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Original Article

Diagnostic approach for the rare anterior variant of mandibular bone depression often misdiagnosed as tumorous lesions

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Diagnostic errors;
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Abstract *Background/purpose:* This study analyzed the clinical and imaging features of lingual mandibular bone depression (LMBD) in the anterior mandible, aiming to prevent misdiagnosis and unnecessary surgical procedures.
Materials and methods: The patients who visited a university dental hospital for painless radiolucency in the anterior mandible from January 2010 to December 2022 were retrospectively reviewed. Twelve cases of LMBD in the anterior mandible that are confirmed by biopsy or long-term follow-up were identified. Two oral and maxillofacial radiologists evaluated the imaging features. Additionally, 12 cases were manually collected from case reports published between 2001 and 2022. Clinical and histopathologic data were obtained from both groups and clinical information were compared using Fisher's exact test.
Results: The clinical information of the patients and that from the case reports showed no statistically significant differences, except for the clinical impression ($P = 0.005$). The imaging features of anterior LMBD included the absence of lingual cortical expansion and soft tissue bulging, a mostly round cortical border, and muscle-level attenuation, as observed on multidetector computed tomography (MDCT). Occasionally, the progression of LMBD led to thinning of the labial cortex.
Conclusion: If non-specific clinical features are present, MDCT is recommended to distinguish anterior LMBD from tumorous lesions that require surgical intervention.

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Introduction

Lingual mandibular bone depression (LMBD), also known as the Stafne defect, was first identified in 1942 as a developmental lingual depression near the angle of the mandible, situated below the mandibular canal.[1] It is typically detected incidentally on panoramic radiography as a radiolucent finding, since the defect is asymptomatic. Subsequent to its initial recognition, a multitude of cases have been reported and examined.[2,3] A study classified bone depression based on the extent and content observed in computed tomography.[4] LMBD does not necessitate treatment; however, it is imperative to distinguish it from pathological lesions to prevent unnecessary invasive procedures.

Anterior variants of LMBD, located in or anterior to the premolar region, have been described in individual cases,[5–16] although they are rare.[17] Most patients did not experience pain, yet these cases were often mistaken for pathologic lesions in panoramic radiography due to the superimposition over the roots of the anterior teeth.[6,8,13] Surgical procedures were performed in a few cases,[6,10,16] and one case even underwent mandibulectomy.[10] The majority of these cases were diagnosed as LMBD through additional imaging techniques, such as multidetector computed tomography (MDCT) or cone-beam computed tomography (CBCT) and subsequently confirmed by long-term follow-up.[5–10,13–16] Some cases were also histopathologically diagnosed.[6–10,13–16]

Discussions on LMBD have focused on its prevalence,[3] classification[4] and location.[17] However, no study has yet investigated the diagnostic indicator for the anterior variant of the defect based on a case collection, despite the use of invasive procedures for diagnosis.[6,10,16] The objective of this study was to provide guidance for diagnosing LMBD in the anterior mandible, which is often mistaken for a pathologic lesion. By analyzing clinical and imaging features, this study aimed to prevent misdiagnosis and unnecessary surgical intervention.

Materials and methods

Patient group

Cases of LMBD in the anterior mandible were retrospectively reviewed from the patient records at a university dental hospital, spanning from January 2010 to December 2022. Cases that were confirmed as LMBD through biopsy, surgical exploration, or long-term follow-up were included and those with significant artifacts in images that hindered diagnosis, or those lacking clinical evaluation were excluded. As a result, 12 cases were included in this study, and both clinical information and imaging features were assessed. Informed consent was waived due to the retrospective nature of the study by the Institutional Review Board (IRB) of Yonsei University Dental Hospital (IRB No. 2-2023-0068).

Case report group[5–16]

Published case reports of LMBD in the anterior mandible were reviewed, and 12 case reports, each detailing a single case of anterior LMBD confirmed by biopsy or long-term follow-up, were included. The clinical information and imaging features described in the literature were collected in the same manner as for the 12 cases acquired for this study. When any evaluation criterion was not reported, it was recorded as "unknown."

Clinical information

Demographic information, including age and sex, along with anatomical location details, symptoms, reasons for the visit, clinical impressions, imaging results, course of treatment (including any surgery), and histopathological diagnoses, were all obtained through electronic medical records.

Imaging features

The evaluation criteria for imaging features were analyzed in both the bone and soft tissue portions as depicted in CBCT, MDCT, magnetic resonance imaging (MRI), or contrast-enhanced MDCT (Fig. 1). For the bone portion, CBCT, MDCT, or pre-contrast MDCT images were assessed with reference to panoramic radiographs (Fig. 2). The criteria included cortical border (total, partial, none), shape (round, irregular), lingual cortical expansion (present, absent), and labial cortical thinning (present, absent). The soft tissue portion was evaluated using MDCT and

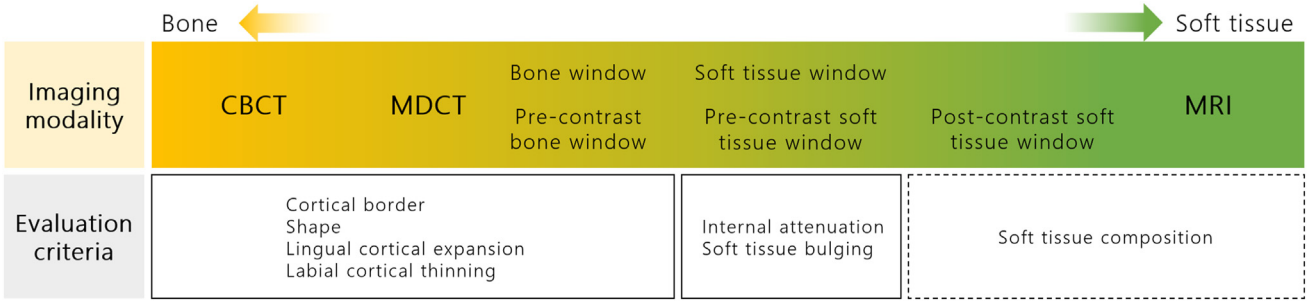

Figure 1 Schematic diagram of evaluation criteria for the imaging features of lingual mandibular bone depression for each imaging modality in the spectrum of bone to soft tissue visualization. Since most cases included MDCT, all six criteria in the lined boxes were assessed. The criterion in the dotted box was assessed in a few cases with post-contrast MDCT or MRI. MDCT, multidetector computed tomography; MRI, magnetic resonance imaging.

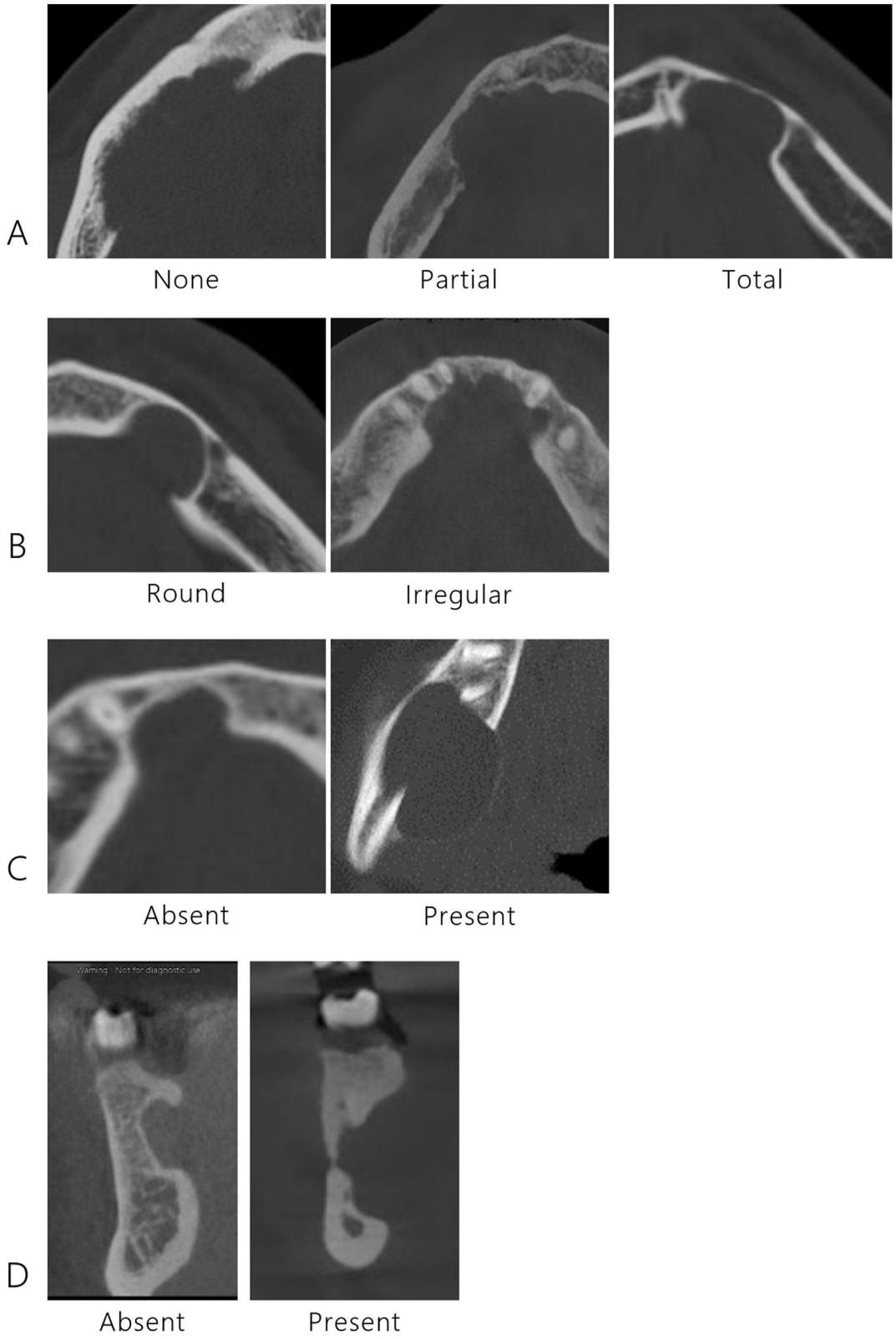

Figure 2 Examples of CBCT or bone-window MDCT images of each evaluation criterion for bone portion imaging features. (A) Cortical border, (B) shape, (C) lingual cortical expansion, and (D) labial cortical thinning. CBCT, cone-beam computed tomography. MDCT, multidetector computed tomography.

pre-contrast MDCT images (Fig. 3), considering overall internal attenuation (low, resembling fat attenuation; intermediate, resembling muscle attenuation; high, resembling cortical bone attenuation), and the presence or absence of soft tissue bulging. Additionally, the composition of the soft tissue was assessed in two cases using contrast-enhanced MDCT or MRI. Two experienced oral and maxillofacial radiologists, familiar with anterior LMBD, reviewed the images. In instances of differing opinions, the radiologists engaged in discussion until a consensus was reached.

Statistical analysis

The mean ages of the patient group and the case report group were compared using the Mann–Whitney test with a significance level set at 95%. Clinical information from the two groups was compared using Fisher's exact test, also at a 95% significance level. Imaging features of both groups were analyzed descriptively.

Results

The clinical information for the cases is summarized in Table 1. The mean age of the patient group was 49.3 ± 15.9 years, while the mean age of the case report group[5–16] was 49.1 ± 17.2 years. The difference was not statistically significant ($P = 0.799$). No significant differences were observed in other clinical data between the groups ($P > 0.05$). However, the clinical impression varied significantly, ranging from LMBD to malignancy and from inflammation to benign tumor in the patient and case report groups,[5–16] respectively ($P = 0.005$). Histopathologic diagnoses were available for 2 cases in the patient group (Fig. 4) and 6 cases in the case report group.[5–16] Most were diagnosed as salivary gland tissue (7 of combined 8 cases)[6,10–13,16] and one case in the case report group also contained fat and vasculature,[6] indicative of non-pathologic findings. One case in the patient group was surgically explored where only vacant space was found.

The imaging features of each case are summarized in Table 2. Overall, there was no evidence of lingual cortical expansion or soft tissue bulging in any of the cases, and the cortical border was at least partially intact in all cases, with most being completely corticated. The lesions were predominantly round, and the internal attenuation was mostly intermediate, similar to that of muscle. Occasionally, the bone depression extended through the bone marrow to the labial side, resulting in cortical thinning. In the contrast-enhanced MDCT and MRI of a few in the patient group, as well as some in the case report group,[7,16] the enhancement of the soft tissue and the signal intensity were continuous and comparable to that of the sublingual gland (Fig. 5).

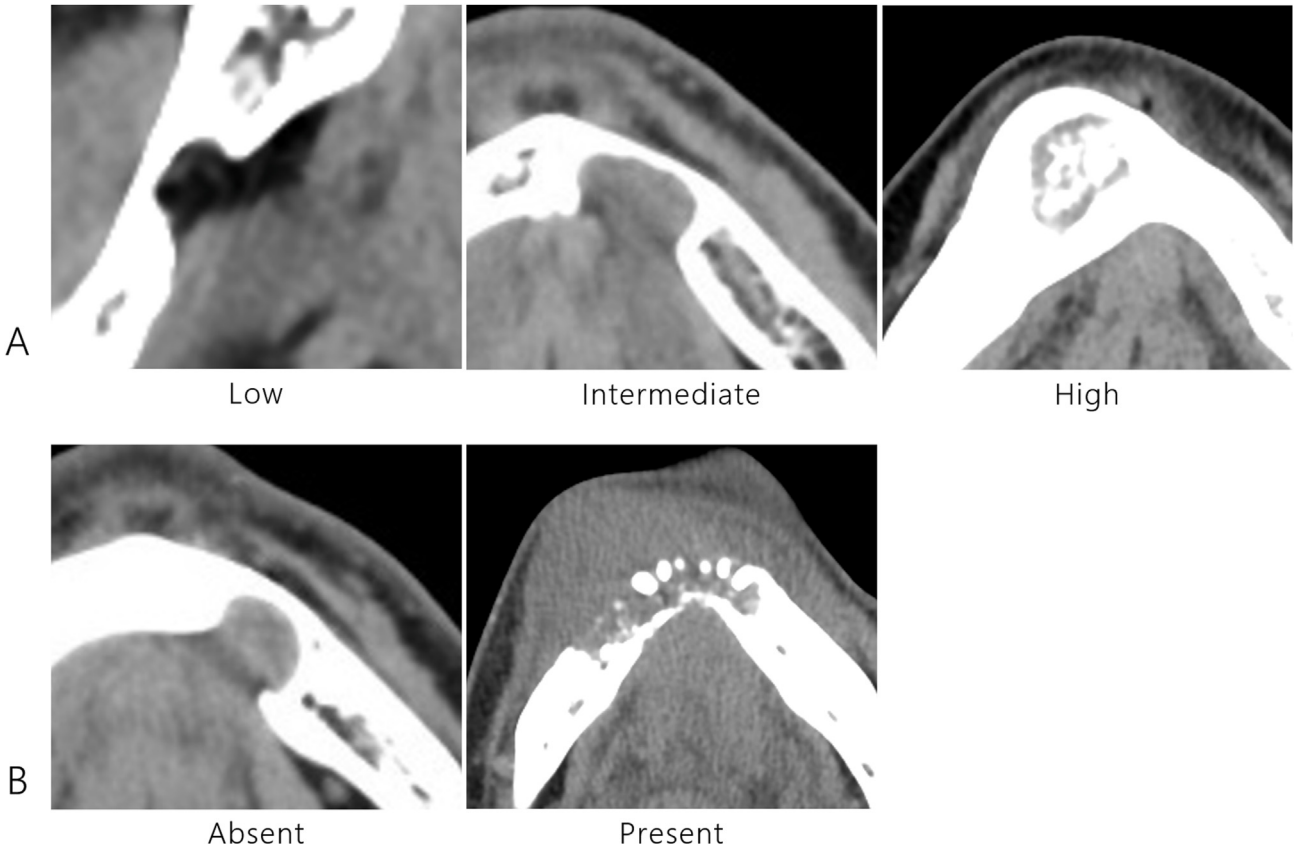

Figure 3 Examples of soft-tissue window MDCT images of each evaluation criteria for soft tissue portion imaging features. (A) Internal attenuation, and (B) soft tissue bulging. MDCT, multidetector computed tomography.

Table 1 The clinical information of the patients and case reports reviewed.[5–16].

Clinical information	Patients	Case reports	P-value
Sex			0.660
Male	9	8	
Female	3	4	
Location			0.140
Right	5	2	
Left	6	8	
Midline	1	0	
Bilateral	0	2	
Center in mandible			0.569
Premolar	6	7	
Canine	4	4	
Incisor	2	1	
Symptom			0.268
No symptom	11	10	
Tooth mobility	1	0	
Pain	0	1	
Unknown	0	1	
Reason for visit			–
Incidental finding	12	12	
Others	0	0	
Clinical impression			0.005*
Benign tumor	4	2	
LMBD	4	0	
Cyst	3	3	
Malignancy	1	0	
Inflammation	0	4	
Unknown	0	3	
Imaging			
Panoramic radiography			0.317
Yes	12	11	
No	0	1	
Computed tomography			0.599
MDCT	8	8	
CBCT	2	2	
CBCT + MDCT	2	0	
Other imaging modality			0.546
MRI	1	2	
Course of treatment			0.397
Follow-up	9	7	
Surgery	3	5	
Total	12	12	

CBCT, cone-beam computed tomography; LMBD, lingual mandibular bone depression; MDCT, multidetector computed tomography; MRI, magnetic resonance imaging.

Discussion

The objective of this study was to provide guidance for diagnosing LMBD in the anterior mandible, which is often mistaken for a pathologic lesion. By analyzing clinical and imaging features, this study sought to prevent misdiagnosis and unnecessary extensive surgical procedures. The only significant difference ($P = 0.005$) was observed in the clinical impression between the patient group and the case report group; other clinical features did not show significant differences. As the anterior presentation of LMBD is very rare,[17] the clinical impressions significantly differed among clinicians, ranging from cysts to malignant tumors. Thus, surgical procedures were done in some cases, as complex as mandibulectomy,[10] but the histopathologic findings were non-specific.[6,10–13,16]

LMBD is most commonly found in the posterior mandible, below the mandibular canal. Its diagnosis is typically straightforward, even when using panoramic radiography.[1–3] The typical findings are a round or ovoid radiolucency with thick and well-defined margins, and CBCT is usually sufficient for a conclusive diagnosis.[1–3] The radiolucency is often filled with fat-level attenuation, which can be visualized using MDCT.[4]

On the contrary, the anterior variant of LMBD is uncommon and some imaging features differ from LMBD in the posterior mandible.[5–16] Typically, it is asymptomatic and presents as a radiolucent area, similar to the classic presentation of LMBD.[5–16] However, the distinguishing features can complicate the diagnosis. Occasionally, LMBD may be located in the periapical region or above the mental foramen, the cortical margin may be partially absent, and the shape may be irregular. Additionally, the radiolucency often appears filled with soft tissue equivalent to muscle on MDCT.[14,15] Lingual cortical bone expansion is not found[6–10,13–16] but labial cortical thinning is occasionally seen.[7–10,13,14] Due to its location and these imaging features, anterior LMBD is frequently mistaken for a cyst or tumor.[5,7,8,10–13,15,16] This explains the significant difference in clinical impressions between the patient and case report groups.[5–16]

The absence of soft tissue bulging on MDCT is a critical diagnostic indicator of anterior LMBD, which is corroborated by post-contrast MDCT or MRI findings that show the extension of the salivary gland.[7,16] This observation helps to rule out the possibility of a cyst or tumor, particularly when the shape is irregular[7,10,11,16] or the cortical margin is partially absent.[7,10–13,16] Therefore, MDCT is sufficient for the imaging diagnosis of anterior LMBD, and additional post-contrast MDCT or MRI can be obtained for confirmation.

The most critical pathologies in the differential diagnosis of anterior LMBD include salivary gland tumors.[18–21] The sublingual gland abuts the anterior mandible, and extension of the tumor is a distinct possibility.[19] Moreover, intraosseous malignant salivary gland tumors are known to occur in the mandible, with the premolar area being the most common site.[18,19,21] In this way, misdiagnosing anterior LMBD as a salivary gland tumor could lead to unnecessary surgery on a large scale, resulting in irreversible changes for the patient. Therefore, to exclude pathologies such as salivary gland tumors, as well as other odontogenic cysts, tumors, or inflammatory changes suspected in this study, symptoms and clinical presentation must be thoroughly evaluated first. If clinical features suggest a positive finding, contrast-enhanced imaging would be necessary for an accurate differential diagnosis. However, if the site is asymptomatic and presents with non-specific features, as in the cases described in this study and as formerly mentioned,[22] plain MDCT would suffice.

One of the significant aspects of this study is that it presents and rigorously analyzes the largest number of total cases to date, comprising 24 cases from 12 patient cases and 12 reported cases. Given the rarity of the anterior

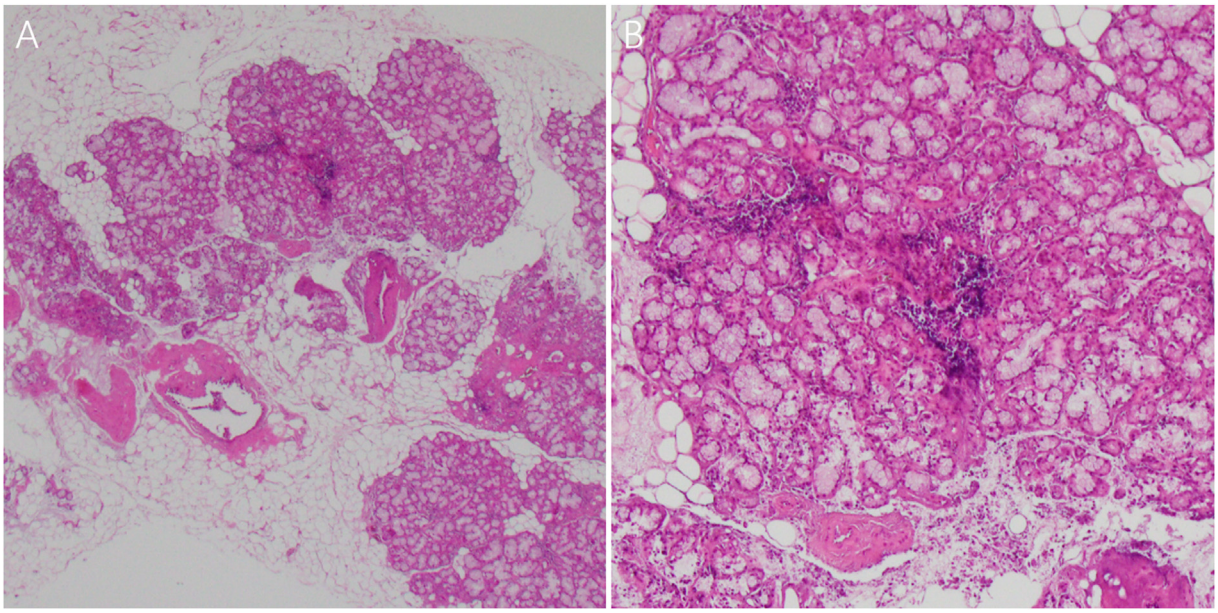

Figure 4 Histopathologic images of a case in the patient group (hematoxylin and eosin staining). (A) The specimen included minor salivary glands, adipose tissues, and blood vessels (× 12.5). (B) Infiltrate of inflammatory cells was observed partially (× 40).

Table 2 Imaging features of the patients and case reports reviewed.[5–16].

Case number[a]	Bone				Soft tissue	
	Cortical border[b]	Shape[c]	Lingual cortical expansion	Labial cortical thinning	Overall attenuation[d]	Bulging
P1	Total	Round	−	+	Intermediate	−
P2	Total	Round	−	−	Intermediate	−
P3	Total	Round	−	+	Intermediate	−
P4	Total	Round	−	+	Intermediate	−
P5	Total	Round	−	−	Intermediate	−
P6	Total	Irregular	−	+	Intermediate	−
P7	Total	Round	−	−	Intermediate	−
P8	Total	Round	−	−	N/A	N/A
P9	Total	Round	−	−	Low	−
P10	Partial	Round	−	+	Intermediate	−
P11	Partial	Irregular	−	+	Intermediate	−
P12	Total	Irregular	−	+	N/A	N/A
C1	Total	Round	−	−	Low	Unknown
C2	Total	Round	−	−	Low	−
C3	Partial	Irregular	−	+	N/A	N/A
C4	Total	Round	−	+	Unknown	Unknown
C5	Unknown	Round	−	+	Low	−
C6	Partial	Irregular	−	+	N/A	N/A
C7	Partial	Irregular	Unknown	Unknown	N/A	N/A
C8	Partial	Round	Unknown	Unknown	N/A	N/A
C9	Partial	Round	−	+	Unknown	Unknown
C10	Total	Round	−	+	Intermediate	−
C11	Total	Round	−	−	Intermediate	−
C12	Partial	Irregular	−	−	Unknown	−

N/A, not applicable.
[a] P1 to P12 indicate patient cases, and C1 to C12 indicate case reports.
[b] Cortical border was classified as total, partial, or absent.
[c] Shape was classified as round or irregular (partially or completely irregular).
[d] Attenuation was classified as low (fat-level), intermediate (muscle-level), and high (cortical bone-level).

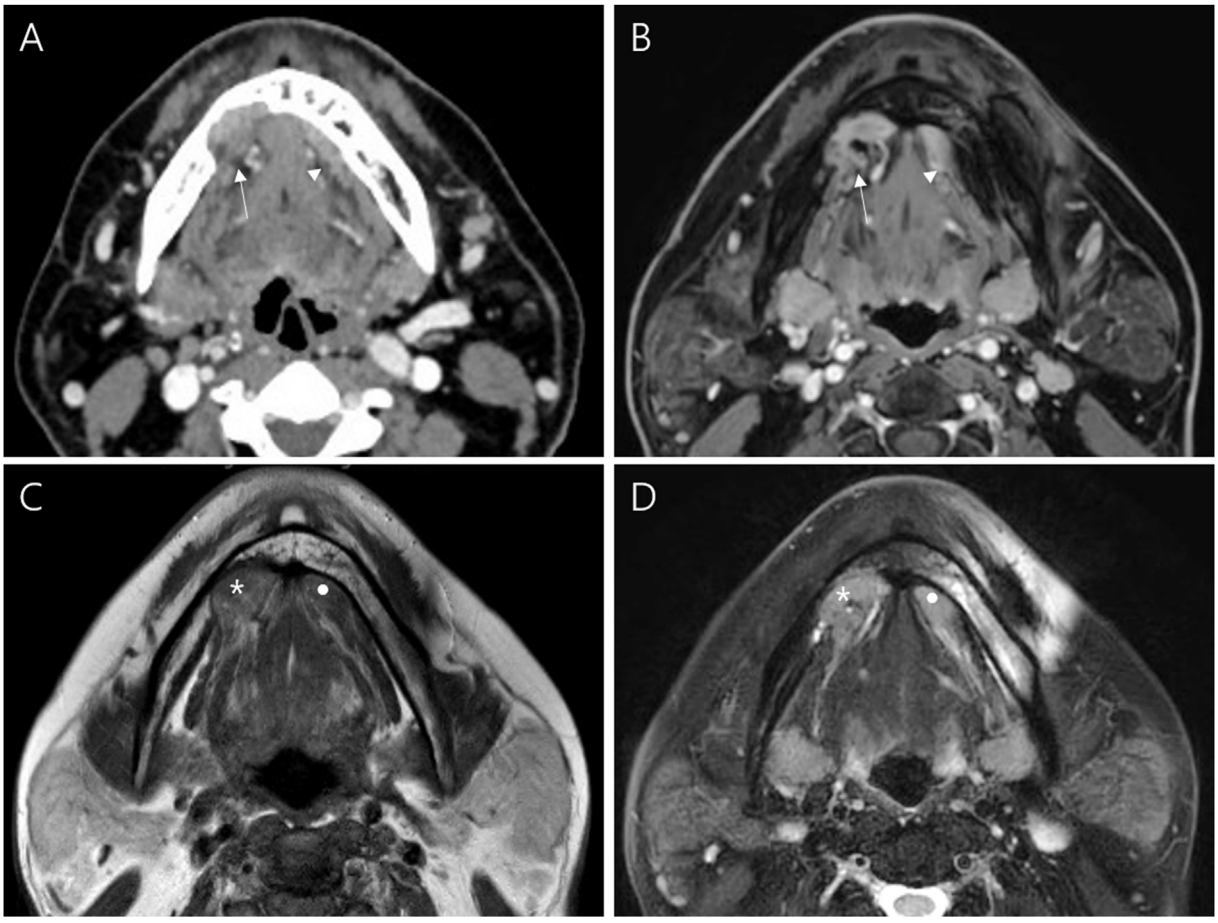

Figure 5 (A) Post-contrast image of MDCT and (B) contrast-enhanced T1-weighted image of MRI indicate that soft tissue within the bone depression in right mandible (arrows) is enhanced at the same level as contralateral sublingual gland (arrowheads) without any bulging. (C) T1-weighted and (D) T2-weighted MRI scans also show the soft tissue (asterisks) with an equivalent signal intensity to that of the sublingual gland (dots). The histopathologic diagnosis was confirmed as salivary gland. MDCT, multidetector computed tomography; MRI, magnetic resonance imaging.

variant of LMBD, previous case reports typically presented a single case, and the most extensive literature review on LMBD to date included only six cases.[17] This study is also the first to propose critical imaging features for diagnosing anterior LMBD, drawing on a comprehensive review of case reports and patient cases. The collected case information was organized to analyze clinical and imaging features, providing suggestions for diagnostic criteria.

This study has several limitations. First, the number of patient cases is relatively small, though it represents the largest collection to our knowledge. The prevalence of LMBD is reported to be 0.17%[3] and among LMBDs, approximately 5.5% are anterior variants.[17] The low prevalence of anterior LMBD inherently leads to small case series. In addition, the patient cases involved different types of imaging modalities, including CBCT, MDCT, and MRI.[5–16] The choice of imaging was influenced by the initial clinical impression, which varied, leading to the use of different imaging modalities.

In conclusion, the differential diagnosis of LMBD in the anterior mandible can be challenging in a clinical setting. When a non-specific clinical feature is present, MDCT is recommended to exclude tumorous lesions by identifying key imaging features and diagnosing the anterior variant of LMBD.

Declaration of competing interest

The author has no conflicts of interest relevant to this article.

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None.

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