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(CASE REPORT)

Positive surgical margins and durable disease control: A case of adenoid cystic carcinoma of the parotid gland treated with adjuvant radiotherapy

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Abstract

The parotid gland, the main site of adenoid cystic carcinoma (ACC) among the major salivary glands, represents a clinically significant location, particularly when the deep lobe is involved. This case illustrates the clinical and evolutionary heterogeneity of parotid adenoid cystic carcinoma. Despite the presence of positive surgical margins, adjuvant radiotherapy achieved durable locoregional control.

We report the case of a 45-year-old woman who presented with left-sided otalgia and headaches evolving over a two-year period. Magnetic Resonance Imaging (MRI) revealed a mass arising from the deep lobe of the left parotid gland. Total parotidectomy demonstrated adenoid cystic carcinoma with positive margins but without perineural invasion. Exclusive adjuvant radiotherapy was administered. After three years of clinical and radiological follow-up, the outcome remains favorable, with no evidence of local recurrence or distant metastasis.

We present the clinical, radiological, and therapeutic challenges raised by this adenoid cystic carcinoma of the deep lobe of the parotid gland, then analyze its underlying mechanisms and compare our observation with data from the literature.

Keywords: ACC; Positive Margins; Radiotherapy; Prognosis

1. Introduction

Adenoid cystic carcinoma (ACC) is a rare malignant tumor of the salivary glands, accounting for nearly 10% of malignant salivary gland tumors. It occurs predominantly in the minor salivary glands, which represent approximately two thirds of cases, making the parotid gland a much less frequent site; moreover, only 2% to 3% of all parotid tumors, whether benign or malignant, are ACCs (1). This tumor is classically characterized by a paradoxical biological behavior, combining slow growth with insidious local aggressiveness (2).

The management of this disease is primarily based on radical surgery, often combined with radiotherapy and, in some cases, systemic treatment.

This case highlights the subtle presentation of adenoid cystic carcinoma of the parotid gland and the value of a combined approach involving surgery and radiotherapy when complete resection is not achieved.

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2. Case Presentation

A 45-year-old woman with no significant past medical history began experiencing left-sided otalgia associated with headaches toward the end of 2019. These symptoms were later accompanied by mild retromandibular swelling, which led to an initial consultation with a general practitioner in early 2022. Symptomatic treatment was prescribed, but no clinical improvement was observed.

She was subsequently evaluated by an otolaryngology specialist at a regional hospital. Head and neck computed tomography revealed enlargement of the left parotid gland, along with several ipsilateral jugulocarotid and subangulomandibular lymph nodes suggestive of an inflammatory process. This assessment was further complemented by cervicofacial MRI, which showed an enlarged left parotid gland harboring a suspicious multilobulated soft-tissue lesion, predominantly involving the deep lobe, with no clear demarcation from the normal parotid parenchyma, measuring 31 × 34 × 30 mm. In addition, a left level II lymph node measuring 8 × 10 mm was identified (figure 1). As part of the staging workup, chest computed tomography (CT) was also performed and was unremarkable.

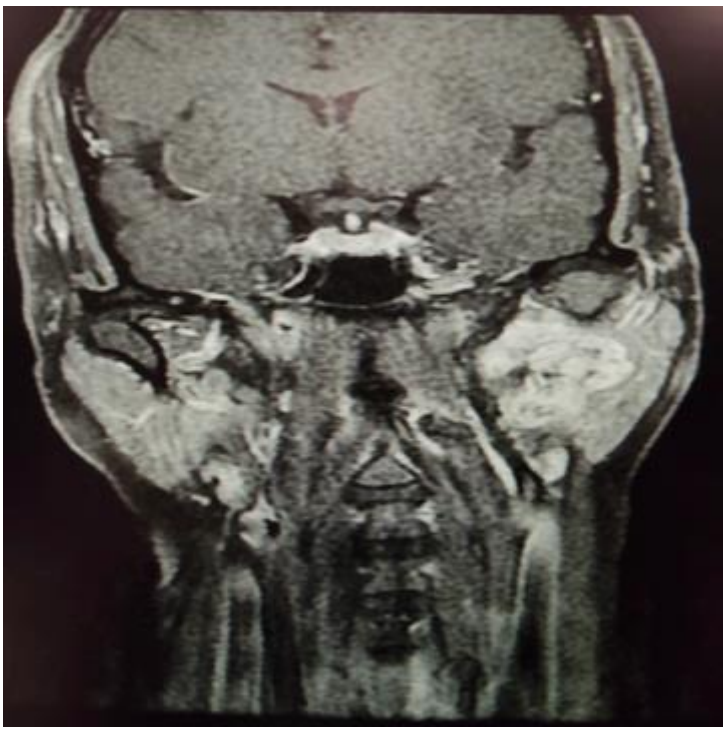

Figure 1 Cervicofacial MRI, coronal section, T1-weighted sequence after contrast medium injection, showing a suspicious mass arising from the deep lobe of the left parotid gland

The initial otolaryngological examination revealed a left retromandibular swelling that was firm in consistency, mobile relative to the superficial plane but fixed to the deep plane, and painful on palpation, with no local inflammatory signs, no peripheral facial nerve palsy, and no trismus. The bilateral cervical lymph node areas were free, and the intraoral examination showed no abnormalities.

The patient underwent a left total parotidectomy with level II adenectomy. Histopathological examination of the surgical specimen from the deep l lobe showed a tumoral proliferation composed of cribriform nests and trabeculae, made up of regular round cells with monomorphic, homogeneous, basophilic, round to oval nuclei, sometimes angular or triangular, and scant basophilic cytoplasm. The cribriform nests contained rounded lumina, sometimes filled with eosinophilic material. The stroma was fibrous. No perineural invasion was identified. A positive surgical margin was noted, leading to the diagnosis of adenoid cystic carcinoma.

It should be noted that the superficial lobe was free of tumor, and the adenectomy specimen was consistent with normal salivary gland tissue without lymphoid tissue.

After discussion at the multidisciplinary team meeting (MDT meeting), the therapeutic decision was to proceed with exclusive adjuvant radiotherapy, given the malignant nature of the tumor and the inadequate surgical margins.

As decided during the multidisciplinary team meeting, the patient underwent exclusive external radiotherapy using the intensity-modulated radiation therapy (IMRT) technique delivered to the high-risk clinical target volume (CTV) corresponding to the parotid tumor bed, with a total dose of 66 Gy according to a fractionation schedule of 2 Gray (Gy) per fraction. Furthermore, the ipsilateral nodal areas, including the course of the facial nerve as well as nodal levels Ib, II, III, and IV, were treated prophylactically to a total dose of 56 Gy, delivered in fractions of 1.8 Gy per fraction according to a simultaneous integrated boost (SIB) technique. An opinion from the medical oncology team was requested in order to discuss the value of concomitant chemotherapy with radiotherapy, but no indication was retained in this context.

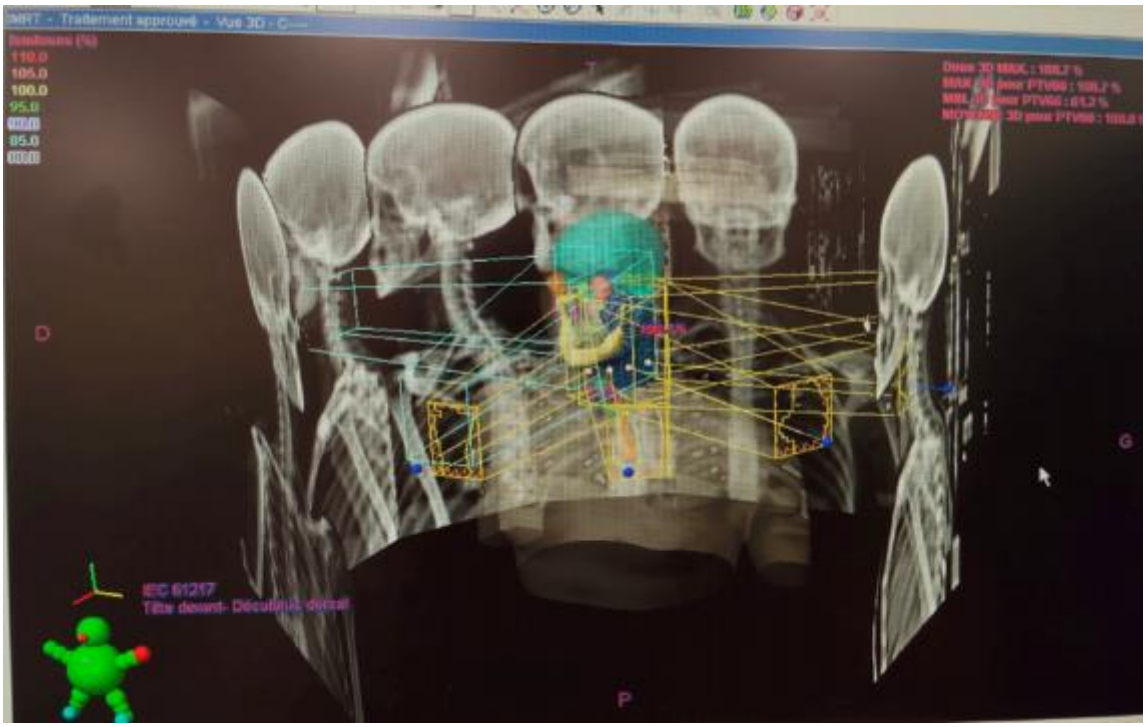

Figure 2 3D reconstruction of the IMRT treatment plan with beam arrangements and irradiated volumes (left parotid tumor bed and ipsilateral cervical lymph node regions)

Treatment was initiated in September 2022 and was completed in November of the same year.

Three years after the end of treatment, follow-up consultations and the latest cervico-thoracic CT scan showed no sign of local or metastatic recurrence, reflecting a favorable clinical course.

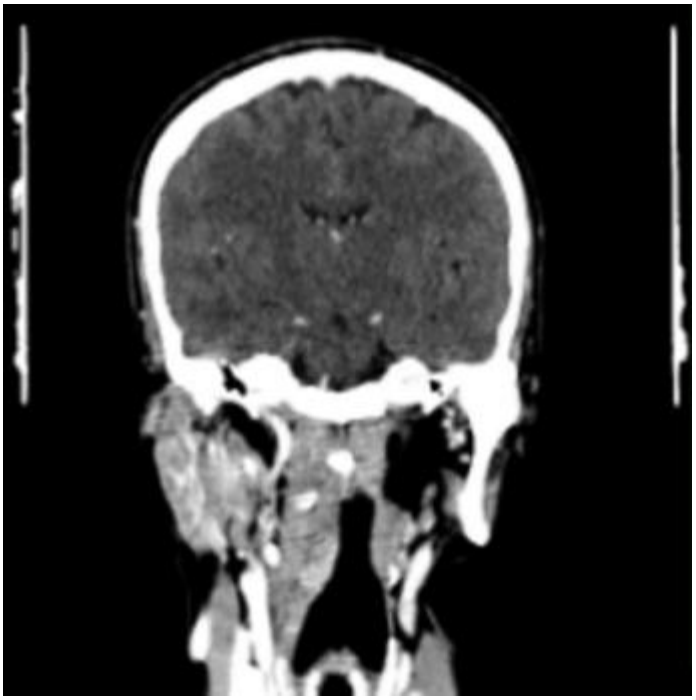

Figure 3 Coronal cervicocranial CT scan showing satisfactory locoregional control 3 years after completion of adjuvant radiotherapy

3. Discussion

ACC of the parotid gland accounts for a non-negligible proportion of malignant tumors of the parotid gland, representing approximately 15% of parotid cancers. However, it remains less frequent than other histological types more commonly encountered in this gland, particularly mucoepidermoid carcinoma (3). Although AdCC is a disease of middle-aged to older adults, typically occurring between the sixth and seventh decades of life, with a slight female predominance (4), in our case, the patient developed adenoid cystic carcinoma at the age of 45 years, which constitutes a relatively early presentation. Such an occurrence at a young age may be related to normal biological variability, genetic predisposition, early environmental exposures, molecular heterogeneity of the tumor, or transformation from a pre-existing lesion, although the literature does not allow the identification of a precise cause (4,5). Clinically, ACC often presents in a subtle manner, most commonly as a painless mass, which explains the frequent delay in diagnosis. Although its progression is slow, this tumor remains aggressive because of its perineural tropism, which is responsible for progressive local extension and sometimes for pain or neurological signs. Its natural history is marked predominantly by hematogenous dissemination, whereas lymph node metastases remain uncommon. This contrast between initially silent growth and aggressive behavior related to perineural invasion and the risk of late metastasis gives ACC an antinomic evolutionary profile (6). In our patient, the two-year interval between the first symptoms and the consultation fits within this pattern of subtle symptoms leading to delayed diagnosis.

The preoperative workup of our patient was consistent with the American Society of Clinical Oncology (ASCO 2021) recommendations (7), including cervicofacial MRI for locoregional assessment and chest computed tomography as part of the distant staging workup. These same recommendations (7) indicate that the preoperative diagnosis of parotid gland tumors, including adenoid cystic carcinoma, is based primarily on tissue biopsy. They state that fine-needle aspiration cytology (FNAC) or core needle biopsy should be performed before any surgical management in order to distinguish a benign tumor from a malignant one and to guide the therapeutic strategy (7). FNAC remains the most commonly used examination in practice, providing good diagnostic performance in parotid tumors, as reported by Taniuchi et al. (8). However, in ACC, its interpretation is sometimes complex and may lead to confusion with benign lesions or other low-grade tumors. This limitation is illustrated by the series reported by Sharma et al., who found a diagnostic accuracy of 82.7% for FNAC, showing that errors may still occur, particularly in solid forms (9). In this context, ultrasound-guided core needle biopsy appears to be a more accurate alternative, particularly for lesions suspected of malignancy or when FNAC is non-contributory, as demonstrated by Song et al. (10). The radiological meta-analysis by Kassem et al. (2025) also confirms the overall superiority of core needle biopsy for histological subtyping (11). Thus, preoperative histological proof now constitutes an essential element of the diagnostic process, although definitive confirmation of the subtype and grade may sometimes require examination of the surgical specimen when

preoperative samples are insufficient (7). In our patient, no preoperative biopsy had been performed, as she presented late and management was directed straight to parotidectomy. Analysis of the surgical specimen confirmed adenoid cystic carcinoma (ACC) with a cribriform architecture, the most frequently reported pattern and one generally associated with an intermediate prognosis according to Szanto et al. (12). Perineural invasion is present in 29.4% to 62.5% of cases according to Dantas et al. (13), and it is associated with an increased risk of local recurrence. This neurotropic behavior is considered one of the major determinants of the locoregional aggressiveness of ACC (6). In our case, the absence of perineural invasion is consistent with the biological variability described in the literature and constitutes a relatively favorable prognostic parameter. However, positive surgical margins constitute one of the most decisive prognostic factors in this disease, being associated with a significant increase in the risk of local recurrence and a reduction in locoregional control (14; 15; 16). Clinical practice guidelines, particularly those of ASCO and the National Comprehensive Cancer Network (NCCN), recommend postoperative radiotherapy in the presence of positive margins in order to optimize locoregional control (7; 17). In our case, the presence of positive margins led to the indication for adjuvant radiotherapy. Dose escalation to the tumor bed is classically recommended in order to optimize locoregional control, whereas clinically N0 cervical nodal areas are generally treated with a prophylactic dose intended to address possible subclinical disease while limiting toxicity to healthy tissues (18). Furthermore, the role of concurrent chemotherapy with radiotherapy in salivary gland tumors remains a matter of debate. Indeed, in the absence of solid evidence demonstrating a benefit in terms of locoregional control or survival, this strategy is not currently considered a standard therapeutic approach, and its value is still being evaluated in prospective trials (18). In our case, the absence of locoregional recurrence or distant metastasis after three years of follow-up reflects good locoregional control after combined surgery and radiotherapy, which remains the standard therapeutic strategy for localized adenoid cystic carcinomas of the salivary glands (19; 20). However, because of the risk of late hematogenous dissemination, mainly to the lungs, reported in several series, prolonged follow-up remains essential (18; 21; 22).

List of Abbreviations

- **ACC:** Adenoid Cystic Carcinoma.
- **MRI:** Magnetic Resonance Imaging Magnetic Resonance Imaging
- **CT:** computed tomography
- **MDT meeting:** multidisciplinary team meeting
- **IMRT:** intensity-modulated radiation therapy
- **ASCO :**American Society of Clinical Oncology
- **FNAC:** fine-needle aspiration cytology

4. Conclusion

This case illustrates the value of a multimodal approach combining surgery and adjuvant radiotherapy in adenoid cystic carcinoma of the parotid gland presenting risk factors, particularly positive surgical margins. The locoregional control achieved in our patient after three years of follow-up highlights the effectiveness of this therapeutic strategy, while also emphasizing that the unpredictable nature of this tumor requires prolonged clinical surveillance.

Compliance with ethical standards

Disclosure of conflict of interest

No conflict of interest to be disclosed.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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