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Alveolar soft part sarcoma of tongue in a 3-year-old Taiwanese



KEYWORDS Alveolar soft part sarcoma; Tongue; Taiwan

Alveolar soft-part sarcoma (ASPS), which was firstly described by Christopherson et al., in 1952,¹ is a rare soft-tissue sarcoma accounting for approximately 0.5-1.0% of all soft tissue sarcomas and less than 0.1% of head and neck sarcomas.² ASPS occurs predominantly in adolescents and young adult from the age of 15-35, and is very rare in age less than 5 years.^{2,3} Most ASPSs manifest as slow growing, painless intramuscular masses. Head and neck region is the most common affected site for cases of infants and children with 41% at the orbit and 25% in the oral cavity.²

We herein reported a case of ASPS in a 3-year-old boy who complained his tongue stabbed by a fishbone. His parents noted a swelling over his right posterior dorsal tongue for 3 months. Intraoral examination revealed a painless, pink-colored, round-shaped, smooth-surfaced submucosal swelling, about rubbery measured 1.2×0.8 cm in diameter, over the right posterior dorsal tongue (Fig. 1A). Neither tenderness nor cervical lymph node enlargement was noted. Tentative diagnosis was a foreign body granuloma. This lesion was subsequently excised under sedation (Fig. 1B, C). Histopathological examination showed sheets and lobules of tumor cells delineated by fibrous septa in the lamina propria of the mucosa (Fig. 1D). The tumor cells revealed eosinophilic, granular cytoplasm, and central or eccentric nuclei with prominent nucleoli (Fig. 1E). Some dilated veins were present at periphery of the mass (Fig. 1F). Immunohistochemically, the tumor cells were positive for TFE3 (Fig. 1G) and myogenin; focally positive for CD68; negative for S100, desmin, and HMB-45. PAS and PASD stains highlighted the intracytoplasmic crystals (Fig. 1H). Histopathological diagnosis of ASPS was rendered. Due to the involvement of tumor cells of the section margin, second surgery for clear surgical margin was arranged. General survey of brain magnetic resonance imaging, chest computed tomography, general bone scan, and positron emission tomography scan revealed no evidence of distant metastatic lesion. The residual lesion was widely excised with 1.5–2.0 cm clinical safe margin under general anesthesia (Fig. 1I), and the surgical defect was repaired by primary closure (Fig. 1J). The histopathological diagnosis was again ASPS with microscopic safe margin of 0.8 cm. The patient revealed recovered swallowing and phonetic function at one-month postoperative follow-up.

Reviewing English literature, to the best of our knowledge, our case is the first oral ASPS in a Taiwanese of age less than 5-year-old. The survival rates of ASPS in adults and children were estimated to be 77% at 2 years, 60% at 5 years, 38% at 10 years, and 15% at 20 years.⁴ Thus, considering the high rates of late recurrence and metastasis, surgical resection with 1.5-2.0 cm surgical margins and long-term follow-up are adopted in our patient. On the other hand, better prognosis has been claimed for cases in children and cases in oral cavity.^{2,3,5} So, potential favorable clinical outcome could be expected for the current case, which also perhaps due to the relative smaller tumor size and the early detection of oral ASPS.

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Figure 1 Clinical and microscopic pictures of the current case of alveolar soft part sarcoma. (A) A pink-colored, round-shaped, submucosal rubbery swelling with smooth surface over the right posterior dorsal tongue. (B, C) An excisional biopsy with primary closure under sedation was performed. (D) The tumor cells were arranged in sheets, and the lobules were delineated by fibrous septa (hematoxylin and eosin stain, H&E; magnification, $40\times$). (E) The tumor cells revealed eosinophilic, granular cytoplasm, and showed central or eccentric nuclei with prominent nucleoli (H&E; magnification, $400\times$). (F) Dilated veins present at periphery of mass, with partial involvement by tumor (H&E; magnification, $100\times$). (G) The tumor cells were diffusely positive for TFE3 (magnification, $200\times$). (H) PAS stain highlighted the intracytoplasmic crystals of the tumor cells (magnification, $200\times$). (I, J) The residual lesion was widely excised by second surgery under general anesthesia with the surgical defect repaired by primary closure.

Conflicts of interest

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

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.jds.2019.03.004.

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