the operation right pleural cavity drainage was performed, due to the presence of a pleural effusion- 2100 ml of serous fluid was obtained. On the sixth day after the operation the drain was removed- the patient was without pain and shortness of breath. Two days later fluid accumulation was once again observed. The patient was transferred to the Department of Internal Medicine. A chest X-ray was performed showing left pleural cavity effusion and possibility of left pulmonary metastases. CT of the chest demonstrated metastatic lesions in both lungs and enlarged periaortic and tracheal bifurcation lymph nodes- characteristic of cancer dissemination.

The histopathological examination of the tumor revealed the presence of a mucoepidermoid carcinoma within the pleomorphic adenoma: malignant mixed tumor of the salivary gland- high-grade mucoepidermal carcinoma. Multiform adenoma texture predominated with mucoepidermoid carcinoma foci. One observed necrotic lesions and blood extravasations. The tumor infiltrated the skin causing ulcerations. The base of the tumor was composed of a fibrous capsule, which the tumor did not exceed (examination nb.14430).

Based on the clinical picture the patient was diagnosed with T4N1M1 cancer (stage IV). The patient was directed for further treatment (radiotherapy), which she did not report too.

DISCUSSION

The presented case deserves special attention, due to the atypical location of the tumor and long-time growth, during which there was malignant transformation of the pleomorphic adenoma.

In most cases the mixed tumor develops in the parotid glands. Its location in the submandibular salivary glands is 8-fold less frequent (12%). The tumor affects patients of all ages, more often women in the fifth and sixth decades of life. Malignant transformation concerns 1.9% to 23.3% of all pleomorphic adenomas (7). The risk of malignant transformation increases with the duration and progres-
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A tumor growing slowly over many years, followed by rapid growth in recent times is a classic example of the clinical course of cancer developing within the mixed tumor (9).

Most often the malignant mixed tumor is diagnosed in the seventh and eighth decades of life. The above-mentioned tumor is not encapsulated, being partially necrotic with hemorrhagic effusions, cysts and perineural infiltration. Lymph nodes metastases are diagnosed in 15% and distant metastases in 30% of patients.

Submandibular gland carcinomas are rare malignant neoplasms representing less than 1% (10) of all malignant neoplasms of the head and neck, and 2.5-7.5% of all salivary gland tumors (submandibular gland tumors constitute 50%). Due to the rare occurrence of the above-mentioned little information concerning survival and tumor predicting factors can be found in medical literature. Neil Bhattacharyya et al. demonstrated that mean survival in case of patients with diagnosed mucoepidermoid carcinomas amounted to 78 months, while the five-year survival was 55.4% (10). According to Stanisław Bien cancer within the pleomorphic adenoma is one of the worst prognostic tumors of the salivary glands (apart from ductal and adenoid cystic carcinomas), where ten-year survival does not exceed 30% (6). Mucoepidermoid carcinoma is the second most common neoplasm of the submandibular gland.

The stage of the disease is an important prognostic factor in case of mucoepidermoid carcinoma of large salivary glands (11). High malignancy cancer is more likely to develop distant metastases and thus, is burdened with worse prognosis (12). The above-mentioned tumor has the ability of infiltrating tissues along the lymphatic and blood vessels. This leads to the development of metastases in the lymph nodes, skin, bones, liver and brain (12). Local lymph nodes metastases are observed in 20-30% of cases (13, 14).

In case of our patient we observed all the characteristics of malignant tumor transformation.

The treatment of choice in case of multiform adenoma is to excise the tumor along with the salivary gland. The patients’ concern considering surgery and the difficult social condition were the reasons for not arriving to planned surgery eight years ago. This enabled the mass of the tumor to grow to such large sizes. If surgery was performed in the slow tumor growth stage malignant transformation could have been avoided. The best therapeutic and preventive method considering cancer within the pleomorphic adenoma is the radical excision of the salivary glands tumor. It is important to raise the awareness of the patient of the possibility of malignant transformation of the pleomorphic adenoma, as well as the fact that final histopathological evaluation is possible only after tumor excision. BAC in the differentiation of cancer is insufficiently sensitive (15).

REFERENCES


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