

Short Case Report

Multiple synchronous rhabdomyomas in the tongue and floor of the mouth: case report

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Abstract – Observation: A 61-year-old male patient presented with tongue and base floor of the mouth firm swelling with intact overlying mucosa with ballottement by bimanual palpation. Histopathological examination and immunodiagnosis revealed this mass as adult rhabdomyoma. **Comments:** Any tongue base or floor of the mouth mass with intact overlying mucosa should be approached by magnetic resonance imaging with great concern of rhabdomyoma.

Observation

A 61-year-old male patient presented with a slowly growing intraoral swelling within several months. He complained of mild discomfort, globus pharynges, intermittent chewing problems and articulation problems. On clinical examination, there was a non-tender, firm mass in left side of the tongue – floor of the mouth felt in the submandibular triangle in the neck via bi-digital palpation, but there was no mucosal ulceration, no cervical lymphadenopathy and no cranial nerve affection (Fig. 1).

MRI (magnetic resonance imaging) was requested, which revealed multiple well-defined soft tissue masses in the tongue and floor of the mouth. This mass involved left side of the tongue extending medially to right side, posteriorly to tongue base and oropharynx, inferolaterally to floor of the mouth; iso-intense to intermediate signal on T1-weighted image (Fig. 2a), high intensity on T2-weighted image (Fig. 2b,c) and STIR (short tau inversion recovery) (Fig. 2d) with mild homogeneous enhancement on T1-weighted with contrast (Fig. 2e). FNAC (fine needle aspiration cytology) was requested but was not conclusive, showing chronic inflammatory lesion.

Surgical management was planned in form with elective tracheotomy; then, surgical excision of these multiple masses through combined intraoral approach and transcervical approach was performed (Fig. 1). Complete tumor excision was obtained in form of large mass (7.2 × 4 × 1.8 cm) and

multiple small satellite masses (variable from 0.3 to 2.00 cm in axial diameter). All these masses were capsulated, lobulated, firm and had smooth surface.

Overlying mucosa, left submandibular gland, left hypoglossal and lingual nerves were preserved during the surgery. Primary closure was done, and a drain was inserted for 48 h.

Intraoperative preliminary biopsy and definitive biopsy with immunohistochemistry-aided diagnosis (+ve Desmin/ –ve S100) confirmed adult rhabdomyoma.

Weaning from tracheotomy tube started after the third day postoperatively and the patient was discharged after 5 days with monthly follow-up. No complication in early follow-up period (6 months), no nerve defect, no oral fistula and no residual or recurrent masses were reported.

Comments

Rhabdomyomas were uncommon benign tumors of striated muscle, slowly growing lesions varying in size from a few millimeters to 15 cm [1], commonly affecting muscles of pharyngeal arches in the head and neck region, especially the oral cavity (base of the tongue and floor of mouth), pharynx and larynx [2].

Adult extra-cardiac rhabdomyomas were mostly found in elderly male patients about 55–60 years with male:female ratio (ratio 4: 1), without racial predilection [3].

Multifocal rhabdomyomas may occur simultaneously, or these lesions develop several years apart, or may be due to incomplete removal of first mass (residual) with

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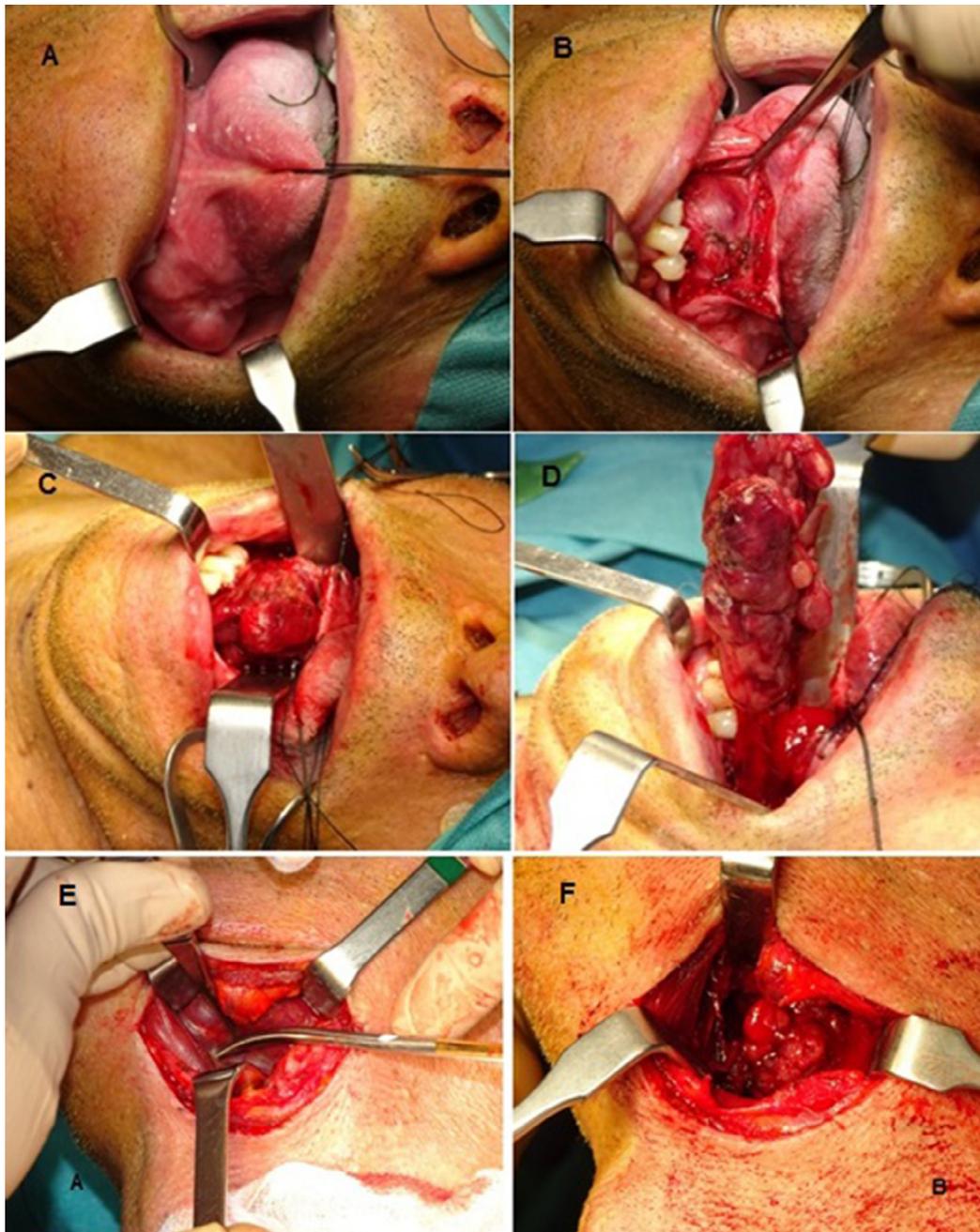


Fig. 1. (A) left tongue base swelling with intact mucosal covering; (B) intraoral approach for mass excision; (C) dissection around capsule of mass; (D) complete excision of huge tongue mass; (E) transcervical approach; (F) dissection till tongue base to expose the mass to facilitate its delivery.

development of a new nidus of rhabdomyoma (recurrence) [3]. Recurrence occurred in 16% of cases reported in the literature, but incomplete removal caused most recurrences. Recurrences have been reported even after 30 years [4].

The treatment of choice for rhabdomyoma was the conservative excision even with a high possibility for recurrence [5]. Though mandibulotomy may be needed

for complete excision of even a benign tumor, complete excision is obtained with combined transoral-transcervical approach.

Tumors rather than squamous cell carcinoma tumors within floor of the mouth were characterized by intact oral mucosa; they showed a wide range of differential diagnosis as minor salivary gland tumor, lymphomas, neurogenic tumors

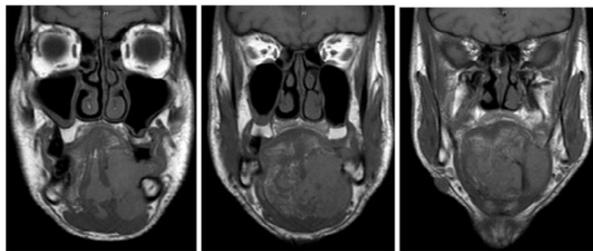


fig 2 a

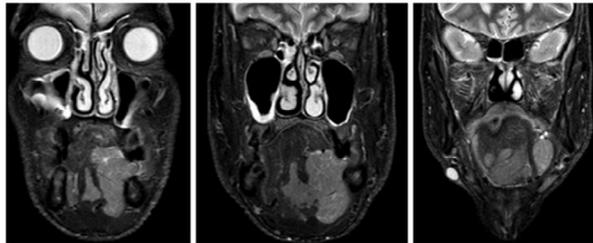


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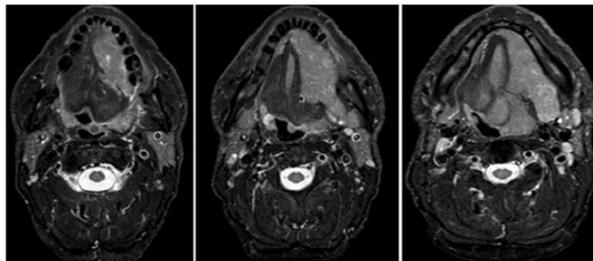


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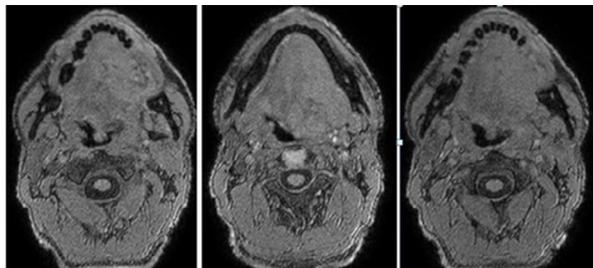


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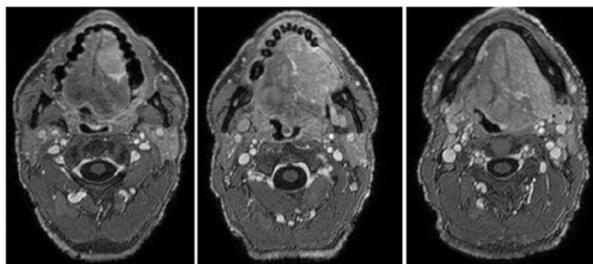


fig 2 e

Fig. 2. MRI findings of rhabdomyoma; (a) coronal T1WI iso-intense signal; (b) coronal T2WI high signal; (c) axial T2WI high signal; (d) axial STIR (short tau inversion recovery) fat suppression MRI high signal; (e) T1 with contrast homogenous enhancement.

and myogenic tumors [5]. So, preoperative cytology and MRI, also histopathological examination with immunohistochemistry, were recommended for accurate diagnosis and management.

The current case is another reported extra-cardiac adult rhabdomyoma in the head and neck region besides about 100 reported cases in the literature [5].

Up to our knowledge, it is the first reported case showing satellite appearance (huge mass $7.2 \times 4 \times 1.8 \text{ cm}^3$ surrounded by multiple small rounded masses variable 0.3–2.00 cm) or in other words, multiple synchronous rhabdomyoma masses in the tongue and floor of the mouth. Moreover, this case is one of rare well-documented cases report regarding MRI characteristics.

Conflicts of interests: The authors declare that they have no conflicts of interest in relation to this article.

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