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Sialadenoma papilliferum-like intraductal papillary tumor of the mandible with BRAF V600E mutation: A rare case report

Sialadenoma papilliferum (SP) is a benign salivary gland tumor with both exophytic and inward papillary proliferation of mucosal and salivary ductal epithelium.[1] Sialadenoma papilliferum-like intraductal papillary tumor (SP-IPT) refers to a neoplasm characterized by the presence of only endophytic ductal proliferation with BRAF mutation.[1] The entity of such tumor has not yet been clearly established, and their nomenclature varies depending on the reporting author, such as tubulopapillary hidradenoma-like tumor of the mandible, or sialadenopapillary ductal tumor.[2,3] Including four cases previously reported by our hospital, the number of reported cases of this entity in the literature is very limited.[2–5] Here, we presented an additional case of large sized SP-IPT with BRAF mutation occurring in the mandible.

A 68-year-old male patient presented with swelling and pain in the left mandible, which had started a week ago. On intraoral examination, a slightly smooth-surfaced swelling was observed in the retromolar region of the left mandible (Fig. 1A). The panoramic radiograph revealed a radiolucent lesion with a scalloped border in the retromolar region of the left mandible (Fig. 1B). CT imaging revealed a low-attenuated osteolytic lesion with destruction of the buccal and lingual cortical bone as well as the mandibular canal (Fig. 1C and D). Excision was performed. On the low-power view, multiple ductal structures forming endophytic papillary proliferation or simple cystic structures were observed, while exophytic squamous proliferation was not identified (Fig. 1E). The ductal components exhibited various sizes, with some cystic structures being notably large (Fig. 1F). On high-power view, abluminal layer of the ductal components consisted of flat to cuboidal cells, while the luminal layer was composed of cuboidal to columnar cells. Oncocytic metaplasia and apocrine snouting of the luminal layer were also observed (Fig. 1G and H). No cytologic atypia or mitotic activity was identified. Immunohistochemical analysis revealed positivity for CK7, VE1 and S100 in both abluminal and luminal cells (Fig. 1I and J). p63, p40, and SMA were selectively positive in abluminal cells (Fig. 1K). Sanger sequencing analysis of the BRAF (exon 15), PIK3CA (exon 9), and HRAS (exon 3) gene revealed that only the BRAF p.V600E mutation was detected (Fig. 1L).

Three months have passed since the surgery, the patient's pain has disappeared, and no specific findings such as recurrence have been observed. Recently, some researchers have proposed unifying SP, SP-IPT, and other tumors with similar histological and molecular features into a single entity named sialadenopapillary ductal tumor.[3] However, further analysis of additional cases will be needed to reach a consensus. When it occurs in the mandible, it is reported to have a relatively larger size.[3,4] Our case also presented with a large lesion measuring approximately 3.5 cm in diameter. Additionally, most patients have been reported to be elderly males, consistent with the findings in our case.[2–5] The present case highlights that SP-IPT is a rare tumor that predominantly arises in the posterior mandible, occurs more commonly in elderly males, and exhibits locally aggressive growth, emphasizing the importance of careful diagnosis.

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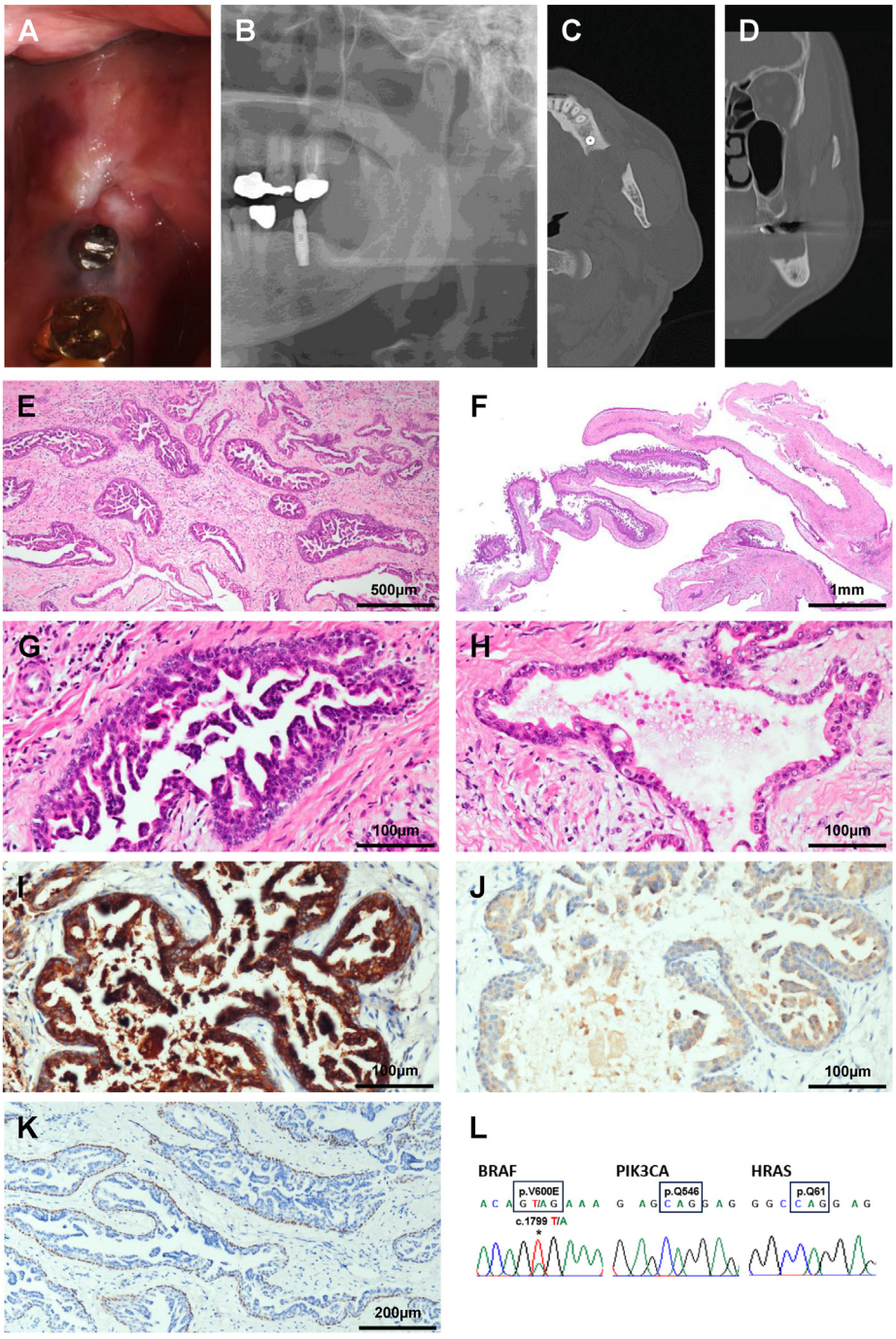

Figure 1 Clinical, radiological, histopathological, and immunohistochemical (IHC) features of sialadenoma papilliferum-like intraductal papillary tumor. (A) Clinical photograph shows a slightly smooth-surfaced swelling in the retromolar area of the left mandible. (B) Panoramic radiograph shows a radiolucent lesion with a scalloped border in the left posterior mandible. (C and D) CT images reveal a low-attenuated lesion with destruction of the buccal and lingual cortical bone (C) and the encroachment of mandibular canal (white arrow) (D). (E) The lesion shows multiple ductal structures forming endophytic papillary proliferation or simple cystic lining. (F) Large cystic structures are also observed. (G and H) A high-power view shows intraluminal papillary proliferation with oncocytic metaplasia (G) and apocrine snouting (H). (I, J and K) IHC results show that diffuse positivity for CK7 (I) and VE1 (J) in both luminal and abluminal cells, while p63 (K) was positive only in abluminal cells. (L) Sanger sequencing results show the presence of a BRAF p.V600E (c.1799T>A) missense mutation, while no mutations were detected in PIK3CA or HRAS.

Declaration of competing interest

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

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None.

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