CASE REPORTS

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High-Grade Mucoepidermoid Carcinoma with Sarcomatous Features of the Tongue in a Seven-Year-Old Boy: A Case Report

Arjay Franz Joseph A. Alberto, MD
Eduardo G. Dizon, Jr. MD
Ronaldo G. Soriano, MD
Miguel C. Aragon III, MD

Department of Otorhinolaryngology Head and Neck Surgery
Dr. Paulino J. Garcia Memorial Research and Medical Center

ABSTRACT

Objective: To report an unusually aggressive presentation of high-grade mucoepidermoid carcinoma (MEC) with sarcomatous features of the tongue base in a seven-year-old boy, characterized by early recurrence, rapid progression and poor response to treatment.

Methods:

Design: Case Report
Setting: Tertiary Government Training Hospital
Patient: One

Results: A seven-year-old boy presented with a left tongue base mass that rapidly enlarged with associated dysphagia and weight loss. Biopsy of the pedunculated mass attached to the left tongue base, abutting the anterolateral tongue, revealed high-grade MEC. Following left subtotal glossectomy with radical neck dissection, histopathology confirmed high-grade MEC with sarcomatous features with negative margins but positive lymph nodes. Post-operative recurrent masses were twice excised at three weeks (left tonsil) and another 12 days (oropharynx extending to neck). Despite tumor debulking and three radiotherapy sessions, the residual mass enlarged further and metastasized to the lungs and liver six weeks postoperatively. The patient's condition further deteriorated and he expired prior to starting chemotherapy.

Conclusion: High-grade MEC of the tongue base is exceedingly rare in children. This case exhibited sarcomatous features and demonstrated unusually aggressive behavior characterized by early recurrence, rapid progression, and poor response to treatment. Continued reporting of such rare presentations is vital to augment knowledge of pediatric MEC with sarcomatous features.

Keywords: *mucoepidermoid carcinoma (MEC); minor salivary gland; tongue; sarcomatous features*

Correspondence: Dr. Ronaldo G. Soriano
Department of Otorhinolaryngology
Head and Neck Surgery
Dr. Paulino J. Garcia Memorial Research and Medical Center
Mabini St., Cabanatuan City, Nueva Ecija 3100
Philippines
Phone: +63 998 551 3139
Email: ronsorianomd@gmail.com

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Mucoepidermoid carcinoma (MEC) is the most common malignant tumor of the salivary glands; however, its occurrence at the tongue base is rare, particularly among pediatric patients.[1,2] Complete surgical excision remains the standard of care with recurrence rates reported at less than 10% for minor salivary gland MEC in children.[3] A literature search using HERDIN Plus, the ASEAN Citation Index, MEDLINE (PubMed and PubMed Central), and Google Scholar with the keywords "mucoepidermoid carcinoma," "child," "base of the tongue," "minor salivary glands," and "sarcomatous features" revealed no published local studies on MEC of the tongue. Furthermore, while several reports have described MEC arising from minor salivary glands, none have documented cases exhibiting sarcomatous features.[2-9] This case adds to the limited body of literature by describing a rare histologic variant of MEC with sarcomatous features in a pediatric patient. We report an unusually aggressive case of high-grade MEC of the tongue base in a seven-year-old Filipino boy characterized by early recurrence, rapid progression, and poor response to treatment.

CASE REPORT

A seven-year-old boy observed an approximately 1 x 1 cm pinkish, dome-shaped mass over the left base of the tongue two weeks prior to consultation. There was no associated pain, bleeding, dysphagia, impaired tongue mobility, voice change, or neck swelling, and the mass did not resolve with antibiotics. The patient had no comorbidities, family history of malignancy, or history of smoking or radiation exposure.

On initial consultation, the mass had enlarged to an exophytic, pedunculated, non-friable 3 × 3 × 2 cm lesion arising from the left tongue base. *(Figure 1)* Its base was located at the posterior tongue, with the bulk abutting but not infiltrating the left anterolateral tongue and floor of the mouth. The mass was non-tender, non-pulsatile, non-bleeding, and did not restrict tongue movement. There was also a firm, movable, non-tender left infra-auricular level II lymph node measuring 2 × 1 × 1 cm.

While awaiting biopsy, the boy developed an acute episode of dyspnea with oxygen desaturation, likely secondary to progressive airway obstruction caused by the enlarging tongue base mass. Endotracheal intubation was unsuccessful due to upper airway distortion and an emergency tracheostomy was performed. Contrast-enhanced CT scans revealed a large, heterogeneously enhancing mass arising from the left posterior tongue, measuring 4.4 × 3.7 × 4.8 cm. As shown in *Figure 2A & 2B,* the lesion extended posteromedially, causing obliteration of the oropharyngeal lumen and superior extension into the nasopharynx. Multiple enlarged, enhancing lymph nodes were also identified in the left submandibular and upper cervical regions, the largest measuring 2.2 cm in diameter. No bony invasion was noted.

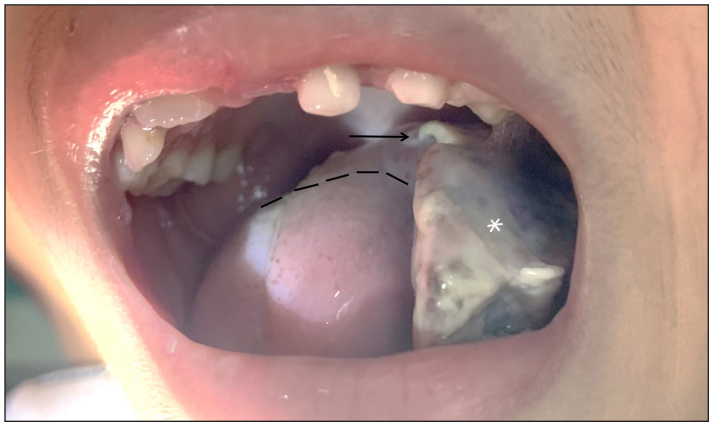

Figure 1. Preoperative intraoral photograph showing an exophytic and pedunculated left tongue base mass (asterisk). The black arrow shows the base of the mass at the foramen cecum. The black dashed line delineates oral tongue from the tongue base.

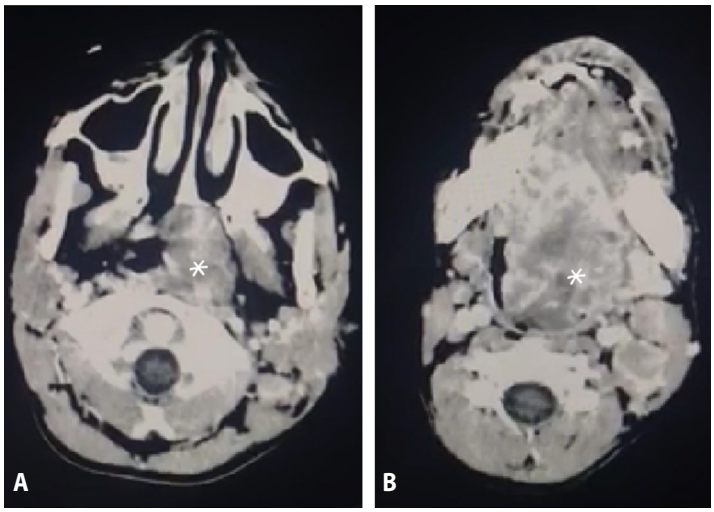

Figure 2. Contrast-enhanced CT images, axial views at the level of the: **A.** nasopharynx; and **B.** oropharynx; revealing a lobulated, heterogeneously enhancing soft tissue mass (asterisks) centered at the left tongue base, extending across the midline with infiltration of adjacent musculature and inferior extension toward the floor of the mouth, causing significant airway narrowing.

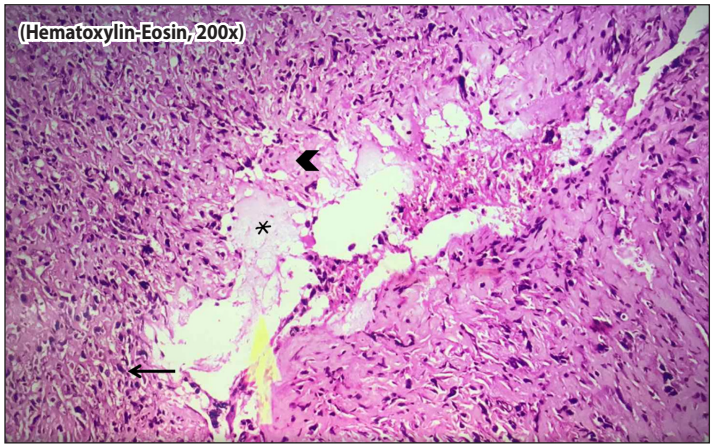

Figure 3. Histopathologic section of the tumor (Hematoxylin-Eosin stain, 200x) demonstrating the characteristic cellular components of mucoepidermoid carcinoma: intermediate cells (arrowhead), mucous cells (asterisk), and squamoid cells (arrow)

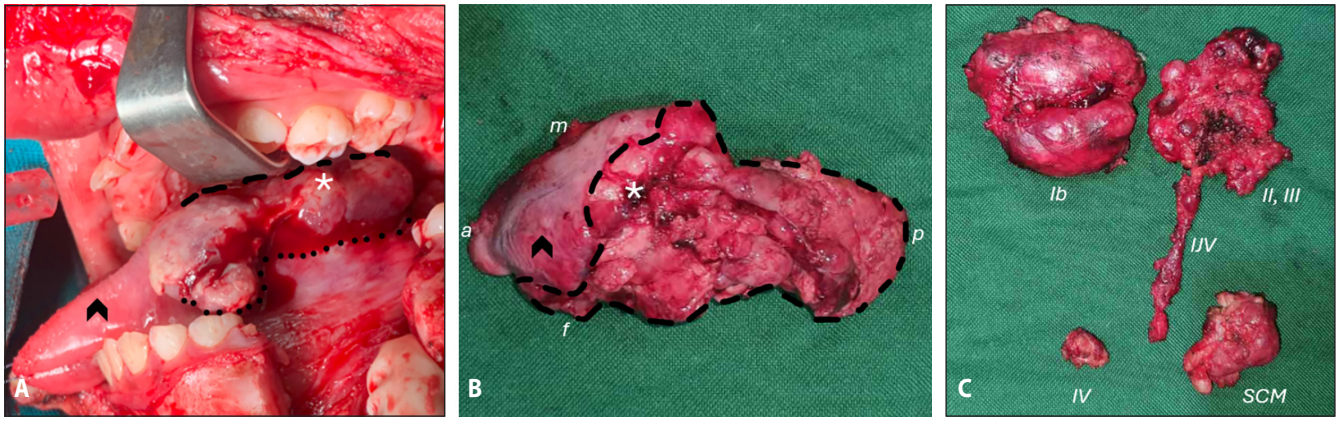

Figure 4. Intraoperative photos showing: **A.** lobulated, firm, irregular mass (asterisk) arising from the left tongue base, extending anteriorly toward the oral tongue (arrowhead), inferiorly into the floor of the mouth (dotted line), and medially near the midline (dashed line), with infiltration into adjacent musculature; **B.** Gross specimen, resection included the left anterior tongue (arrowhead) and tongue base (asterisk). The bulk of the mass is encircled (dashed line), medial aspect, (m); floor, (f); anterior, (a); posterior, (p); and **C.** Gross specimens of enlarged cervical lymph nodes excised during radical neck dissection (levels 1b, II, III, IV); internal jugular vein (IJV); sternocleidomastoid muscle (SCM)

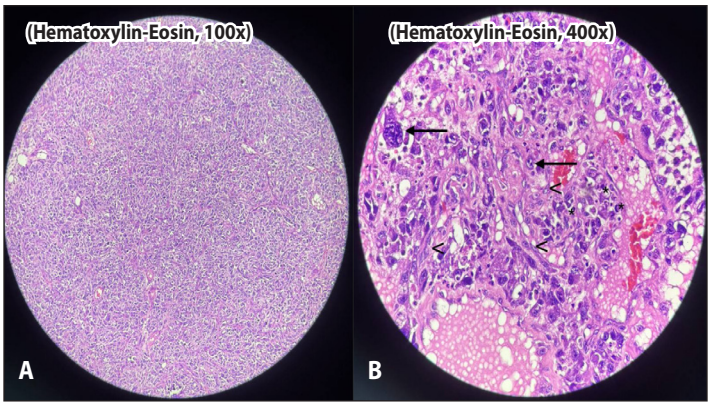

Figure 5. Histopathologic sections of the tumor showing sarcomatous features (Hematoxylin and Eosin stain): **A.** Sheets and haphazard fascicles of atypical cells on low-power objective (LPO, 100×); and **B.** Bizarre nuclei (arrows), elongated nuclei (arrowheads), and mitotic figures (asterisks) on high-power objective (HPO, 400×)

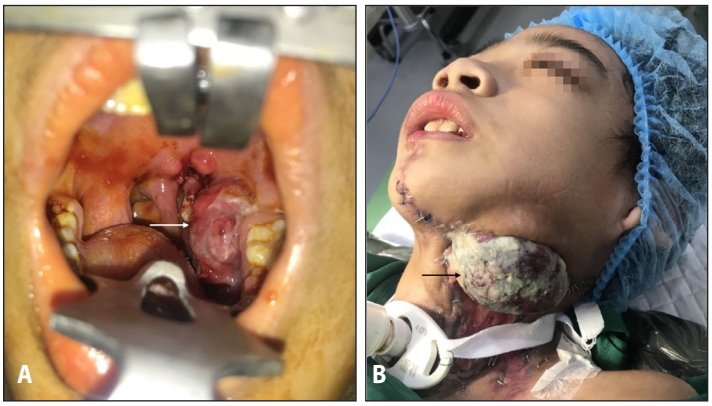

Figure 6. Postoperative images at three weeks: **A.** intraoral view showing a lobulated mass at the left tongue base (white arrow); and **B.** wound breakdown with orocutaneous fistula and exposed necrotic tissue along the left neck incision site (black arrow). Photos published with permission.

Histopathologic examination of a biopsy specimen demonstrated nuclear atypia, numerous mitotic figures and perineural invasion. *Figure 3* illustrates the presence of three cell types—intermediate, mucous, and squamoid—findings characteristic of high-grade MEC. Immunohistochemical studies to aid further characterization were not available at our institution at the time.

A left subtotal glossectomy via mandibulotomy and left radical neck dissection was performed. Intraoperatively, a lobulated, firm, grayish-pink mass with an irregular, ulcerated surface was identified at the left tongue base. *(Figure 4A)* The lesion measured 5 × 4 × 3 cm and extended anteriorly to the posterior half of the oral tongue (1.3 cm from the tip), inferiorly into the floor of the mouth, and medially toward the midline. *(Figure 4B)* It was inseparable from the intrinsic musculature and infiltrated the extrinsic tongue muscles, with induration palpable to the oral tongue tip, floor of the mouth, and midline. Enlarged lymph nodes were present at levels Ib–IV, with close involvement of the spinal accessory nerve. *(Figure 4C)*

Reconstruction was performed using primary closure of the residual tongue tissue with the buccal mucosa and the remaining tissues of the floor of the mouth to restore tongue bulk and preserve tongue mobility as much as possible. The surgical bed was closed in watertight manner to prevent orocutaneous fistula while ensuring adequate tension-free repair to optimize healing. Tracheostomy was maintained to secure the airway, minimize incidence of aspiration and facilitate pulmonary toilet.

Histopathology revealed negative surgical margins and demonstrated features of MEC, but including sheets and haphazard fascicles of atypical cells (spindle-cell morphology) with elongated to bizarre nuclei and fibrillar cytoplasm. These are unusual for

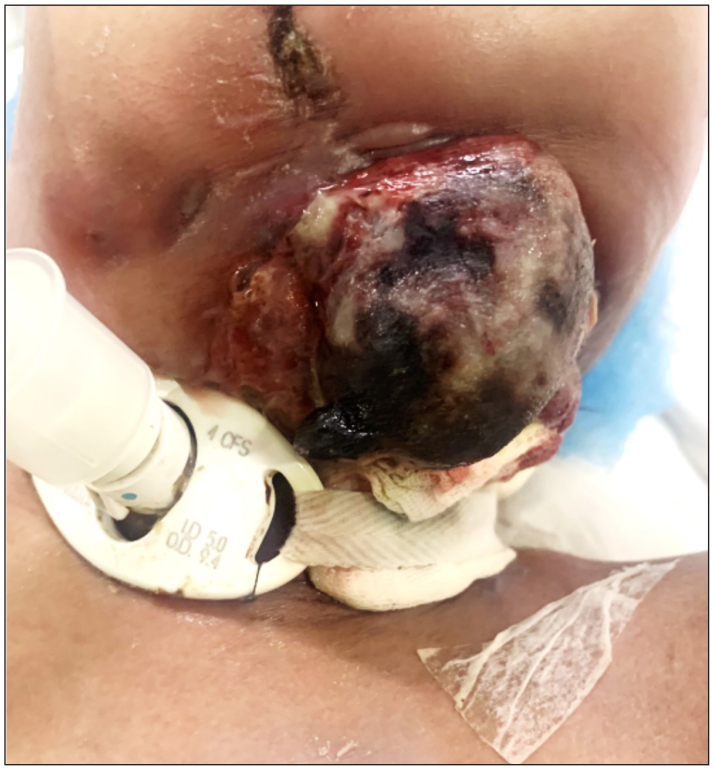

Figure 7. Five-week postoperative image of oropharyngeal mass extending into the neck through the orocutaneous fistula

MEC and more indicative of sarcomatous differentiation. *(Figure 5)* Immunohistochemical staining for cytokeratin and actin was negative, while desmin demonstrated only focal positivity. Following review and consensus by multiple pathologists, the case was diagnosed as high-grade MEC with sarcomatous features. Metastatic tumor was identified in 19 of 22 examined lymph nodes.

Within the first postoperative week, wound dehiscence with orocutaneous fistula formation was noted. Conservative management was initiated, including meticulous wound care, pressure dressings, nasogastric tube feeding to reduce salivary contamination, and broad-spectrum antibiotics. Despite these measures, the dehiscence progressed. Three weeks postoperatively, a mass was identified along the left lateral pharyngeal wall, occupying the tonsillar region and extending inferiorly through the dehiscence in the floor of the mouth and neck incision. *(Figure 6A)* The lesion, measuring 6 × 4 × 3 cm, was externally visible through the wound defect. *(Figure 6B)* Surgical excision was performed, and histopathology once again confirmed high-grade MEC.

Twelve days after the second surgery, an oropharyngeal mass was again noted. *(Figure 7)* CT scans demonstrated an orohypopharyngeal lesion measuring 3.8 × 2.4 × 2.0 cm, causing complete obliteration of the airway, along with bilateral upper cervical lymphadenopathies. The patient underwent tumor debulking and initiated radiotherapy, completing three of the planned 34 fractions. Despite these interventions, the mass continued to enlarge, accompanied by clinical deterioration. Repeat CT imaging revealed a necrotic lobulated soft tissue density in the left lateral neck measuring 6.9 × 3.7 × 3.0 cm, suggestive of tumor extension or confluent necrotic lymph nodes. Further chest imaging showed bilateral pleural effusion with lower lobe atelectasis, multiple pulmonary parenchymal nodules, mediastinal lymphadenopathy, and multiple hypodense hepatic lesions, all consistent with metastases.

The patient's course was further complicated by facial edema, anemia, thrombocytopenia, hypoalbuminemia, hyperkalemia, ascites, pleural effusion, and sepsis. Despite maximal supportive and medical management, the patient ultimately suffered cardiopulmonary arrest.

DISCUSSION

Salivary gland (SG) tumors represent only 3–5% of all head and neck malignancies, with a global incidence estimated at 0.4–13.5 per 100,000 individuals.[1] Among these, mucoepidermoid carcinoma (MEC) is the most common, accounting for 10–15% of cases and typically arising in the parotid and minor salivary glands.[2] Within the minor salivary and submandibular glands, however, MEC ranks second in frequency to adenoid cystic carcinoma (AdCC).[1] The hard palate is the most common site of involvement among the minor salivary glands, followed by the buccal mucosa, tongue, lips, and floor of the mouth.[6] MEC arising from the base of the tongue is particularly uncommon with limited data on management and long-term outcomes.[7,8]

An analysis of the SEER database (2004–2016) identified only 145 such cases, with a female predominance (56.5%) and a mean age at diagnosis of 58.8 years.[7] In children and adolescents, MEC also demonstrates a female predilection and often presents as a fluctuant submucosal mass with bluish discoloration, even mistaken as mucoceles.[3] Our patient, however, developed an exophytic lesion that enlarged rapidly, further highlighting the atypical aggressiveness of this tumor. It typically presents as a low- to intermediate-grade tumor, generally associated with favorable outcomes following complete excision.[9,10] In contrast, our case describes a rare high-grade MEC with sarcomatous features which exhibited unusually aggressive behavior.

Histopathologically, MEC demonstrates varying proportions of mucous, intermediate, and squamoid cells. High-grade tumors are characterized by marked pleomorphism, increased mitotic activity, and perineural invasion.[3] Review of the resected specimen in our case

revealed high-grade MEC with sarcomatous features—a particularly rare histologic variant associated with aggressive behavior and poor outcomes.[4,5] This phenomenon may explain the unusually rapid recurrence and widespread metastasis observed in our patient despite adequate initial surgical management.

Standard management for high-grade MEC consists of wide surgical excision with negative margins and appropriate neck dissection. In children, this approach must be balanced with the need to minimize long-term morbidity.[3,11] Despite achieving negative surgical margins in our patient, tumor persistence was observed within three weeks, initially in the tonsillar fossa and subsequently in the oropharyngeal and cervical regions. This rapid recurrence underscores the aggressive potential of high-grade MEC with sarcomatous features. Surgical excision is followed by adjuvant radiotherapy specifically for tumors with nodal metastasis or perieural invasion.[1,6] Our patient began intensity-modulated radiotherapy (IMRT) and received only three fractions of the prescribed 5100 cGy due to rapid disease progression and clinical deterioration. Debulking was performed at this stage, not with curative intent, but to relieve airway compromise and facilitate tracheostomy care, as the enlarging tumor compromised tracheal tube function. This highlights the role of surgery as palliation in advanced recurrent disease, particularly for airway management.

Chemotherapy has not been established as standard treatment for MEC but may be considered in advanced or metastatic cases, especially in rare aggressive variants.[3,4] Previous reports of MEC with carcinosarcomatous features describe patients who remained recurrence-free for at least six months after surgery and adjuvant radiotherapy only.[4,5] In our case, chemotherapy was initially deferred given the lack of robust evidence for its benefit; however, as the disease progressed and in light of the sarcomatous component, chemotherapy was contemplated. Unfortunately, the patient's rapid decline precluded its initiation.

This case represents a rare and highly aggressive tumor overwhelming all available management strategies and ultimately resulting in the patient's demise. Achieving adequate surgical margins while minimizing post-operative morbidity posed significant challenges, given potential impacts on swallowing, aspiration, nutrition, and speech. In hindsight, earlier initiation of chemotherapy may have altered the disease course, particularly given the sarcomatous features and multiple recurrences; however, further evidence was required to support this approach. Despite early recognition and maximal multidisciplinary care, our patient succumed to this unusually aggressive tumor. Continued reporting of such rare presentations that deviate from established patterns may address gaps in the knowledge of pediatric MEC with sarcomatous features.

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