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Case report

A rare case of static bone cavity in the anterior mandibular region of a 10-year-old boy

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ABSTRACT

Introduction: Static bone cavity (SBC) is a bone defect that develops as a result of localized pressure from tissues surrounding the mandible. It is most commonly observed in the mandibular angle of adult males caused by the submandibular gland. The condition is asymptomatic and requires no treatment. The frequency of onset is rare, especially in the anterior mandible, and SBC is extremely difficult to diagnose in children.

Presentation of case: This report is on a case of SBC in the anterior mandible of a 10-year-old boy. The condition could not be diagnosed after panoramic radiograph and Magnetic Resonance Imaging (MRI) analyses. Computed-topography (CT) imagery revealed an oval-shaped depression 6 × 5 × 3 mm in size at the lingual apex of the mandibular left lateral tooth. Diagnosing the patient was difficult and a tumor was suspected. For treatment, the tumor-like lesion was resected and the fistula in the periosteum was sutured and closed. One year later, the defect on the lingual mandibular bone had ossified and recovered.

Conclusion: The incidence of SBC in children is rare and the condition is difficult to diagnose because of the small size of the lesions, however in this study, CT imagery proved to be useful. SBC was detected in the subject in early childhood, and his postoperative course suggested that the lesion developed as a result of glandular tissues herniating through the periosteum and causing compression on the mandible, which resulted in bone resorption. In other words, herniation of normal salivary glandular tissues were a cause of SBC.

1. Introduction

In 1942, Stafne described static bone cavity (SBC) as a pathological defect on the lingual mandibular bone filled with salivary gland tissues [1,2]. The condition was generally referred to as an asymptomatic developmental disorder detected behind the mandible of men between ages 50 and 70 [3–6]. The frequency of onset has been reported to be 0.1% to 0.48%, and is considered rare [7–8]. The only pathology is a change in the morphology of the mandible, however, no treatment is required. SBC is seldomly observed in the anterior and premolar tooth region, however when it is, a sublingual gland tumor needs to be considered as a differential disease [9,10]. Lesions that develop in such rare locations make differential diagnoses very difficult [11]. The etiology of SBC is still unknown however, some theories have been hypothesized. This Case report has been written in line with the 2020 SCARE Criteria [12].

2. Case presentation

In November 2018, CT imagery was taken of a 10-year-old boy for orthodontic evaluation and as a result, a bone defect was found on the lingual apex region of the mandibular left lateral incisor (Fig. 1). He was referred to the department of Oral Surgery in December 2018 for examination due to the suspicion that he had a neoplastic lesion in his mandible. The boy was 150 cm in height, 41 kg in weight, well-nourished, and healthy. He had no history of trauma or myofunctional habits. His face was symmetrical and no dysesthesia in the mental region was reported. There was no pain or swelling around the lesion and no abnormality in salivary flow (Fig. 2). The anterior teeth were all vital. The maxillary sinus, mandibular condyle, and the mandibular body appeared normal in the panoramic radiograph taken during his initial visit. There were no indications suggesting that the lesion was a tumor (Fig. 3A). CT imagery revealed a 6 × 5 × 3 mm oval shaped defect on the lingual cortical bone at the apex of the mandibular left lateral incisor (Fig. 3B–D). MRI revealed that the sublingual glands were in contact

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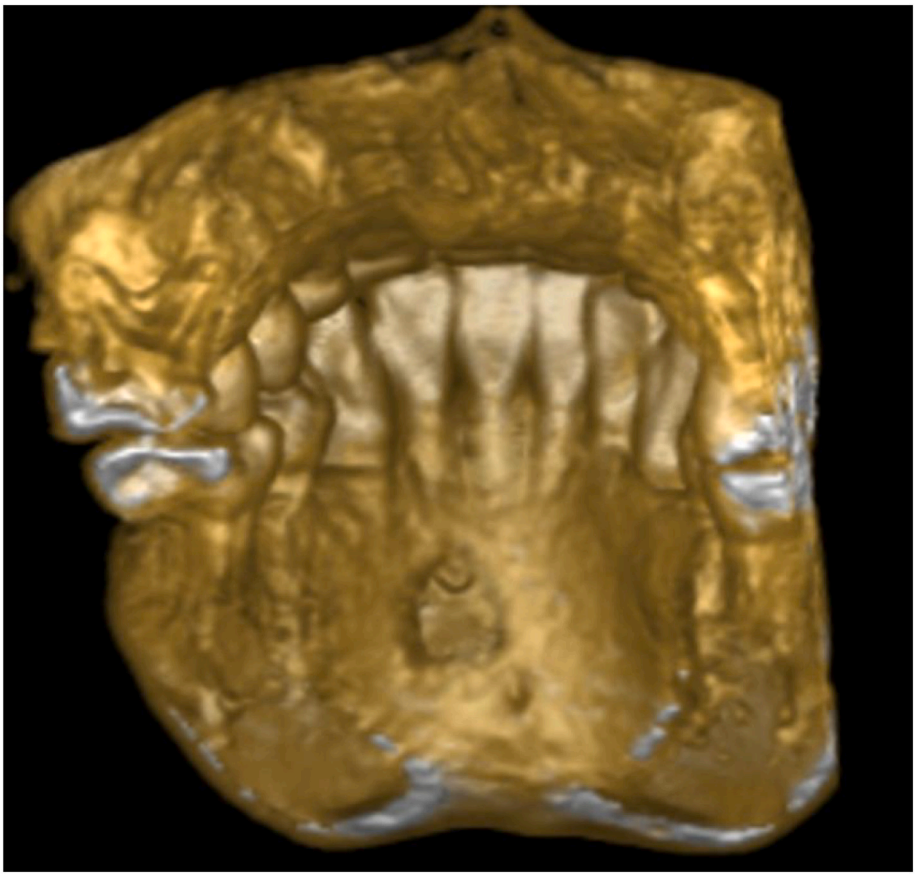

Fig. 1. A bone defect was found in the CT for orthodontic therapy evaluations on the lingual apex region of the mandibular left lateral incisor.

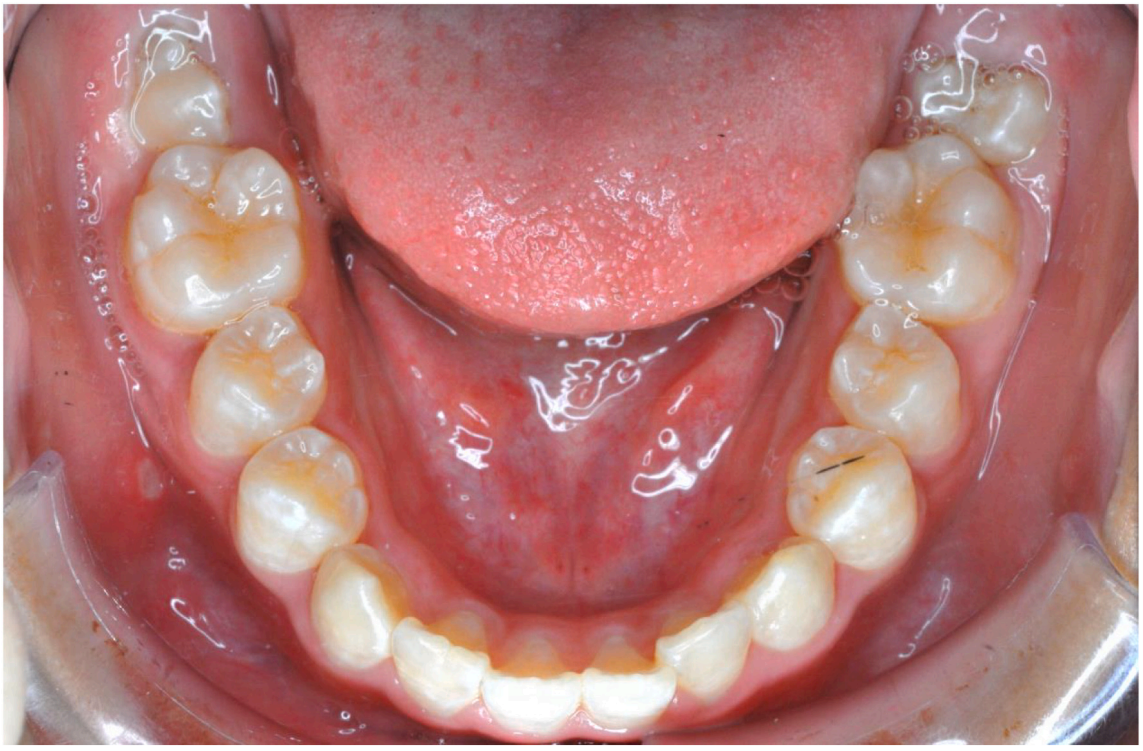

Fig. 2. There was no swelling around the lesion and no abnormality in salivary flow.

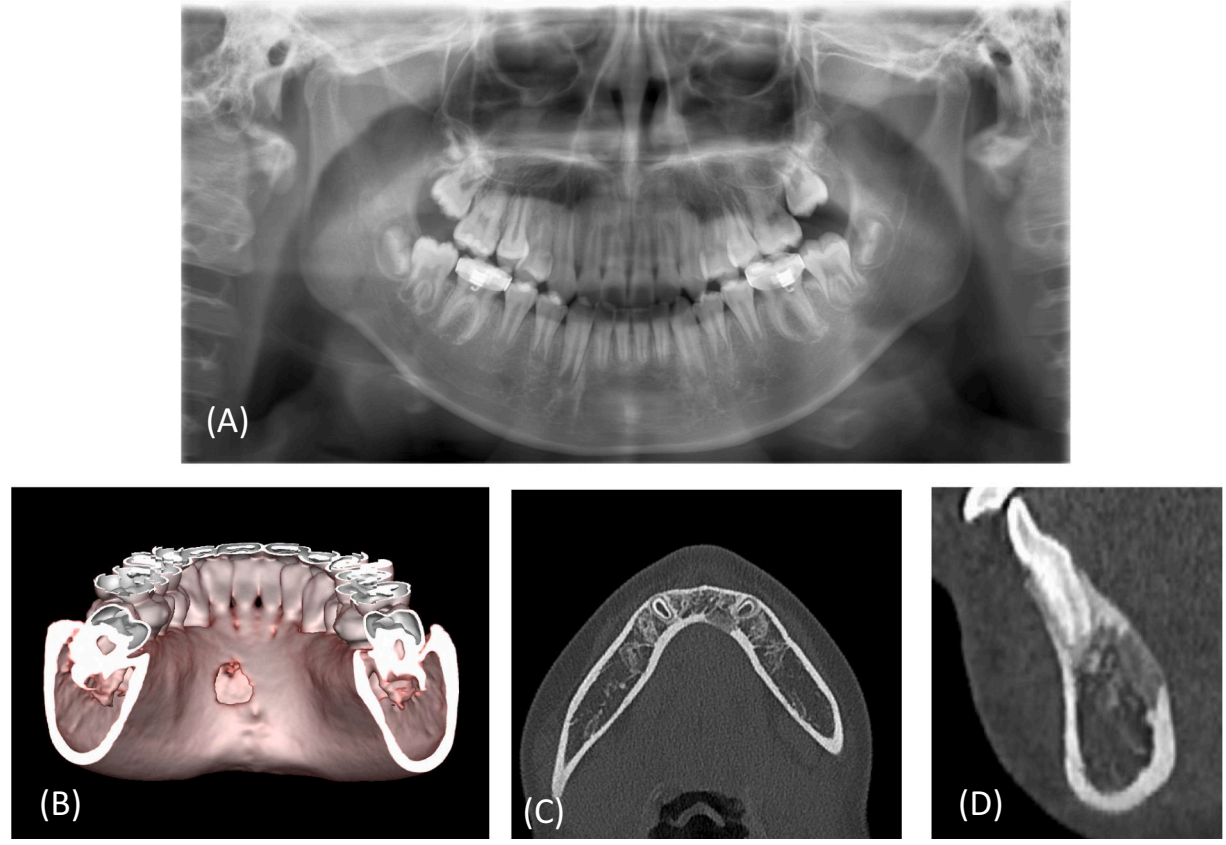

Fig. 3. (A) Panoramic radiograph taken during his initial visit. There were no indications suggesting that the lesion was a tumor. (B) 3DCT imagery revealed a 6 × 5 × 3 mm oval shaped defect on the lingual cortical bone at the apex of the mandibular left lateral incisor. (C) (D) showing a lingal cortical bone defect at left lateral incisor.

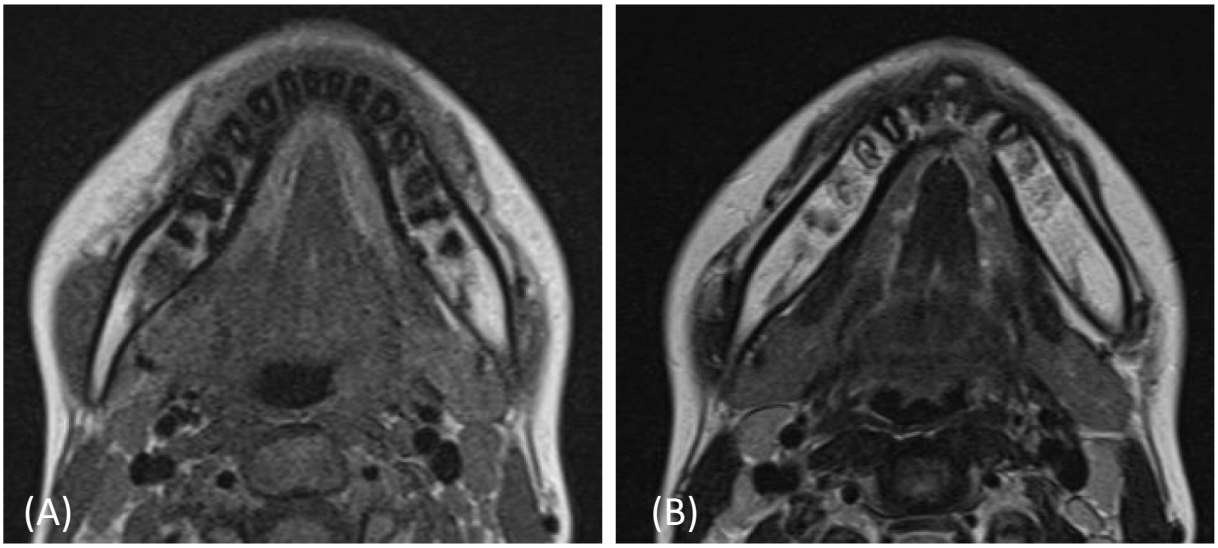

Fig. 4. (A) Axial T1-weighted image (TR/TE = 653/11 ms) (B) Axial T2-weighted image (TR/TE = 3500/100 ms) show the sublingual glands were in contact with lingual cortical bones of the lateral incisor.

with lingual cortical bones of the lateral incisor, however, the lesions were unclear. The size of the sublingual gland was normal (Fig. 4A,B). After the examinations, a neoplastic lesion on the lingual apical region of the mandibular left lateral incisor was suspected. In March 2019, the subject, while under general anesthesia, had the lesion surgically approached from the lingual side of the anterior teeth.

A periosteal flap was raised, revealing defects of the bone and periosteum on the inner surface of the mandible. Herniated tissues were found in the periosteum of the defected area. On the periosteal side, tissues that had herniated through the periosteum were observed on the inner surface of the mandible corresponding to the bone defect (Fig. 5). These tissues were elastic and soft and could easily be placed back into

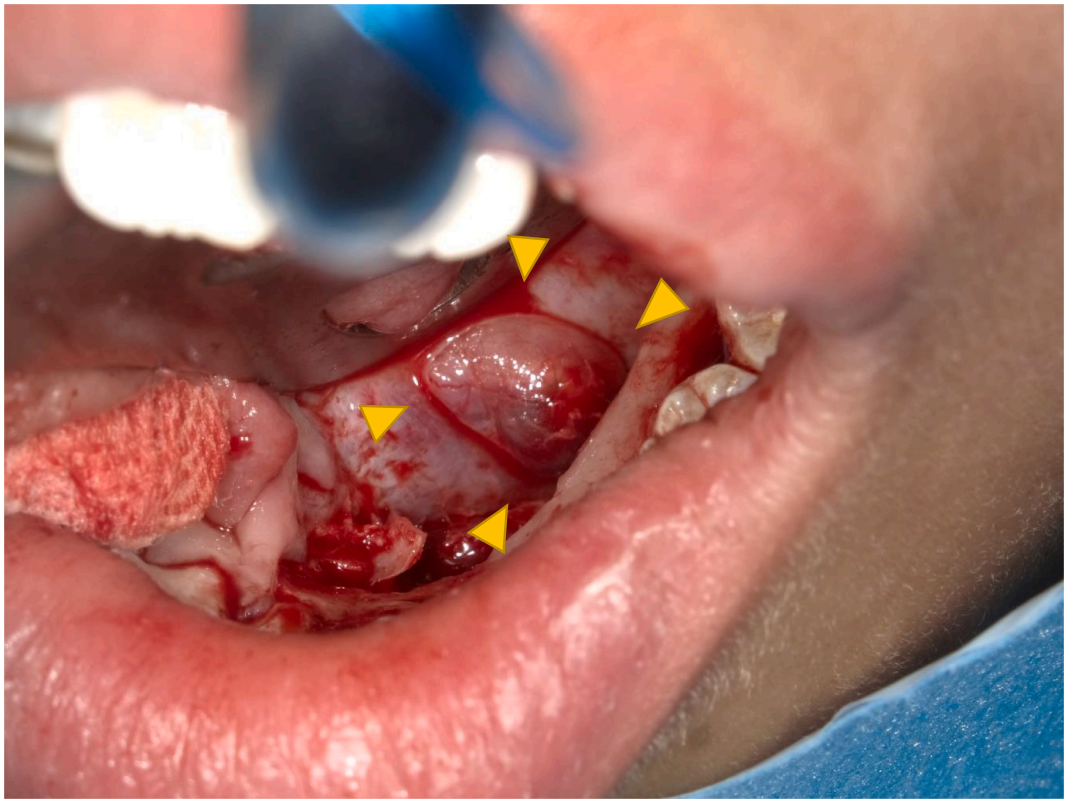

Fig. 5. Herniated tissues were found in the periosteum of the defected area.

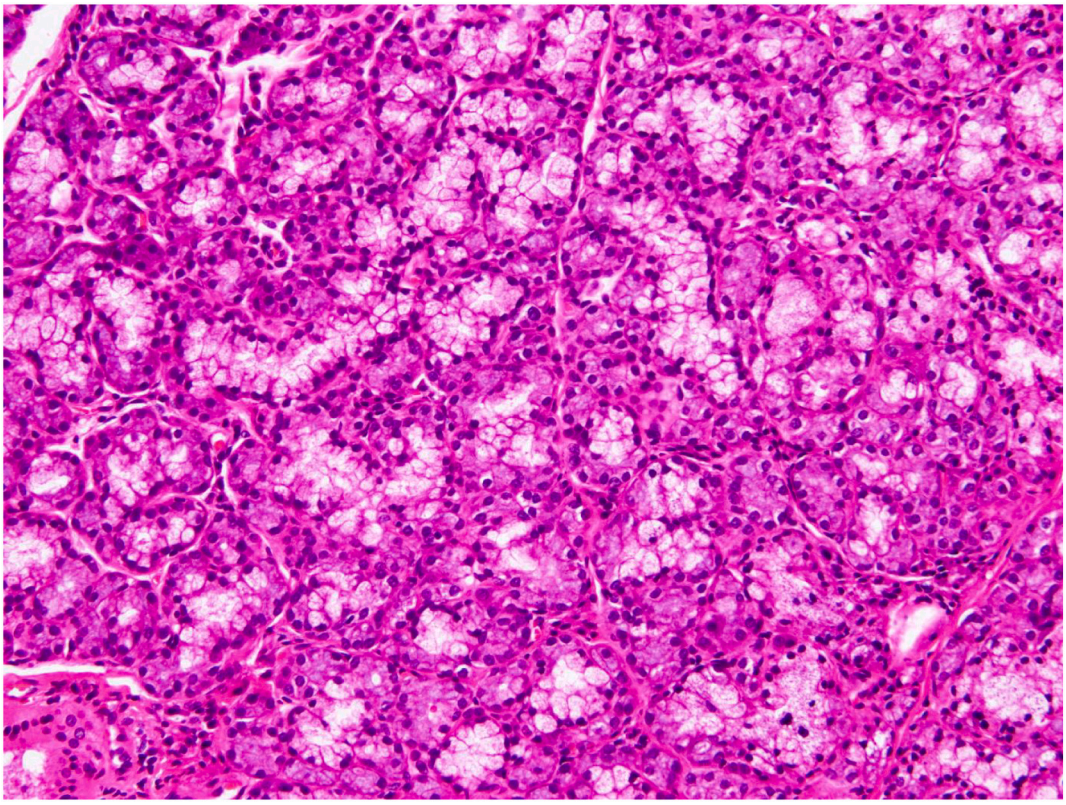

Fig. 6. Photomicrograph of the removed tissue showing normal salivary gland tissues consisting of a mixture of serous and mucous glands.

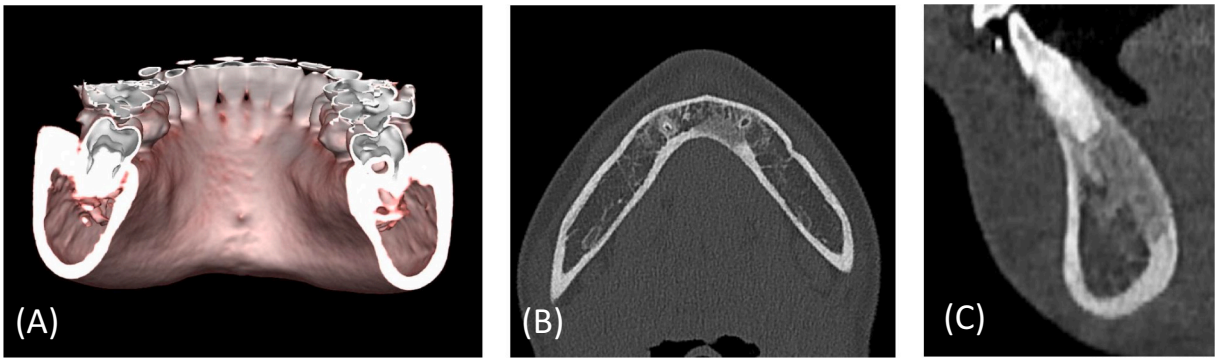

Fig. 7. (A) (B) (C) CT imagery after resection for one year which showed bone hyperplasia and disappearance of the defect.

the periosteum. The herniated tissues were excised and resected. The remaining tissues were ligated and the periosteum was sutured from the inside, with 4–0 Vicryl to close the fistula (tissue suture method). A histopathological examination was performed on the resected material. The tissues included normal salivary glands consisting of a mixture of serous and mucous glands, and no periosteum (Fig. 6). There were no neoplastic changes observed in the tissues.

The diagnosis was a SBC, and it was believed to have been caused by the sublingual gland which herniated through the periosteum and caused compression on the inner surface of the mandible, which led to bone resorption. A year later in March 2020, improvements were observed on CT imagery which showed bone hyperplasia and disappearance of the defect (Fig. 7A–C).

3. Discussion

SBC occurs when tissues surrounding the mandible such as salivary gland tissue, lymphoid tissue, connective tissue, fat, and muscles cause localized pressure on the mandible creating a bone defect [1]. Since its initial report by Stafne in 1942, SBC has been referred to by various names because its etiology and pathophysiology are unclear [13]. These cavities are generally found accidentally in panoramic radiographs, usually in men between the ages of 50 and 70 years old [4–6]. The most common site is the mandibular angle on the inner surface [3]. The frequency of onset is reported to be 0.1% to 0.48%, and it is 7 times rarer for the lesion to occur in the anterior mandible than it is the posterior mandible [14,15]. SBC that occurs in the anterior region is caused by the compression of sublingual or heterotopic salivary glands on the mandible, causing bone resorption [9,10].

The detection rate of SBC in panoramic radiographs is very rare, reported to be 0.009% [4,16]. Differential diseases of SBC include dentigerous cysts, radicular cysts, ameloblastomas, and residual cysts [17]. The legion is relatively distinguishable in the molar region, however is difficult to distinguish in the anterior region because sublingual salivary gland tumors must be considered as a differential disease [11]. Although the incidence of sublingual salivary gland tumors is low among salivary gland tumors, the rate of malignancy is high, and thus, the disease cannot be overlooked. SBC do not usually require treatment, however in the anterior region, it is necessary to consider neoplastic lesions and conduct visual inspections and histopathological examinations.

This 10-year old patient was the youngest case of SBC reported, therefore, the initial stages of this condition were investigated. The defect was observed on the orthodontic CT images however, it was too small to be detected on the panoramic radiograph and MRI. All 3 imagery methods are generally recommended for diagnosing SBC [18]. In this case, CT analysis was effective for early detection.

The sublingual gland begins developing at 8 weeks. These glands are flat and elongated in the antero-posterior direction, existing below the mucosal membrane of the floor of mouth and above the mylohyoid muscle, and are in contact with the inner surface of the mandibular body [19]. It is a mixed salivary gland tissue, consisting mostly of mucous cells with fewer serous cells [20]. Based on anatomical position and histopathological imagery, the subject's sublingual gland was diagnosed as normal.

According to Johan's Review Article, SBC occurs in all areas of the mandible and Johan suggests that the condition may be a developmental lesion [18]. In diabetes and chronic pancreatitis patients, acinar cells swell, and this process leads to morphological changes in the submandibular glands which causes them to compress the mandible [21]. Tongue habits or hand-on-chin habits can cause the sublingual glands to be compressed on the mandible, resulting in a developmental lesion that causes the glands to swell. In this case, the patient was young and showed no abnormal habits, therefore, none of the above were considered to be potential causes. The hypothesized causes of SBC include solitary cysts, developmental anomalies, pulsations of facial arteries, and compressions of hypertrophied salivary gland tissues [22–26]. The true cause is still unknown, however studies reported that 70% of SBC are filled with salivary glands, suggesting most SBC to be of glandular origin.

During the surgical procedure, it was visible that the salivary glands had separated from the periosteum and caused resorption on the inner surface of the mandibular body. The resected tissues were normal and the glandular tissues showed no pathological or morphological changes. By resecting the separated tissue and closing the fistula, the defect on the inner surface of the mandible was improved, demonstrating that resorption and invasion of the inner surface of the mandible was due to pressure by normal glandular tissue. Since the bone defect disappeared after closing the fistula, this suggested that herniated normal salivary tissues were a cause of SBC.

This case showed the initial stages of SBC. Bone formation was confirmed after resection of glandular tissues, therefore, pressure from these tissues likely caused bone resorption and herniated normal salivary glands was determined as a likely cause of SBC.

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Ethical approval

The ethical committee of the hospital of Tokyo Dental college gave agreement to report this case. The writing was performed according to the rule of anonymity.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the

written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Dr.Watanabe, Dr.Yoshida and Dr.Kato performed surgery and perioperative management. Dr. Watanabe drafted the manuscript. Prof. Takano participated in the correction of the work. Prof. Matsuzaka was responsible for pathological diagnosis.

Research registration

Not applicable.

Guarantor

Akira Watanabe is that individual who accepts full responsibility for the work and/or the conduct of the study, had access to the data, and controlled the decision to publish.

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Declaration of competing interest

The authors declare no conflicts of interest associated with this manuscript.

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