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Intraoral myoepithelial carcinoma in a young man with intellectual disability: A highly rare and challenging case

Myoepithelial carcinoma (MC), which take only one percent of all salivary gland neoplasm and most commonly involve parotid gland, is a very rare cancer. The latest review paper to date indicates that there are only 109 cases of MC raised from minor salivary gland worldwide.[1] On the other hand, due to difficulties in communication and compliance, the treatment for oral cancer in people with intellectual disabilities (PWIDs) is seldom discussed and reported. Treating PWIDs with myoepithelial carcinoma becomes a dilemma when there is no established treatment protocol for MC and a lack of evidence regarding oral cancer treatment in this population.

A 22 years-old young man, who had medical history of autism, was brought to Department of Oral-Maxillofacial Surgery, Shuang-Ho Hospital for an oral mass that had been noted for several months. Physical examination revealed no facial asymmetry nor palpable neck mass. Intraorally a soft bulging mass was observed in left retromolar area, measuring approximately 3 × 3 cm with mild induration. Dental panoramic film showed no osteolytic changes. His mother denied any personal habit of cigarette or betel nuts using. The result of incisional biopsy revealed of myoepithelial carcinoma. The clinical stage was cT2N0M0, stage II after finishing the tumor study, which including a head and neck MRI, PET-CT scan, chest PA and abdominal sonogram. Standard surgical treatment plan would typically involve tumor wide excision including marginal mandibulectomy and a free flap reconstruction. However, considering the patient's compliance for post operative care, a more conservative surgical approach was chosen, including wide excision of the soft tissue tumor with peripheral osteotomy, and repair the defect with an artificial skin. Final histopathological report is the myoepithelial carcinoma ex pleomorphic adenoma, pT3N0M0, stage III. Classical histopathological finding such as a solid plasmacytoid myoepithelial cell tumor infiltrated in the tubular structure with background of mucin like tissue, with some plasmacytoid pattern was also founded under high-power magnification. Although there is no treatment protocol for myoepithelial carcinoma, adjuvant radiotherapy with 66 Gy in 33 friction was done based on the NCCN guidelines because of the risk factors of tumor size and close surgical margin (less than 1 mm). Monthly follow-up was scheduled. So far, there is no evidence of local or regional recurrence with acceptable oral function (Maximum mouth opening: 20 mm). All the photos of the pre-operative record, surgical procedures, pathological findings, and post-operative results are showed in Fig. 1.

To our knowledge, only low double-digit number of papers reported malignant oral tumors in PWIDs, but less than 5 cases accepted the standard surgical treatment due to almost impossible post-operative care.[2–4] Furthermore, no case(s) of intraoral myoepithelial carcinoma in PWIDs were reported in the literature to-date. Prognosis is related to tumor size, mitotic rate, mix tumor type (CA ex-PA) and resection margin but nearly no role of adjuvant radiation therapy.[1] Fortunately, as the recommendation from Jeng et al.,[5] our case become cooperative after using tell, show,

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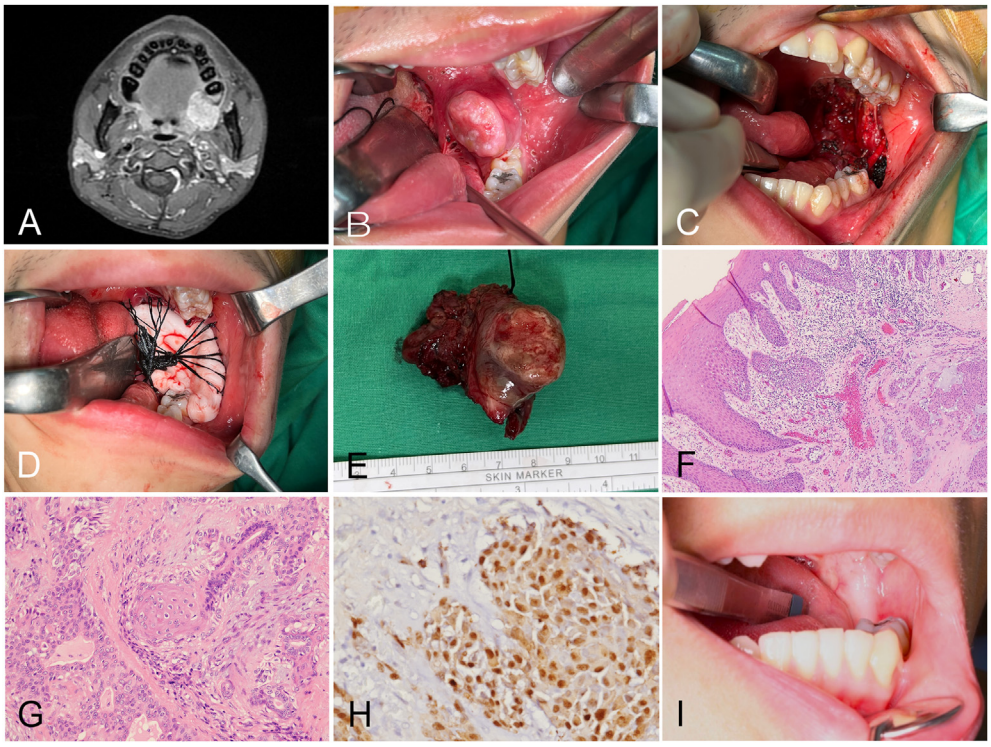

Figure 1 Photographs of our patient with myoepithelial carcinoma ex pleomorphic adenoma depicting surgical procedures, histopathological findings, and immunostaining results. (A) An exophytic enhanced mass was detected in the left retromolar trigon in MRI T1-weighted images with contrast. (B) Protruding mass with intact mucosa. (C) Defect after wide excision of the tumor. (D) Repair with Integra® Dermal Regeneration Template. (Integra Lifesciences Corporation, Plainsboro, NJ, USA). (E) The main specimen was about 5 × 5 cm in size. (F and G) Medium-power and high-power photomicrographs predominantly showing plasmacytoid myoepithelial carcinoma cells and the infiltrative border. Focal ductal or tubular structures and squamous differentiation are noted, compatible with a preexisting pleomorphic adenoma. (hematoxylin and eosin stain; original magnification; F, × 40; G, × 400). (H) A high-power photomicrograph revealing that the cells were S100-positive (immunohistochemical stain; original magnification; × 400). (I) Surgical area after 30 months of postoperative follow-up.

and do approach and provide the possibility of wound care and regular follow-up.

Declaration of competing interest

The authors have no conflicts of interest relevant to this article.

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