

Anterior Lingual Mandibular Bone Depression in an 11-Year-old Child

REINHARD E. FRIEDRICH¹, HANNA A. SCHEUER² and ALEXANDER GRÖBE¹

¹Oral and Maxillofacial Surgery, ²Orthodontics, Eppendorf University Hospital, University of Hamburg, Hamburg, Germany

Abstract. *This report describes physical and imaging findings in a case of anterior lingual mandibular bone depression in a child. This entity is very rarely diagnosed and even more extremely rarely seen in children. We present some characteristic findings depicted on images provided by different sources and briefly address current hypotheses on its pathogenesis.*

The anterior lingual mandibular bone depression (ALMBD) is a rare finding, firstly reported by Richard and Ziskind (1). The pathogenesis of ALMBD is still unknown. ALMBD is categorized as the anterior counterpart to a similar posterior mandibular lingual bone depression (or defect) (PLMBD), preferentially known as Stafne's cavity (2, 3). ALMBD is much rarer than PLMBD. Based on a recent literature survey, the ratio is about 1:6.8 in favour of posterior lesions (4). Both defects frequently contain salivary gland tissue in an otherwise static osseous defect (3, 4). The frequent evidence for the presence of dystopic salivary gland tissue by different imaging techniques and, following surgical interventions, by histological proof, is the main reason explaining defects on both sides by one mechanism, *i.e.* the resorption of bone by continuous pressure of adjacent salivary glands (5). However, the exact mechanism of this bone resorption remains unclear. Both lesions are preferentially diagnosed in adults (2, 6, 7). In the most exhaustive survey on this entity only one case out of 40 ALMBD was diagnosed in the 10- to 19-year-old age group (4). Lingual mandibular bone defects of other sites were also reported, *e.g.* of the mandibular ramus (8, 9). However, these locations of mandibular lingual defects are even less common than ALMBD and PLMBD (4).

Correspondence to: Professor R.E. Friedrich, Oral and Maxillofacial Surgery, Eppendorf University Hospital, University of Hamburg, Martinistr. 52, D-20246 Hamburg, Germany. Tel: +4940741053259, e-mail: rfriedri@uke.de

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We present the extremely rare case of an anterior mandibular lesion in a child that demonstrated characteristic findings allowing for diagnosis of ALMBD.

Case Report

The 11-year-old patient was submitted to our outpatient clinic in order to define a lesion of the anterior mandible that was depicted on an orthopantomogram performed in the course of orthodontic therapy.

On admission, the healthy girl had an age-adjusted normal dentition with class III occlusion, no anomalies of the dental arches and a symmetrically developed face. All teeth were fixed in their alveoli and reacted promptly to adequate stimuli. The patient had been subjected to rapid palatal extension due to maxillary hypoplasia at the age of eight years. She had no further history of trauma or previous surgery in the oral and maxillofacial regions. Two of her sisters are affected by unilateral cleft lip and palate.

On the orthopantomogram, an ovally-osteolytic lesion was projected on the roots of the anterior teeth, predominantly on the left paramedian basal part of the mandible. The lesion was superimposed on the apical parts of the mandibular incisors (Figure 1D). The lesion appeared to be sharply demarcated but showed no sclerotic margin. Earlier orthopantomograms had been performed during the treatment planning and follow-up examinations of orthodontic therapy and were presented for evaluation (Figure 1A-C). One of the orthopantomograms, taken at the age of 9 years, already showed the radiolucency of the anterior mandible in close proximity to the anterior teeth (Figure 1B). However, the radiographs of the patient taken at the age of 8 years showed no such radiolucency (Figure 1A).

We decided to perform further imaging in order to delineate the borders of the lesion and to more accurately define its topography. On cone beam computed-tomograms, the osseous lesion was strictly lingually situated and coved into the bone (Figure 2). The margins of the lesion were distinct but the content of the lesion could not be further specified. On dynamic B-scan ultrasound images, normal anatomy of soft

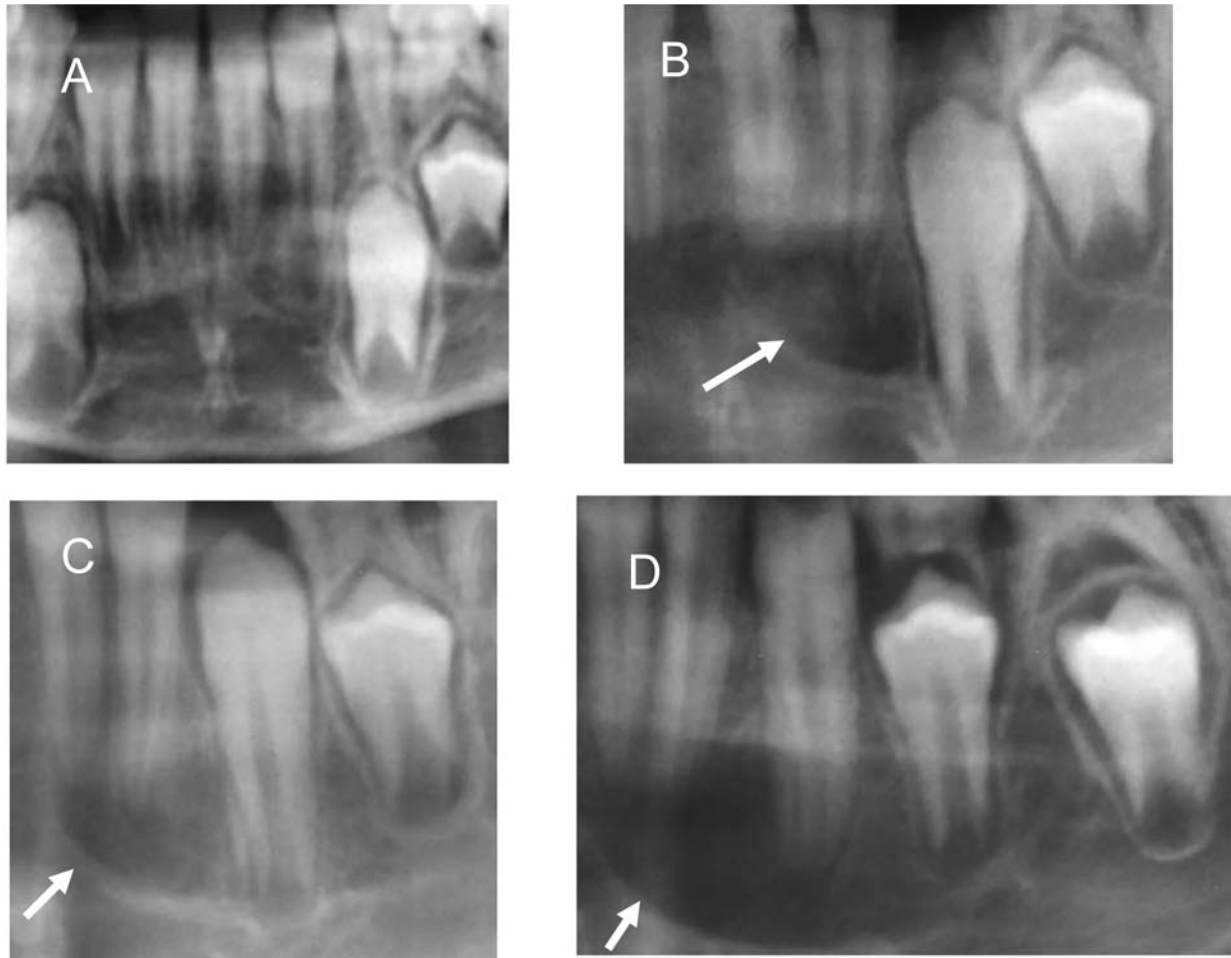


Figure 1. Cropped images of the anterior mandible, depicted on orthopantomograms of a patient at the age of 7 (A), 9 (B), 10 (C), and 11 (D) years. A: The anterior mandible exhibits normal osseous structures in the region of the left inferior lateral incisor and left inferior canine. B: Two years later, a small radiolucent area had become visible below the apex of the left lateral incisor. C: At the age of 10 years, this ovoid-shaped radiolucent zone extends below the incompletely developed root of the left inferior canine. The lesion exhibits a sclerotic margin at its lower border. D: At the time of admission, the lesion even extends to the left inferior first premolar. Arrows point to the mesial border of the lesion (B-D).

tissues located in the anterior floor of the mouth was depicted, with the sublingual gland being closely-attached to the bone (Figure 3C). No enlarged lymph nodes were visible in the region of the sublingual gland. However, transcutaneous ultrasound did not allow the complete mapping of the lingual bony surface. The left sublingual gland appeared to have a slightly greater transverse diameter compared to its counterpart. In order to identify soft tissue possibly located inside the cavity, we decided to perform magnetic resonance imaging (MRI) of the mandibular region. On MRI, the left sublingual gland was shown to extend anteriorly into the curved and sharply-demarcated lesion and completely occupied it (Figure 3A and 3B). These findings were summarized as an ALMBD. The patient and her parents were informed about the diagnosis and the proposed refraining from therapy. A regular follow-up

examination was instructed, preferably in co-ordination with the planned orthodontic therapy.

Discussion

This report presents a distinct osseous lesion of the anterior mandible in a child that fulfils the diagnostic criteria of ALMBD. The occurrence of ALMBD in childhood is extremely rare and was unknown in earlier surveys on this entity (10). Philipsen *et al.* recorded only one individual with an ALMBD in their large survey on buccal and lingual mandibular depressions (4). Based on the analysis of 42,600 orthopantomograms, the incidence of ALMBD was calculated to be 0.009% (4). ALMBD and PLMBD are expected to be findings identified occasionally on radiographs of the jaws in

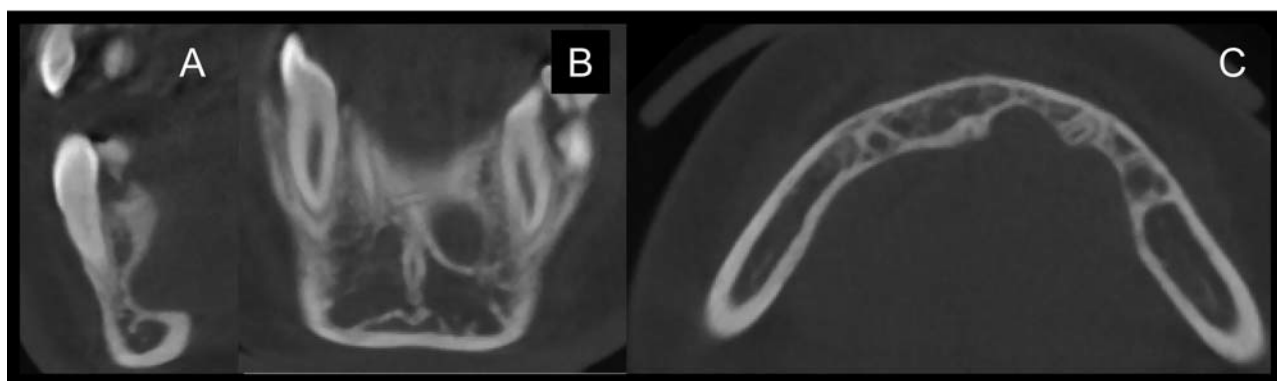


Figure 2. Cropped images derived from cone beam computed tomograms of the anterior mandible in three planes. A: In the sagittal view, the cove enters the mandibular base below the roots of the incisors. The borders appear in this section radiopaque equivalent to cortical bone. B: In the coronal view, the distinct borders at the most anterior part of the cavity are depicted. C: In the occlusal view, the bi-lobed depression becomes evident.

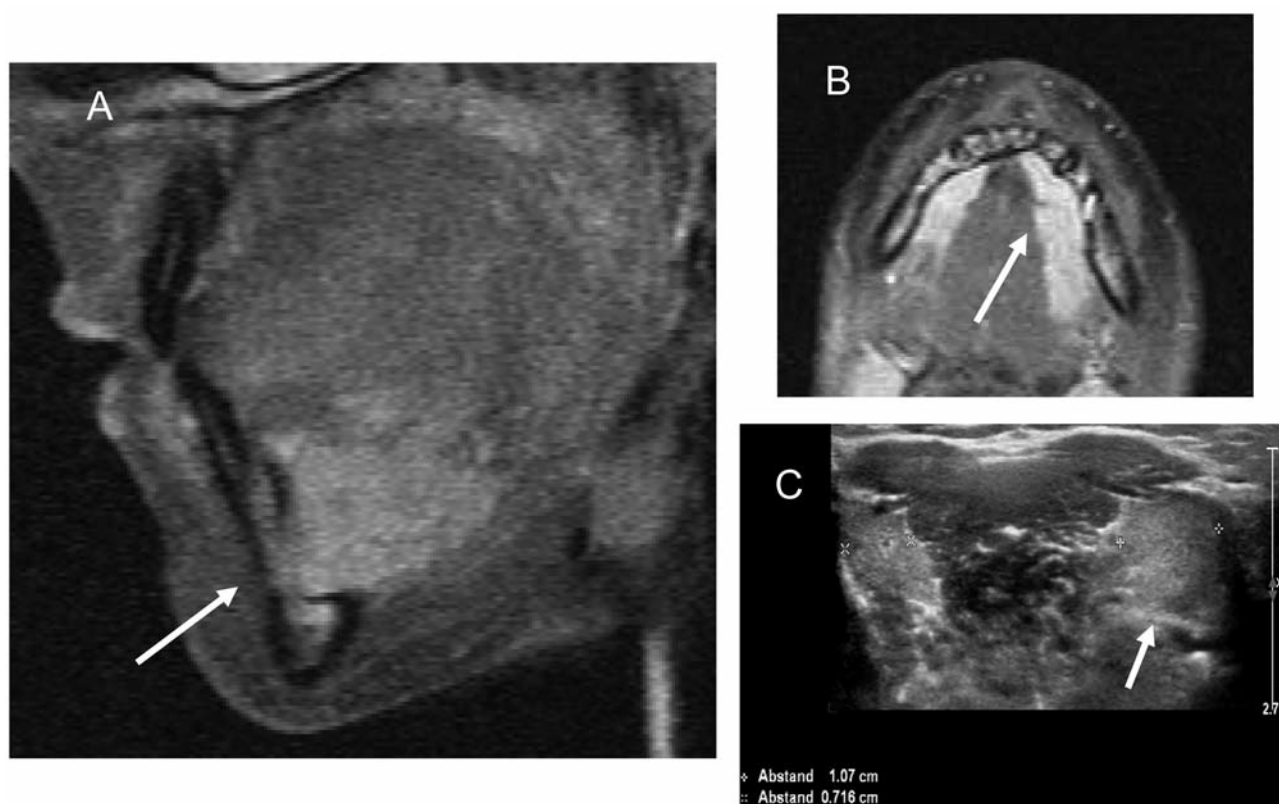


Figure 3. On magnetic resonance imaging, the sublingual gland was found to extend into the defect. A: In the sagittal view the isointense signal of the gland extends into the bone (arrow). B: The axial view delineates the glandular protrusion into the mandibular depression of the left anterior lingual mandibular margin (arrow). C: On transcutaneous ultrasound imaging of the floor of the mouth, the left sublingual gland (arrow) appears to have a slightly greater transverse diameter.

the middle-aged group (11-15). There is a strong preference for men for all locations of lingual mandibular bone defects (4). This preference for males is not linked to selection bias, which could not be ruled out in some earlier studies (11). The

male/female ratio of ALMBD was calculated to be 3:1 (4). In due consideration of the extremely rare diagnosis of ALMBD in childhood (16), several investigations were carried out in order to exclude other entities, in particular odontogenic cysts

or a tumor (17-19). Indeed, the initial presentation of the lesion on an orthopantomogram did not allow the exclusion of a neoplasm, probably of odontogenic origin. On plain radiographs, ALMBD exhibit well-defined sclerotic margins less frequent than do PLMBD (4). However, no physical finding in the area pointed to a tumor. Indeed, in rare instances, the cavity can be palpable and even becomes visible during physical investigation of the floor of the mouth (20).

Several studies have used computed-tomograms (CT) and sialography in order to clearly delineate the osseous margins and to differentiate the soft tissues occupying the lingual bone depression (21, 22). Identification of salivary gland tissue inside the cavity was frequently shown, both by various imaging techniques and following surgery, but was not present in all cases (21, 22). Furthermore, sialography is not routinely applied in these cases and in some cases the imaging of the duct system is insufficient due to technical problems (4).

Cone beam CT sialography was applied in the differential diagnosis of Stafne's cavity and yielded a clear delineation of salivary gland tissue inside the lesion (23). MRI was also used to differentiate the soft tissue inside the cavity complementary to cone beam CT (24). Even bilateral ALMBDs were revealed by this method (24), as initially described by Stafne for PLMBD (2). Indeed, MRI was recommended as a confirmatory diagnostic tool in cases suspected of having developed a Stafne's cavity (25). MRI has the advantage of being able to differentiate soft tissues in the region of interest, in particular to reveal salivary gland tissue, but to prevent x-ray exposure (24, 25). The need for identifying the soft tissue in the defect is reinforced due to reports on the existence of tumor inside similar lesions (19). On the other hand, ALMBD can be devoid of any content (26). In the present case, MRI revealed the continuity of the anterior defect's content with the sublingual gland in different planes. The fine structure of the gland, as shown on MRI, was homogenous throughout the whole organ. Thus, MRI was the main diagnostic tool in establishing the diagnosis in the present case.

Ultrasound (US) imaging has not been yet used in diagnosing ALMBD. The main advantage of US in diagnosis of head and neck pathologies is the easily achievable visualization of soft tissue lesions. Brightness modulation scan US imaging is the first modality applied to differentiate the size and structure of major salivary glands (27). In the present case, US showed a slightly enlarged left sublingual gland. The glandular structure was homogeneous and not suspicious for salivary gland neoplasms (28). However, US usually cannot transmit through bone, unless the bony surface is reduced to a thin cortical layer and the adjacent tissues contain water (29). Therefore, the complementary visualization of the whole organ by MRI was needed to complete the diagnostic work-up.

The late onset of PLMBD and ALMBD is an important argument to reject theories of pathogenesis that propose developmental defects during bone formation as the cause of mandibular bone depressions. This assumption was originally proposed by Stafne (4). The present case links the lingual mandibular defects to much earlier phases in life than usually expected. However, we were not able to define the onset of this lesion more precisely due to the fact that the present ALMBD was an incidental finding with no history of any other disease, or developmental alteration in the region of interest. ALMBD is an idiopathic bone defect that can occasionally be diagnosed in children aged 10 years or younger.

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