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CASE REPORT

A case of odontogenic keratocyst in the buccal space: characterization by multimodality imaging including computed tomography, diffusion-weighted magnetic resonance imaging, and ultrasonography

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Abstract

Odontogenic keratocyst (OKC) is a relatively common non-inflammatory jaw lesion. OKC is known to occur most often in the mandibular angle and mandibular ramus, but rarely outside the bone. In this report, we describe characteristic multimodality imaging of OKC in the buccal space, especially diffusion-weighted MR imaging (DWI) with apparent diffusion coefficient (ADC) mapping, extra-oral and intra-oral ultrasonography. On clinical examination, an approximately 20 mm in diameter mass with elastic hardness was found the left side of the buccal area. Contrast-enhanced CT showed areas of internal non-contrast lesions in the left buccal space. On T1-weighted image, the mass showed multilocular high signal intensity, and homogeneous internal. T2-weighted images revealed high signal at the marginal part and slightly median signal in the internal part. STIR images revealed a heterogeneous high signal in the interior. Furthermore, DWI and ADC map showed high signal and moderate-to-low signal intensity, respectively. ADC value of the lesion was 1.55×10^{-3} mm^2 s^{-1}. On extra-oral ultrasonography, the tumor showed clear boundary, hypoechoic, homogeneous internal architecture and vascular signals, and heterogeneous hard of the lesion. On intra-oral ultrasonography also showed clear boundary, hypoechoic, homogeneous internal architecture, heterogeneous hard of the tumor, and back echo enhance. The histopathologic diagnosis based on a full excisional specimen was odontogenic keratocyst. This case suggests that multimodality imaging, especially MR imaging with ADC and DWI, and extra and intra-oral ultrasonography with color Doppler imaging and elastography, could be effective for evaluating buccal lesions.

Keywords Apparent diffusion coefficient · Diffusion-weighted magnetic resonance imaging · Strain elastography · Shear wave elastography · Odontogenic keratocysts

Introduction

Odontogenic keratocyst (OKC) is a relatively common non-inflammatory jaw lesion to encounter [1]. Cross-sectional imaging is often used for diagnosis and management [2]. OKC is known to occur most often in the mandibular angle and mandibular ramus, but rarely outside the bone [3, 4]. OKC requires extreme caution in diagnosis and management because of its locally aggressive behavior and high recurrence rate [5].

Buccal space is a small and easily missed space in the head and neck region [6]. It is delineated by the facial muscles, the mandible, maxilla, and their muscles [7]. The buccal cavity contains a variety of tissues inside, including fatty tissue, salivary gland ducts, blood vessels, and nerves [8].

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In the buccal cavity, a variety of lesions occur, including tumors, inflammatory lesions, and cysts [9]. And not enough is known about the radiological features to differentiate them [8].

OKC occurring in soft tissues (peripheral OKC: POKC) is very rare, and there are few reports on CT, MRI, and ultrasonography imaging of OKC [10]. In this report, we describe characteristic multimodality imaging of OKC in the buccal space, especially diffusion-weighted MR imaging (DWI) with apparent diffusion coefficient (ADC) mapping, and extra-oral and intra-oral ultrasonography.

Case report

A 59-year-old man presented with mass on the left side of the buccal area for the past 2 years. On clinical examination, an approximately 20 mm in diameter mass with elastic hardness was found the left side of the buccal area. Contrast-enhanced CT was unclear due to artifacts, but showed areas of internal non-contrast lesions in the left buccal space (Fig. 1a), and bone tissue algorithm CT could not point to any obvious lesion area (Fig. 1b). On T1-weighted image, the mass showed multilocular high signal intensity, and homogeneous internal (Fig. 2a). T2-weighted images revealed high signal at the marginal part and slightly median signal in the internal part (Fig. 2b). STIR images revealed a heterogeneous high signal in the interior (Fig. 2c). Furthermore, DWI and ADC map showed high signal (Fig. 2d) and moderate-to-low signal intensity (Fig. 2e), respectively. ADC value of the lesion was 1.55×10^{-3} mm^2 s^{-1}. On extra-oral ultrasonography, the tumor showed clear boundary, hypoechoic, homogeneous internal architecture and vascular signals by color Doppler imaging (Fig. 3a), and heterogeneous hard of the lesion by shear wave elastography (Fig. 3b). The lesion had a maximum hardness of 102.9 ± 30.3 kPa and a minimum of 25.3 ± 11.5 kPa. On intra-oral ultrasonography also showed clear boundary, hypoechoic, homogeneous internal architecture (Fig. 4a), heterogeneous hard of the tumor and back echo enhance by strain elastography (Fig. 4b).

Histopathologic diagnosis on partial biopsy of the lesion was epidermoid cyst. Later, excisional surgery was performed under general anesthesia, and a $24 \times 23 \times 8$ mm large mass was removed. The histopathologic diagnosis based on a full excisional specimen was POKC. The cyst lumen is lined by 4–6 layer parakeratinized stratified squamous epithelium with surface corrugations and the basal layer is well defined and palisaded, with hyperchromatic nuclei and focal areas showing reversed nuclear polarity (Fig. 5a). To confirm the histopathological diagnosis of POKC for the present case, we performed immunohistochemical staining for parakeratinized lining epithelium. CK19, D2-40, and Cyclin D1 were positive in the lining epithelium (Fig. 5b, c, d).

Discussion

Boffano et al. [3] state that OKC occurs most commonly in the mandibular angle and mandibular ramus, while Chi et al. [4] state that OKC occurs rarely in soft tissues. Stoelinga et al. [11] reported that the majority of extra-osseous OKC in the mouth occur in the buccal space, and most of them showed lumps, and OKC occur in soft tissue have been reported to recur, as have OKC occurring within the maxilla and mandible. Therefore, correct diagnosis by preoperative imaging is considered important.

Usually, OKC shows unilocular or multilocular lesions with no internal contrast on contrast-enhanced CT, MR images show medium-to-high signal on T1-weighted images and heterogeneous signal on T2-weighted images,

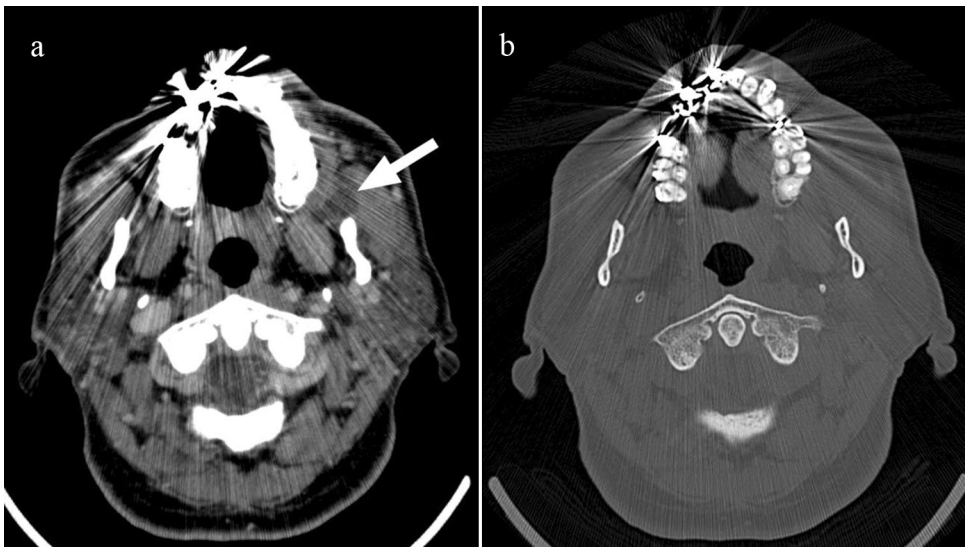

Fig. 1 Contrast-enhanced CT image showed a mass of internal non-contrast lesions in the left buccal space (**a**, arrow), and it is unclear due to artifacts. Bone tissue algorithm CT could not show obvious lesion area (**b**)

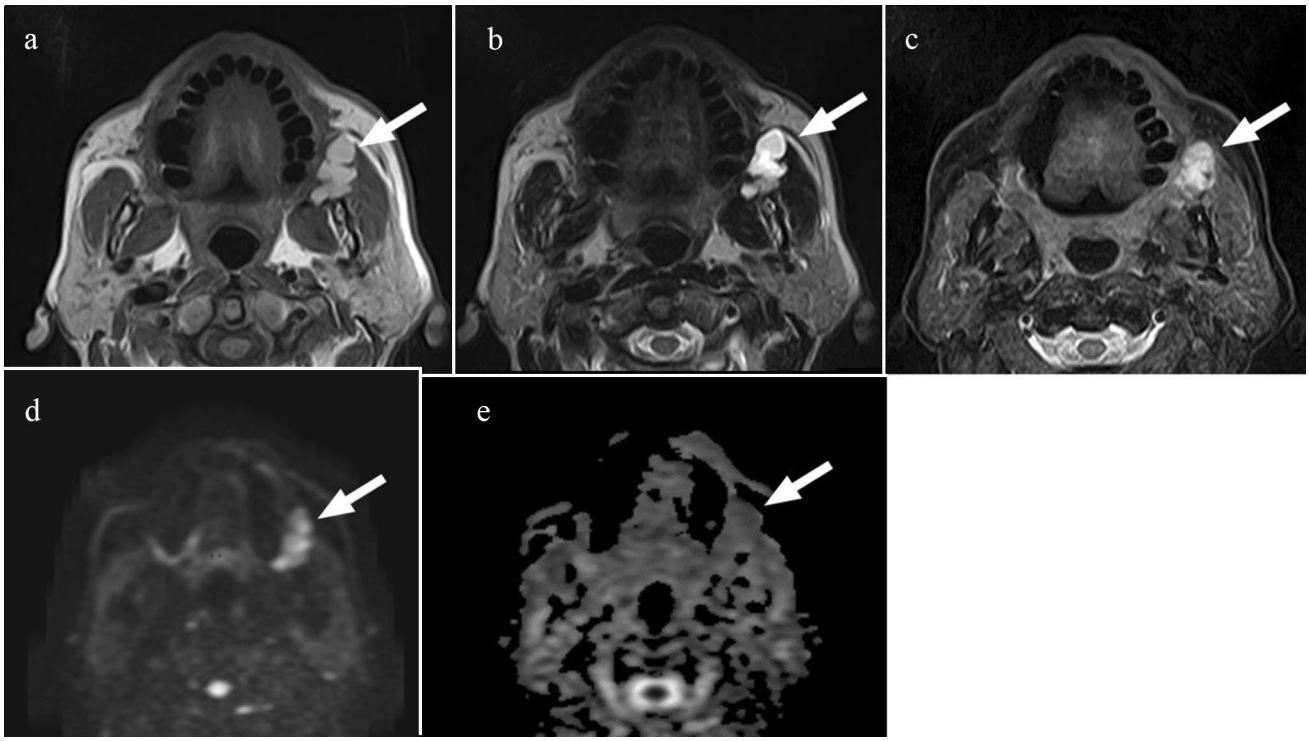

Fig. 2 On T1-weighted image, the mass showed multilocular high signal intensity (**a**, arrow), and T2-weighted images showed high signal at the marginal part and slightly median signal in the internal part (**b**, arrow), and STIR images showed heterogeneous high signal intensity (**c**, arrow), and DWI and ADC map showed high signal (**d**, arrow) and moderate-to-low signal intensity (e, arrow), respectively

and DWI and ADC are known to reflect the microstructure of the lesion and aid in the diagnosis of OKC [12]. Sumi et al. [13] reported the ADC value of OKC as $1.13 \pm 0.56 \times 10^{-3}$ mm^2 s^{-1}, and Zhu et al. [14] reported the ADC value of OKC occurring on the buccal space as 1.29×10^{-3} mm^2 s^{-1}. In our case, the ADC value was 1.55×10^{-3} mm^2 s^{-1}, which is higher than that reported by Sumi et al. [13] ADC may reflect local water diffusion phenomena, which may result in higher values compared to lesions in the bone. We also indicate that the difference in ADC from that reported by Zhu et al. [14] is due to differences in the amount of internal keratin. Beena et al. [15] reported that the ultrasonography of OKC in the buccal region was well-defined homogeneously hyperechoic focal lesion. In our case, the internal homogeneous hypoechoic, which may also be due to different amounts of internal keratin. It is also considered that the distribution of the internal keratin may have resulted in the heterogeneous hardness.

To consider extra-osseous OKC as a preoperative diagnosis, we consider it especially important to determine internal keratin retention and heterogeneous internal by functional MR imaging and ultrasonography. However, OKC has histopathologic characteristics similar to those of epidermoid cysts; therefore, differentiating OKC from epidermoid cysts on imaging may be difficult. According to Anantanarayanan et al. [16], epidermoid cysts have no internal blood flow, but our case shows blood flow within the lesion by ultrasonography with color Doppler imaging, which may be useful in differentiating from epidermoid cysts.

Previous reports indicated that CK19 and D2-40 are positive in the lining epithelium of odontogenic keratocysts and negative in epidermal cysts [17, 18], and Cyclin D1 is positive in the lining epithelium of odontogenic cysts including odontogenic keratocysts [19, 20]. In the present case, CK19, D2-40, and Cyclin D1 were positive in the lining epithelium. The immunohistochemical profile for the present case was identical to those for OKC.

In conclusion, we reported multimodality imaging characteristics of OKC occurring in the buccal space. This case suggests that multimodality imaging, especially MR imaging with ADC and DWI, and extra- and intra-oral

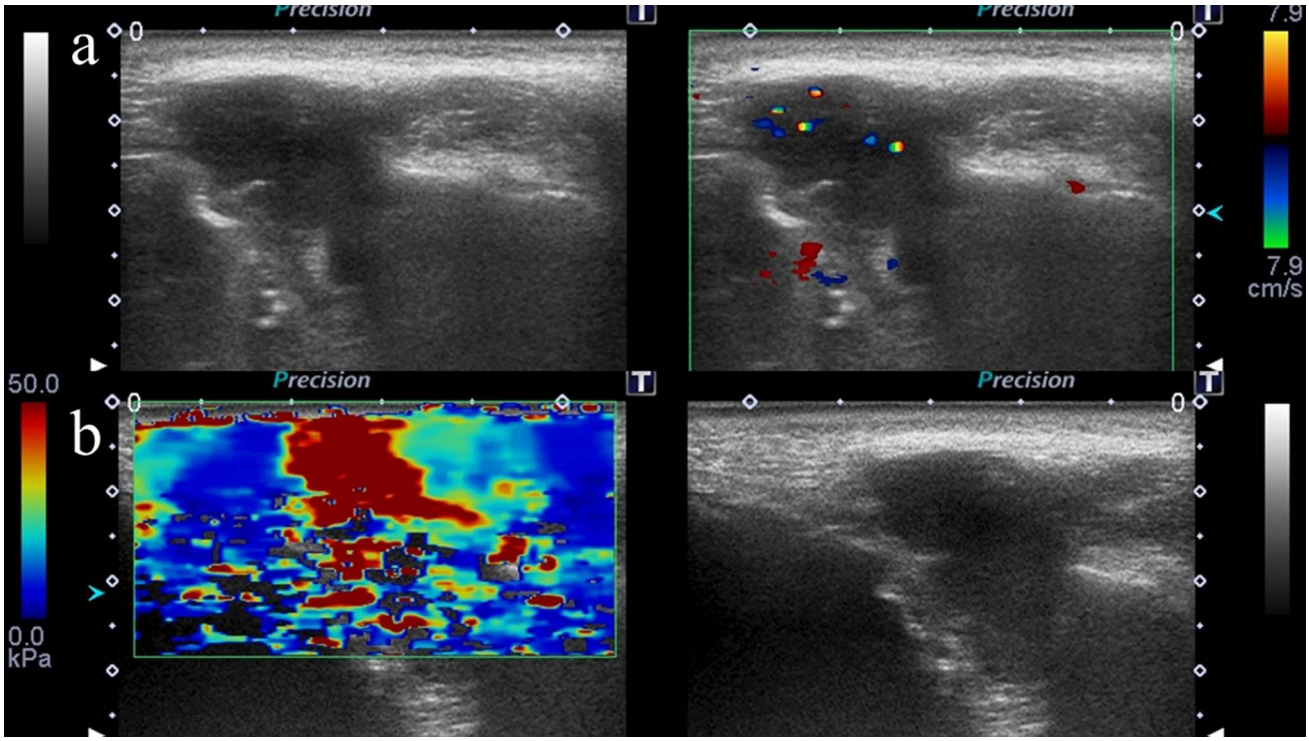

Fig. 3 On extra-oral ultrasonography, the tumor showed clear boundary, hypoechoic, homogeneous internal architecture and vascular signals by color Doppler imaging (**a**), and heterogeneous hard of the lesion by shear wave elastography (**b**). The lesion had a maximum hardness of 102.9 ± 30.3 kPa and a minimum of 25.3 ± 11.5 kPa

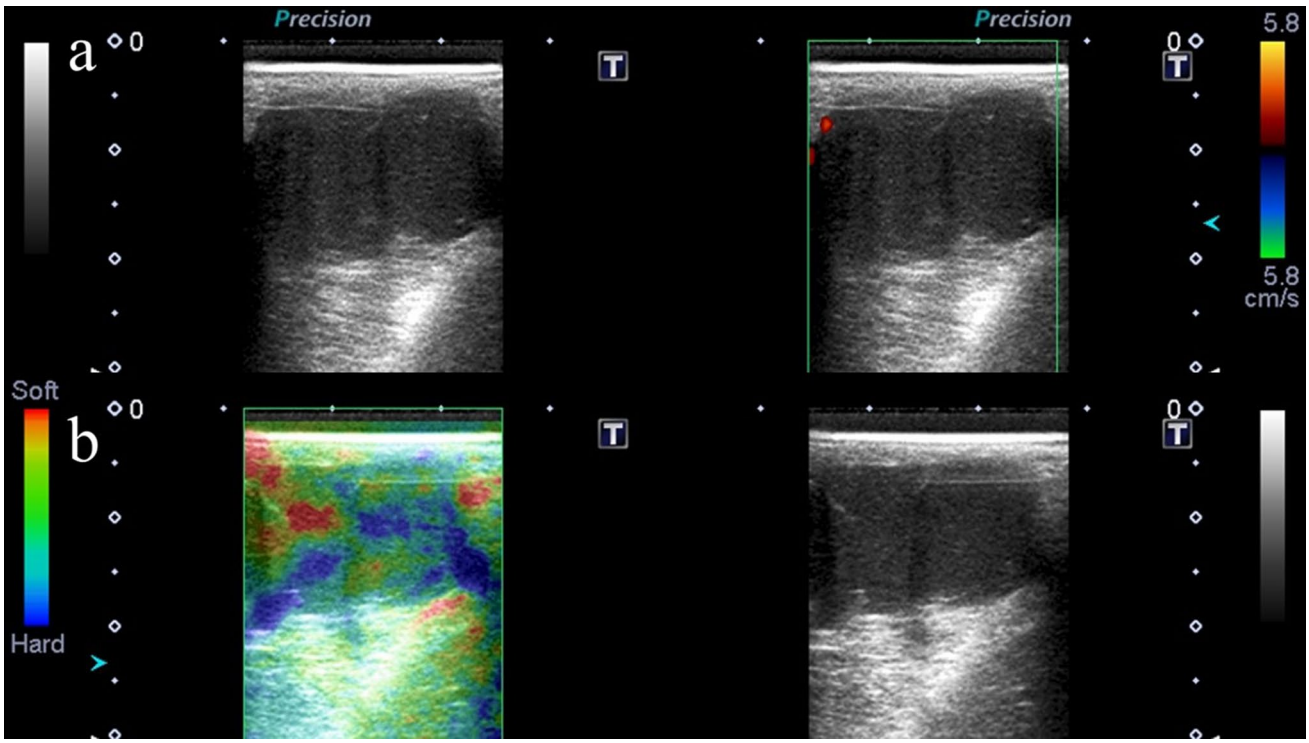

Fig. 4 On Intra-oral ultrasonography also showed clear boundary, hypoechoic, homogeneous internal architecture by color Doppler imaging (**a**), and heterogeneous hard of the tumor and back echo enhance by strain elastography (**b**)

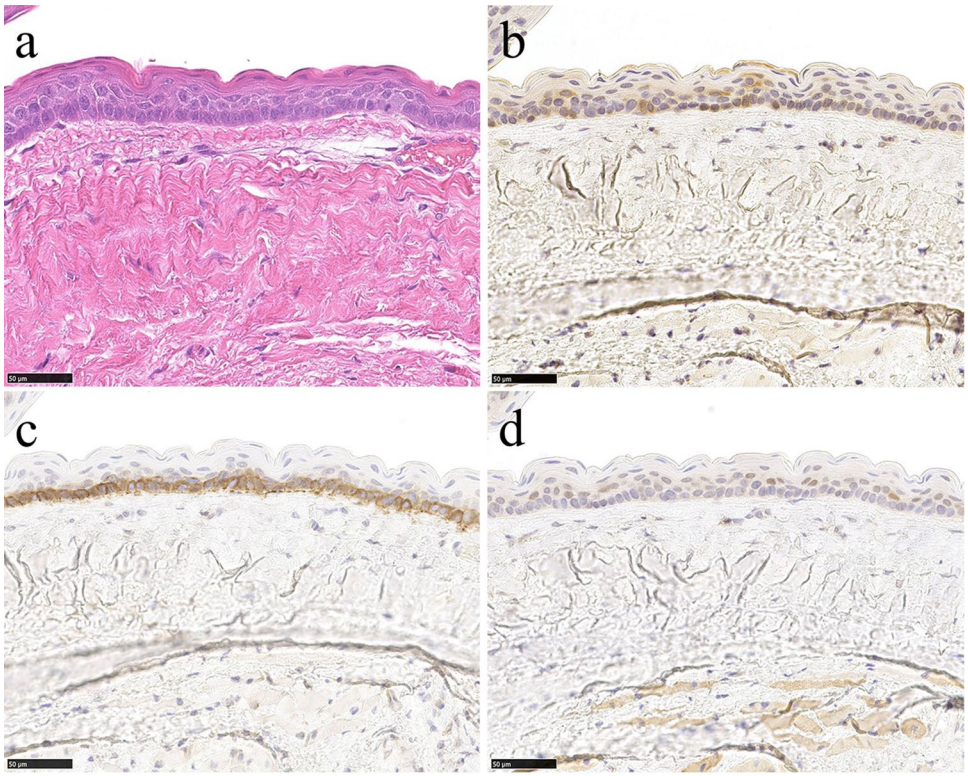

Fig. 5 Histopathological and immunohistochemical findings. The cyst lumen is lined by 4–6 layer parakeratinized stratified squamous epithelium with surface corrugations and the basal layer is well defined and palisaded, with hyperchromatic nuclei and focal areas showing reversed nuclear polarity (**a**), CK19 immunohistochemical staining shows positive finding of the entire lining epithelial cytoplasm (**b**), D2-40 immunohistochemical staining shows positive finding of the basal layer cell membrane (**c**), and Cyclin D1 immunohistochemical staining shows positive finding of the nuclei of spinous cell layer (**d**). Scale bars: 50 µm

ultrasonography with color Doppler imaging and elastography, could be effective for evaluating buccal lesions.

Declarations

Conflict of interest The authors declare that they have no conflict of interest.

Ethical standard All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008 (5).

Informed consent Informed consent was obtained from all patients for being included in the study.

References

1. Rajendra Santosh AB. Odontogenic cysts. Dent Clin North Am. 2020;64:105–19.
2. Vanagundi R, Kumar J, Manchanda A, Mohanty S, Meher R. Diffusion-weighted magnetic resonance imaging in the characterization of odontogenic cysts and tumors. Oral Surg Oral Med Oral Pathol Oral Radiol. 2020;130:447–54.
3. Boffano P, Ruga E, Gallesio C. Keratocystic odontogenic tumor (odontogenic keratocyst): preliminary retrospective review of epidemiologic, clinical, and radiologic features of 261 lesions from University of Turin. J Oral Maxillofac Surg. 2010;68:2994–9.
4. Chi AC, Owings JR Jr, Muller S. Peripheral odontogenic keratocyst: report of two cases and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2005;99:71–8.
5. Pitak-Arnnop P, Chaine A, Oprean N, Dhanuthai K, Bertrand JC, Bertolus C. Management of odontogenic keratocysts of the jaws: a ten-year experience with 120 consecutive lesions. J Craniomaxillofac Surg. 2010;38:358–64.
6. Hasan Z, Tan D, Buchanan M, Palme C, Riffat F. Buccal space tumours. Auris Nasus Larynx. 2019;46:160–6.
7. Kumar M, Kudva A, Vineetha R, Solomon M. Unilateral buccal space masses: a case series. Med Pharm Rep. 2020;93:310–3.
8. Kim HC, Han MH, Moon MH, Kim JH, Kim IO, Chang KH. CT and MR imaging of the buccal space: normal anatomy and abnormalities. Korean J Radiol. 2005;6:22–30.
9. Ogura I, Kaneda T, Sasaki Y, Sekiya K, Tokunaga S. Characteristic power Doppler sonographic images of tumorous and non-tumorous buccal space lesions. Dentomaxillofac Radiol. 2013;42:20120460.
10. Hornillos-de Villota M, Pampin-Martínez MM, Moran-Soto MJ, Cebrián-Carretero JL. Peripheral odontogenic keratocyst. A Case report. J Clin Exp Dent. 2023;15:e169–72.
11. Stoelinga PJ, Grillo R, da Silva YS. The extra-osseous odontogenic keratocyst: an anachronism? J Stomatol Oral Maxillofac Surg. 2022;123:e790–3.
12. Borghesi A, Nardi C, Giannitto C, Tironi A, Maroldi R, Di Bartolomeo F, et al. Odontogenic keratocyst: imaging features of a benign lesion with an aggressive behaviour. Insights Imaging. 2018;9:883–97.
13. Sumi M, Ichikawa Y, Katayama I, Tashiro S, Nakamura T. Diffusion-weighted MR imaging of ameloblastomas and keratocystic odontogenic tumors: differentiation by apparent diffusion coefficients of cystic lesions. AJNR Am J Neuroradiol. 2008;29:1897–901.

14. Zhu L, Yang J, Zheng JW. Radiological and clinical features of peripheral keratocystic odontogenic tumor. Int J Clin Exp Med. 2014;15(7):300–6.
15. Beena VT, Meleveetil DB, Cheriyan LM, Angamuthu K. Mucosal keratocyst of buccal mucosa: a rare entity. J Oral Maxillofac Pathol. 2020;24:589.
16. Anantanarayanan P, Manikandhan R, Bhargava D, Sivapathasundaram B. Sub-lingual epidermoid cyst. Head Neck Pathol. 2010;4:136–8.
17. Koizumi Y. Odontogenic keratocyst, orthokeratinized odontogenic cyst and epidermal cyst: an immunohistochemical study including markers of proliferation, cytokeratin and apoptosis related factors. Int J Oral-Med Sci. 2004;2:14–22.
18. Ide F, Kikuchi K, Miyazaki Y, Mishima K, Saito I, Kusama K. Keratocyst of the buccal mucosa: is it odontogenic? Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2010;110:e42–7.
19. Taghavi N, Modabbernia S, Akbarzadeh A, Sajjadi S. Cyclin D1 expression in odontogenic cysts. Turk Patoloji Derg. 2013;29:101–7.
20. Júnior JF, de França GM, da Silva Barros CC, Felix FA, da Silva WR, de Lucena HF, et al. Biomarkers involved in the proliferation of the odontogenic keratocyst, glandular odontogenic cyst and botryoid odontogenic cyst. Oral Maxillofac Surg. 2022;26:655–62.