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Case Report

Squamous-cell carcinoma after radiotherapy for a recurring pleomorphic adenoma of the parotid gland: A case report

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ABSTRACT

Pleomorphic adenoma (PA) is the most common tumor of the parotid gland. Even though PA is a benign tumor, it is associated with a significant risk of recurrence, especially if surgical resection is not sufficient. Since the introduction of parotidectomy with facial nerve identification, this recurrence risk has decreased from 20–45% down to 0.4–2% [1,2]. Nevertheless, the treatment of these recurrences, when they occur, is still controversial. Recurrence risk increases after the first episode of recurrence and is associated with a higher incidence of facial nerve damage during surgery and with a higher rate of malignant transformation. Radiotherapy has been proposed to increase local control of the tumor but using this treatment in young patients with a benign disease is controversial due to the risk of radio-induced cancers.

Here we report the case of a squamous cell carcinoma of the left tonsil occurring after radiotherapy for a PA of the left parotid gland.

1. Case report

A 62-year-old woman was referred to our maxillofacial surgery clinic for the management of a recurring PA (pleomorphic adenoma) of the left parotid gland. She had no history of smoking and drinking.

The patient had benefited from total left parotidectomy for a deep lobe PA 30 years prior. The facial nerve had been preserved. Ten years after the initial procedure, she presented with a recurrence of the PA and underwent secondary surgery. Pathology showed multinodular PA without malignant transformation. The tumor was in contact with the limits of the secondary resection. Adjuvant radiotherapy (55 Gy) was decided in order to avoid a second episode of recurrence.

In our clinic, 20 years after having benefited from adjuvant radiotherapy (so 30 years after the initial surgery), the patient presented with a lump regarding the parotidectomy scar. PA recurrence developed from residues of the initial tumor was suspected. Head and neck CT-scan and MRI showed a 14 mm lesion of the left parotid gland enhancing after contrast agent injection (Figs. 1 and 2). The aspect was in favor of PA recurrence. An independent 25 mm lesion of the left tonsil extending to the glosso-epiglottic fold was also found.

Fine needle aspiration of the parotid lesion showed atypical cells. Left tonsil biopsy was in favor of squamous cell carcinoma (SCC). Surgical treatment was decided. Left tonsil resection extended to the soft palate and the base of tongue was performed, associated with the excision of the suspected PA recurrence, left neck dissection and reconstruction using a radial forearm flap. Extemporaneous pathological examination during the procedure showed that the left parotid nodule and the tonsil lesion were both SCCs.

Final pathological results showed a moderately differentiated non-keratinizing exophytic squamous cell carcinoma infiltrating the tonsil with perineural and intravascular invasion. This SCC was not papillary because of the non-branching, thin aspect of the

☆ Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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Fig. 1. Pre-operative head and neck MRI imaging. Fat suppression T1-weighted image with contrast agent injection (gadolinium). (A) Tonsil squamous cell carcinoma and (B) squamous cell carcinoma of the parotid region, both occurring 20 years after radiotherapy (55 Gy) for the treatment of a recurring pleomorphic adenoma.

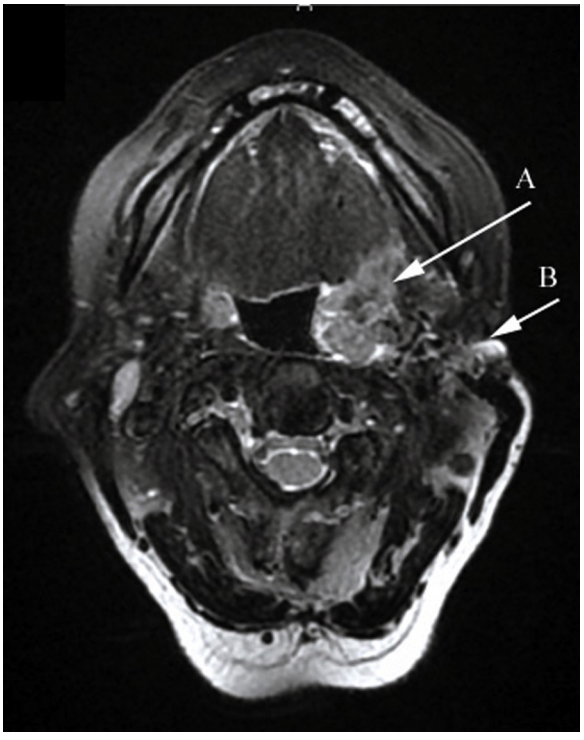

Fig. 2. Pre-operative head and neck MRI imaging. Fat saturation T2-weighted image. (A) Tonsil squamous cell carcinoma and (B) squamous cell carcinoma of the parotid region, both occurring 20 years after radiotherapy (55 Gy) for the treatment of a recurring pleomorphic adenoma.

papillary-like component. The parotid lesion had similar pathological features (Figs. 3 and 4). Immunohistochemical p16 stains were performed using a mouse monoclonal anti-$p16^{INK4a}$ antibody, clone E6H4 (CINtec Histology Kit, REF 9511, mtm laboratories, Heidelberg, Germany), according to the instructions of the manufacturer (Fig. 5). Two out of twenty-two lymph nodes from the left neck dissection contained SCC cells with capsular rupture. Adjuvant radiotherapy with volumetric modulated arc therapy (66 Gy/33 fractions) was decided because of a 30 years gap since the previous irradiation. The patient had no signs of recurrence 6 months after the end of radiotherapy.

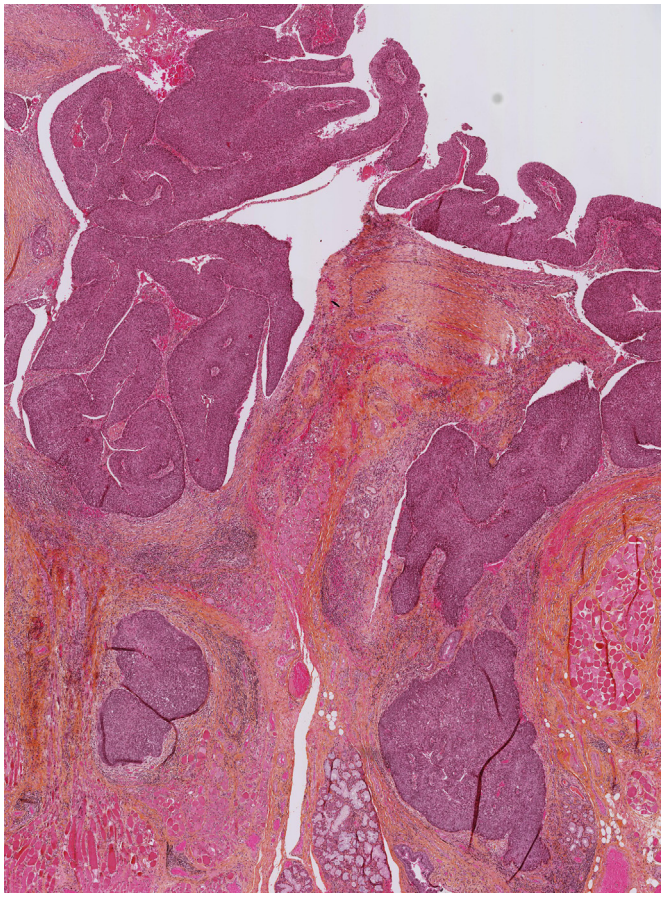

Fig. 3. Tonsil squamous cell carcinoma occurring 20 years after radiotherapy (55 Gy) for the treatment of a recurring pleomorphic adenoma. Poor differentiation, non-papillary architecture, perineural extension and intravascular invasion. Hematein–eosin–safran stain, magnification 1.2×.

2. Discussion

The use of radiotherapy is controversial in the management of PA recurrences because of the risk of radio-induced malignancies [1,2].

Criteria defining radio-induced tumors were first proposed for sarcoma [3] but are now applied to non-sarcoma malignant lesions. There criteria are all fulfilled in the case we report: (1) the tumor must arise in the irradiated area, (2) the tumor must be histologically distinct from the original benign tumor, (3) radiation must be above 2 Gy, (4) the tumor must not be present at the time of the irradiation, (5) there must be a prolonged latency period between radiation delivery and tumor development (at least 5 years), (6) metastatic tumor, tumor recurrence, genetic predisposition for