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## Case report

# Sebaceous variant mucoepidermoid carcinoma in a pediatric patient: Diagnostic pitfalls and management

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## ABSTRACT

Mucoepidermoid carcinoma (MEC) is the most common salivary gland malignancy of childhood. Early diagnosis is essential in determining prognosis and guiding treatment. This can be difficult as it has a spectrum of histological appearances. Here we present the case of a 14-year-old with a high-grade, sebaceous variant. Though initially diagnosed as benign sebaceous adenoma, fine needle aspiration performed for suspected recurrence revealed MEC. After parotidectomy confirmed this finding, a substantial defect remained and reconstruction using an anterolateral thigh free flap was performed. Furthermore, radiation was implemented and thought to have played a role in the successful outcome of this patient.

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## 1. Introduction

Salivary gland malignancies affect less than one child per million and comprise 8–10% of pediatric head and neck cancers [1]. The parotid gland is the most common location of pediatric salivary gland malignancies and mucoepidermoid carcinoma (MEC) is the most common subtype, accounting for 45–50% of all cases [1]. Many such individuals have a history of prior radiation exposure and present in the second decade of life [1]. Accurate histological diagnosis is critical in determining treatment and prognosis.

Classic MEC is defined by a mix of mucous, epidermoid (squamous), and intermediate cells [2]. Low grade variants have large areas of mucin-producing cells and cystic areas, often greater than 50%, while higher grades are more cellular and difficult to differentiate from normal gland [3]. In addition, the classification “high grade” encompasses many different histological variants including clear cell, oncocyctic, sclerosing, and sebaceous [3]. These tumors are notoriously difficult to identify due to histologic appearances that often overlap with benign salivary gland tumors. Once identified,

complete tumor resection with parotidectomy is the treatment of choice regardless of grade, often leaving children with substantial surgical defects, thus posing a reconstructive challenge. While low grade tumors may be managed with excision and reconstruction alone, higher grade tumors merit consideration of adjuvant radiation and neck dissection [3].

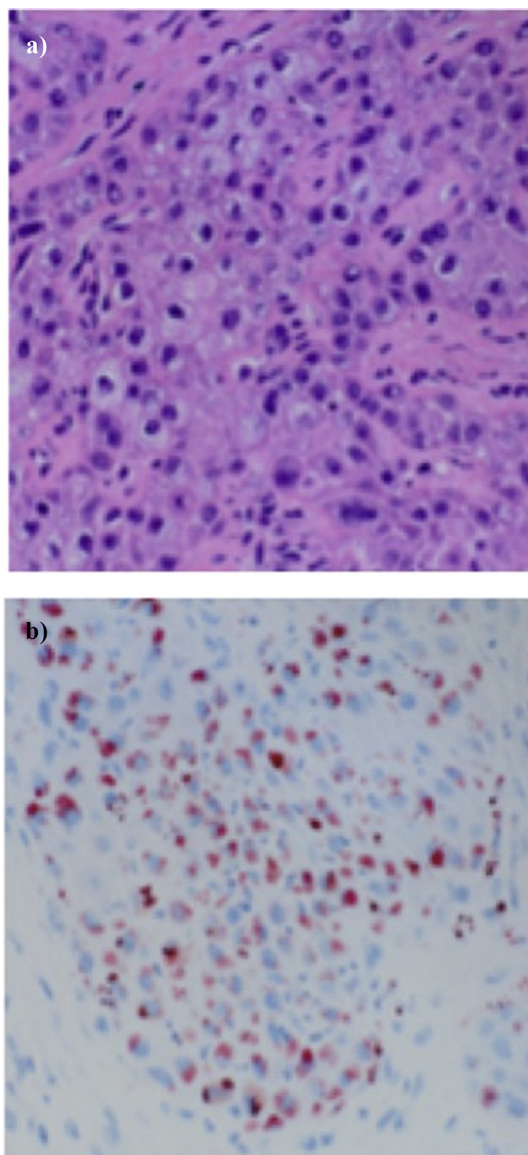
Here we report the case of a 14-year-old patient with sebaceous variant high-grade MEC of the parotid gland initially misdiagnosed as sebaceous adenoma treated with radical parotidectomy, selective neck dissection, and buried anterolateral thigh (ALT) free flap reconstruction followed by adjuvant radiotherapy.

## 2. Case report

A 14-year-old girl presented with a history of a left-sided parotid mass. She had previously undergone a total parotidectomy with facial nerve preservation for what was diagnosed as benign sebaceous adenoma on surgical pathology. Histologic evaluation disclosed a relatively well circumscribed neoplasm with a cell population characterized by microvesicular cytoplasm showing strong positivity for Oil Red-O, indicative of sebaceous differentiation (Fig. 1). Unlike most sebaceous adenomas, this tumor recurred quickly and aggressively over the next year. During a routine follow up appointment it was noted that the mass had recurred in the

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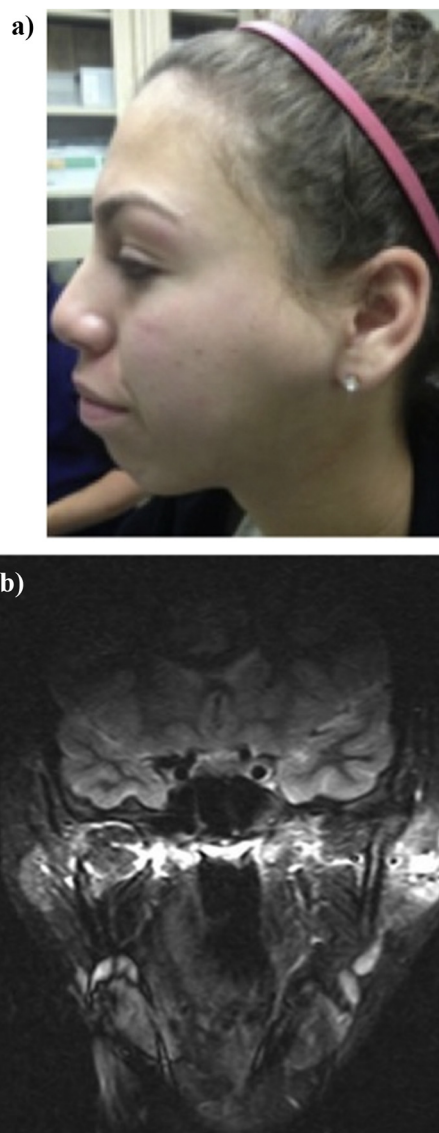
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**Fig. 1.** a) The tumor initially showed prominent sebaceous differentiation characterized by cells with abundant vacuolated cytoplasm consistent with benign sebaceous adenoma (H&E, 20 $\times$ ); b) Positive staining with oil red O confirms sebaceous differentiation (Oil Red O, 20 $\times$ ). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

preauricular region (Fig. 2) and the patient complained of occasional facial pain but no weakness. Repeat MRI imaging revealed a parotid mass extending from the angle of the mandible to the level of the zygoma in the preauricular space, measuring approximately 4.2  $\times$  4.2  $\times$  2.4 cm. A fine needle aspiration biopsy was consistent with low-grade MEC. The patient's case was discussed at a multidisciplinary tumor board conference and the consensus was to proceed with aggressive surgical management. A radical parotidectomy and mastoidectomy, neck dissection, and free flap reconstruction was planned. Given the size and location of the tumor, it was determined that sternocleidomastoid flap reconstruction was unlikely to adequately reconstruct the resultant defect and plastic surgery was consulted. Additionally, plans were made to harvest a nerve from the donor site to be used for cable nerve graft reconstruction given the high likelihood of facial nerve sacrifice.

A dense tumor with significant involvement of the deep lobe as



**Fig. 2.** Recurrent mass was noted in the preauricular region, found to be 4.2  $\times$  4.2  $\times$  2.4 cm extending from the angle of the mandible to the zygoma on MRI (a,b).

well as encasement of the facial nerve was encountered upon operation. Radical parotidectomy with facial nerve sacrifice was performed given the infiltrative nature of the tumor. Resected margins were sampled, as well as the main trunk of the facial nerve, and all intra-operative margins were negative. A selective supraomohyoid neck dissection of levels 1–3 was performed and all nodes returned negative for malignancy.

The radical parotidectomy defect was substantial, and a significant amount of soft tissue was needed for repair. A 7  $\times$  10 cm adipo-fascial free flap was harvested from the anterolateral thigh with blood supply from the descending branch of the lateral femoral circumflex artery. The donor site was extended and the lateral femoral cutaneous nerve of the thigh was harvested for cable grafting of the facial nerve. After completion of the parotidectomy and neck dissection, the temporal branch of the facial nerve was grafted and anastomoses were made at the common facial artery and external jugular vein. The adipo-fascial flap was then inset into the parotidectomy defect and the cheek skin flap was re-draped and closed primarily, burying the flap. The patient

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