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Case Report

Long-Term Recurrence-Free Survival After Salivary Duct Carcinoma of the Parotid: A 10-Year Follow-Up Case Report

D. Schneider¹ and P.W. Kämmerer^{2*}

¹Private Praxis, Rostock, Germany

²Department of Oral, Maxillofacial and Plastic Surgery, University Medical Center Mainz, Mainz, Germany

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ABSTRACT

Salivary duct carcinoma (SDC) represents a very rare and aggressive parotid neoplasm. A 70-year-old male was admitted to the hospital with a swelling of the right parotid region, persisting for twelve weeks. Based on sonography and contrast-enhanced computed tomography, a suspicious lesion showing mass expansion and invasion of surrounding tissues was seen. Intraoperative biopsy brought evidence of SDC. In accordance, surgical treatment included parotidectomy with preservation of the facial nerve as well as ipsilateral neck dissection due to suspicious lymphatic nodes. Afterwards, an adjuvant radiation therapy was undertaken. At 10 years of follow-up, the patient was alive and free of recurrence with full function of the facial nerve. In conclusion, keeping in mind its poor prognosis, the rare parotid salivary duct carcinoma needs aggressive therapy consisting of a surgical as well as a radiation therapy approach.

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Introduction

Salivary duct carcinoma (SDC) is characterized as an aggressive tumor with early metastasis, local recurrence, and significant-high mortality. Histopathologically, there is a resemblance to the ductal carcinoma of the breast. A total of 85% of the cases occur in the parotid gland, followed by the submandibular gland (approximately 8% of cases) [1]. A single case was reported with an arising of the sublingual gland [2]. Among all salivary gland tumors, the incidence of SDC is 1-3% [1]. The goal of the present case report was to represent the course of patients' disease and discuss the relevant current literature.

Case Report

A 70-year-old male was referred to the Department of Oral, Maxillofacial and Plastic Surgery, University Medical Centre Mainz, Germany, with a suspicious parotid gland swelling of the right half of

the face (Figure 1). At this time, the hardened, not painful tumor had a size of about 3 x 3cm. The swelling had been apparent without further increase in size for 12 weeks. Sensibility and motoric function were not impaired. Constitutional symptoms were not seen. In the ultrasound sonography examination, a homogenous dense, non-encapsulated lesion with dorsal acoustic shadow was seen (Figure 2). For staging, the patient was examined using computed tomography (CT) (Figure 3), showing a heterogeneous tumor at the bottom on the right parotid. Furthermore, a suspicious lymph node was seen on the right side of the neck (level IIA in accordance with the classification of Robbins; Figure 4).

The primary surgical treatment for the patient consisted of a total parotidectomy conserving the facial nerve and an ipsilateral neck dissection (Figures 5 & 6), as the intraoperative histology suggested a malignant tumor. In the final histopathological analysis, a moderate differentiated salivary duct carcinoma with lymphangiosis carcinomatosa was seen. Microscopically, comedo-type necrosis,

*Correspondence to: PD Dr. med. Dr. med. dent. Peer W. Kämmerer, M.D., D.M.D., M.A, Ph.D., Department of Oral, Maxillofacial and Plastic Surgery, University Medical Center Mainz, Augustusplatz 2, 55131, Mainz, Germany; Tel: 00496131173572; Fax: 00496131176602; E-mail: peer.kaemmerer@unimedizin-mainz.de

calcification and cystic changes were also described (Figure 7). The resection margins (>5 mm) were free from residual neoplasia. There was no vascular involvement. The lymph node specimens contained malignant cells in two of thirteen lymph nodes. The final pathological staging was pT1, pN2b (2/13), pM0, G3, pR0. In accordance, an adjuvant radiation therapy with a total dose of 60 Gy was carried out. At 10 years follow-up, the patient was alive, free of recurrence and with the full motoric function of the right facial nerve.



Figure 1: Clinical picture of the patient at the first consultation. At this time, the hardened, not painful tumor was approximately 3 x 3cm. The swelling had been persistent for twelve weeks.

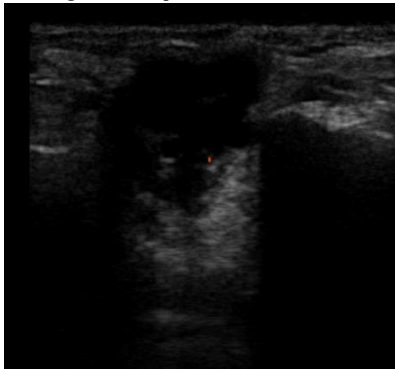


Figure 2: Ultrasound sonography of the dense lesion within the right parotid gland showing a dorsal acoustic shadow and infiltration in the surrounding tissue.

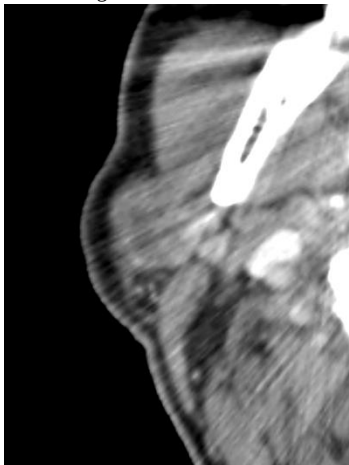


Figure 3: Contrast-enhanced computed tomography in axial direction showing a suspicious tumor in the parotid gland with infiltration of the surrounding soft tissue.

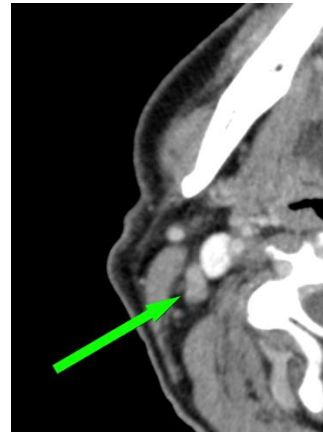


Figure 4: Contrast-enhanced computed tomography in axial direction showing a lymphatic node with signs of malignancy (green marking arrow).

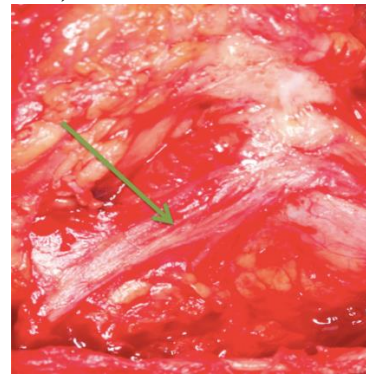


Figure 5: Intraoperative picture showing the site within the right parotid gland after resection of the SDC together with the preservation of the facial nerve (green marking arrow).



Figure 6: Macroscopic tissue after tumor excision.

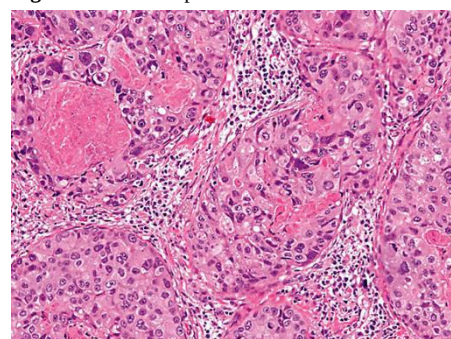


Figure 7: Histopathological specimen of salivary duct carcinoma showing cribriform ductal component with comedo necrosis (H & E; original magnification x5).

Discussion

Salivary duct carcinoma (SDC) is a highly aggressive tumor of the glands, mostly within the parotid [1]. Due to its aggressive behaviour (local relapses ~33%, early distant metastases ~46% and high mortality ~65%), a rapid diagnosis and treatment is obligatory. SDC occurs mainly in male (2:1 to 3:1) patients between 60 and 70 years of age [3]. Interestingly, other case reports demonstrated the occurrence of SDC in young patients as well [4]. In addition, other studies reported SDCs in locations other than salivary glands, like the larynx or the nose [5, 6]. In the present case, the tumor was located in parotid without any symptoms. In the contrary, Wee *et al.* reported facial nerve dysfunction or paralysis in more than a quarter of patients [7].

SDC arises from the larger intralobular and extralobular ducts. Same as the intraductal and infiltrative ductal carcinoma of the breast, SDC shows intraductal and infiltrating components [8]. Macroscopically, SDCs are non-encapsulated, poorly circumscribed, and multi-nodular tumors. Histologically, comedo-type necroses, calcifications and cystic changes are seen. In about 60% of the cases, perineural invasion and approximately in 30% of the cases, lymphovascular invasion are seen [7]. Furthermore, SDC usually metastasizes early in the regional lymphatic nodes. Salovaara and colleagues demonstrated that in 56% of the cases, lymphatic node involvement was seen [9]. In the present case, two of thirteen lymph nodes contained malignant cells. Frequent localizations of distant metastasis of the salivary duct carcinoma are liver, lung, bone, brain and skin. In the present case, no distant metastasis was found. The general five-year survival rate is 35%-65% and the main causes of death are distant metastases [1].

Treatment of SDC consists of surgical excision with tumor-free margins. The involvement of the facial nerve may have a negative influence on survival. Postoperative radiation therapy, as seen in the presented case, is recommended, whereas additional chemotherapy is reserved for distant metastatic forms. Negative prognostic factors include tumor size, lymph node involvement, distant metastases and HER-2/neu, which was negative in the presented case. For HER-2 positive SDCs, the application trastuzumab, a human monoclonal antibody, has shown promising results, even for cases with distant metastases [10].

The median overall survival is approximately 3 years. The five-year recurrence-free survival is about 30%. In accordance, SDC has shown to have the lowest 5 five years overall survival rate of all parotid carcinomas [1]. In the present case, the patient was alive and free of

recurrence at over 10 years of follow-up. The particularities of this case were the rarity of the salivary duct carcinoma, insidiously occurrence and the associated aggressiveness of the tumor.

Conflicts of Interest

None.

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