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A Connective Tissue Neoplasm of the Mandibular Angle Mimicking Stafne's Bone Cavity on Panoramic View

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Abstract. *Background: When assessing sharply delineated bone lesions of the mandibular angle on X-rays, numerous diagnoses must be considered. The static bone cavity (Stafne's bone cavity, SBC) is a harmless lingual bone depression of the mandibular angle that usually does not require any treatment. It is essential to differentiate this bone deformity from other lesions that may require treatment. Case Report: The 22-year-old patient was referred for further diagnosis and therapy after osteolysis of the mandible was noticed on a panoramic view (PV). The location and size of the lesion was typical of SBC. Only the three-dimensional representation of the lesion on cone beam computed tomographs revealed an intraosseous lesion. Histological examination of the lesion provided evidence of a fibrous neoplasm. Conclusion: The typical image of SBC is ambiguous on plain radiographs such as PV. The radiological diagnosis of the lesion should be based on the representation of the region of interest in different planes.*

On standard X-rays of the jaw, distinct alterations of the mandibular angle are relatively often diagnosed as being radiotranslucent lesions. However, similar lesions on radiographs may comprise different entities (1). The nosological spectrum of radiotranslucent lesions ranges from osseous neoplasm and infections to anomalies in bone structure and shape (2). Stafne's bone cavity (SBC), a radiotranslucent lesion limited in size and topography, is viewed as a harmless variant of mandibular lesions at this site (3). SBC refers to a lingual bone depression in the region of the mandibular angle typically located below the mandibular canal and does not require treatment (4). Sexual dimorphism of SBC in favor of the male sex is considerable (5). Furthermore, diagnosis of SBC is predominantly based on plain radiographs (5). The lesion is not diagnosed radiologically until later in life (3). Well-founded doubts have been expressed assessing the lesion as a developmental disorder in the formation of the mandible (6). The present report supports the assessment that SBC diagnosis should preferably be based on X-ray examinations presenting the region of interest in several dimensions since other lesions requiring treatment may appear like SBC on plain radiographs.

Case Report

The 22-year-old patient was referred to the outpatient clinic of the Department of Oral and Maxillofacial surgery for further diagnostics and treatment of the incidental finding of a jaw lesion. The otherwise healthy male patient was undergoing dental treatment, where a panoramic view (PV) was obtained, showing a lesion in the region of the left angle of the lower jaw (Figure 1).

On admission, the external findings of the facial skin were normal, as was the oral mucosa. There were no sensitivity disorders of the trigeminal nerve and the teeth responded adequately to cold stimuli. Based on PV, it was initially assumed that the lesion was SBC because the location (anterior to mandibular angle, below mandibular canal) and size (oval, sharply demarcated osteolysis) of the lesion corresponded to the radiological criteria of the entity. Although the patient's gender was typical for SBC, his age at the time of diagnosis was well below the expected age. A cone-beam computed tomography (CBCT) was made for advanced diagnostics

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Figure 1. *Panoramic view (PV) of the patient depicts a well-demarcated radiotranslucency of the left mandibular angle. The lesion is situated below the mandibular canal.*

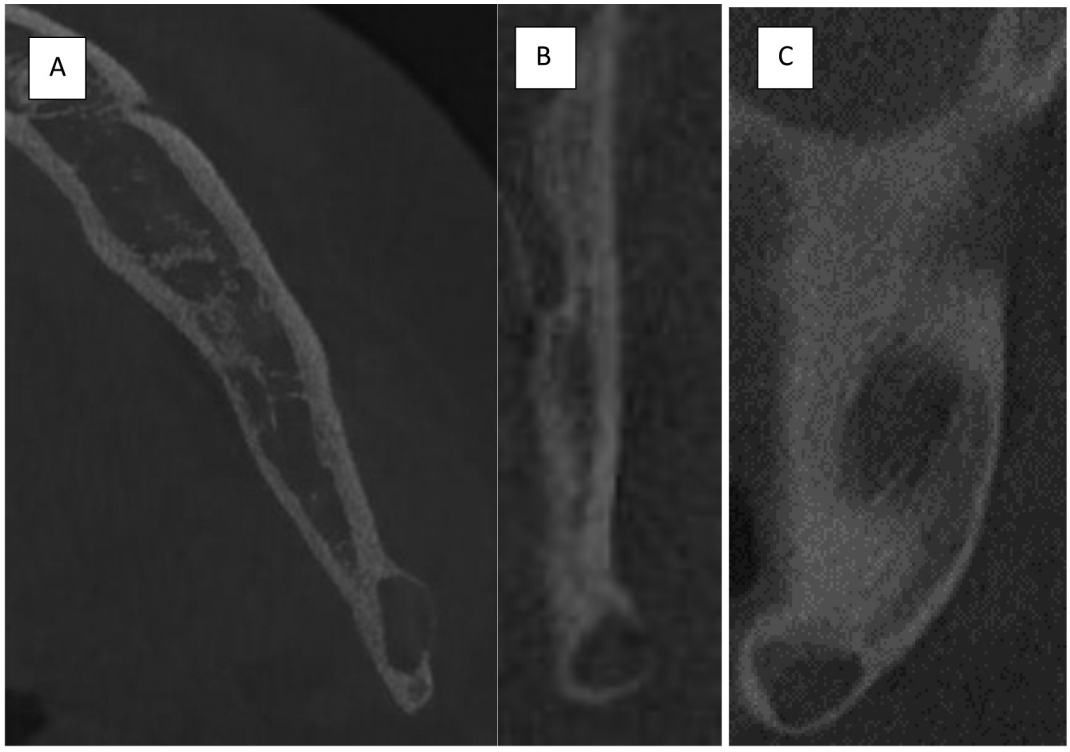

Figure 2. *Cone beam computed tomography (CBCT) of the left side of the mandible (cropped images). A) Axial plane. The lingual side of the lower jaw is smoothly delimited to the soft tissues of the floor of the mouth and has not developed any osseous depression in the region of the strictly osseous lesion. Vestibular and lingual cortical bone layers confluent anteriorly to the lesion. The lesion is radiotranslucent. The lesion bulges in the vestibular direction and covered by a thin layer of bone. B) Coronal section. Lesion protrudes in vestibular direction. The basal cortical layer is intact. C) Lesion on sagittal plane.*

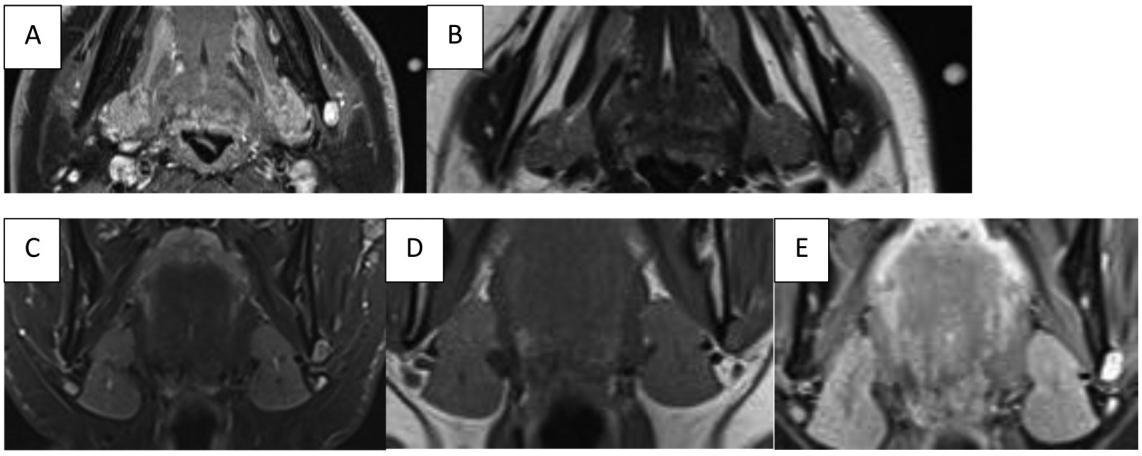

Figure 3. *Magnetic resonance images (MRI) of the left mandibular lesion. A) Axial section. Hyperintense lesion of oval shape in left mandibular angle [T1 fat suppressed Dixon method (FS-Dixon)]. B) Axial section. Incomplete, narrow hyperintense margin of the lesion. This narrow hyperintense border is marked more clearly on the T2-weighted coronal image depicted in C), whereas the lesion is homogeneously isointense to muscle on T2 weighted turbo-spin echo (TSE) coronal image shown in D). E) Coronal image of the lesion on TSE (FS-Dixon) weighted image.*

(Figure 2). This X-ray showed a rounded intraosseous lesion. The lingual cortical bone of the mandible was not depressed in the region of the lesion, as expected in SBC. Rather, the homogeneously soft tissue-dense lesion, which was completely surrounded by bone, protruding in vestibular direction (Figure 2). On the vestibular side, the cortical layer of the bone appeared to be considerably thinned. The CBCT visualization of the lesion made an intraosseous lesion very likely. Magnetic resonance imaging (MRI) of the head and neck region was performed to adequately display the surrounding soft tissue. MRI confirmed a hyperintense oval lesion limited to the lower edge of the mandible, with a sharply defined border and no adjacent soft tissue infiltration (Figure 3).

Based on these findings, it was decided to explore the lesion. Under general anesthesia, the vestibular side of the bone was exposed following a mucoperiosteal incision. The cortical bone was smooth and evenly developed, except for the small protuberance located in the angle of the jaw and close to the lower edge of the bone (Figure 4). The bony protuberance was removed with a chisel and bone cavity exposed. The soft tissue contents of the bone cavity were excised and the oral soft tissue wound closed with sutures by primary intention. The postoperative course of the patient was uneventful.

Figure 4. *Exposed surgical site: the dome-shaped bone lesion consists of intact cortical bone (located on the left of the inserted hook).*

Histology. The histological examination revealed a fibro-osseous lesion composed of hypercellular fibroblastic stroma and trabeculae of woven bone. The nuclei of the fibroblasts showed characteristic hyperchromatic nuclei without atypia. Some areas presented prominent osteoblastic rimming. Cementum-like tissue was not detectable. However, the amount of tissue types may vary considerably in ossifying fibroma. Epithelial structures such as salivary gland tissue, oral mucosa, or soft tissue without any bone formation that could indicate entrapment, such as that seen in SBC, were not evident (Figure 5A and B).

Discussion

This case report shows a mandibular lesion which, when viewed from a PV, meets the diagnostic criteria of SBC. Thus, diagnosis

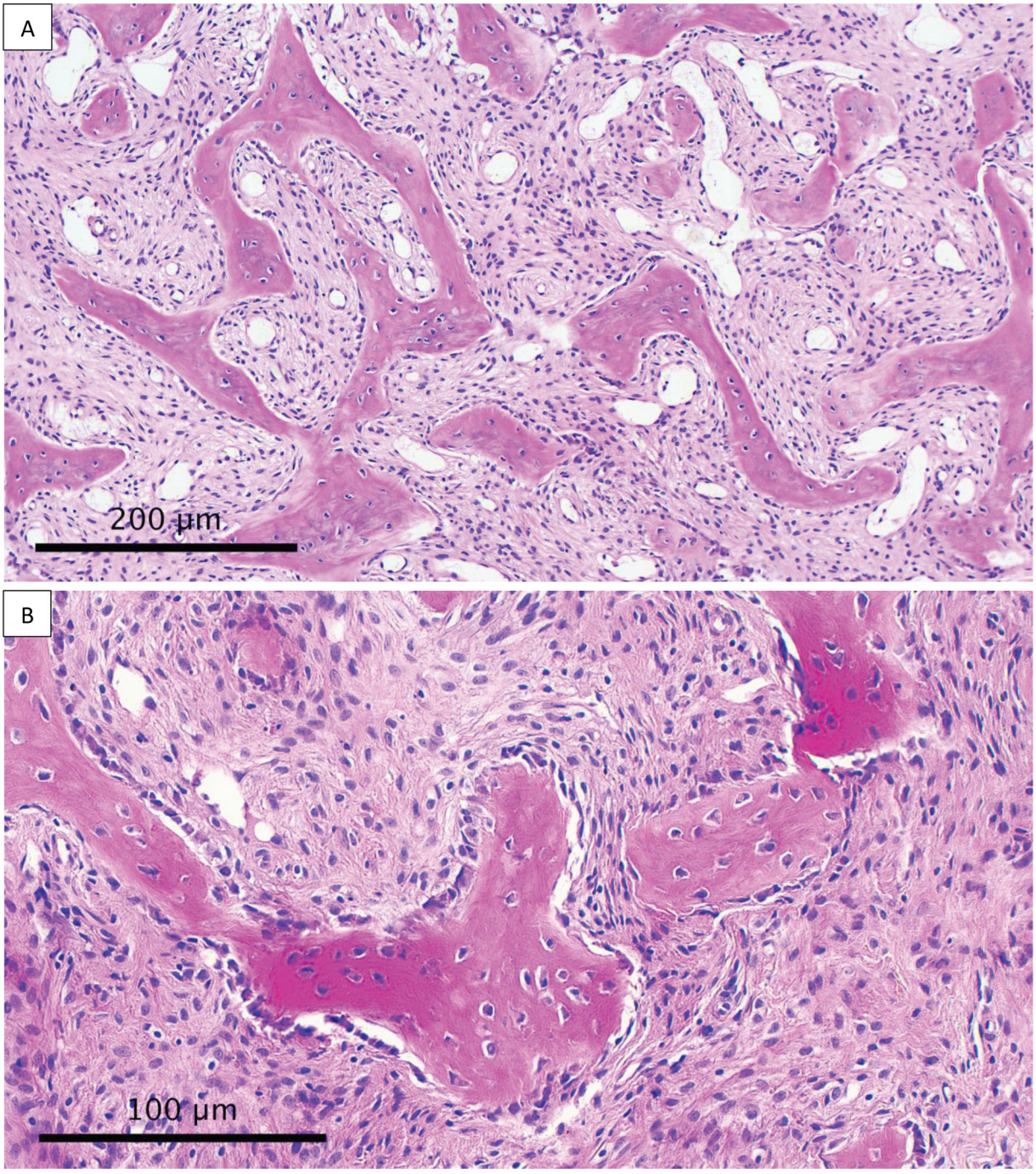

Figure 5. *Ossifying fibroma of the mandible. A) Fibro-osseous tumor composed of spindle cell-rich stroma and woven bone. B) Areas with prominent osteoblastic rimming. (HE, EDTA-decalcified).*

based on PV would have justified the assessment that the lesion is a bone change not requiring treatment. Only the supplementary three-dimensional radiography revealed an intraosseous lesion indicating the need for surgical intervention. Reliance on a sufficiently precise SBC diagnosis using plain radiography like PV is no longer justified. Alternatively, three-dimensional sectional imaging is the preferred imaging modality to establish a lesion's adequate topographical representation (7).

SBC. The eponymous name SBC gives credit to the author who described the mandibular anomaly on plain radiographs (1-4). The cup-like depression on the lingual side of the mandible has since been described in several radiological studies (4, 5). The localization in the angle of the jaw is the most common manifestation of SBC and the initial description of the lesion has focused entirely on this region (3). However, systematic analyzes of published studies, case series, and individual reports have shown that such bone depressions can occur in many mandibular sites (8-15). The etiology of the lesion is unknown. The retention of the eponymous definition of the entity in medical terminology may also be due to the lack of a coherent pathogenetic concept that explains the numerous variants of the phenomenon. Some authors prefer the theory of local bone erosion due to pressure from the neighboring salivary glands as the cause of SBC (16). However, some authors have pointed out that no salivary gland tissue can be detected in the lesions (7). Rather, the lesion is filled with adipose tissue, blood vessels, *etc*. (6, 7, 14). The soft tissue of the osseous lesion in this examination confirms the exclusion of salivary gland tissue. Blood vessels and connective tissue were essential components of the lesion mixed up by foci of ossification.

In order to assess the significance of the case presented here, it is crucial to consider that the vast majority of studies on SBC are based on plain radiographs (17). In fact, it is characteristic of SBC that, since it's definition (3), diagnosis has primarily been based on radiological findings depicting the region of interest in two dimensions (1). Until a few years ago, the number of systematic studies on SBC was relatively small (11). Interest in the radiological diagnosis of SBC has increased in recent years, probably due to improved skeletal diagnosis of the facial skull using CBCT (10). Atypical SBCs were identified using CBCT, for example bi- or trilobated types, but also SBC-like lesions of the mandible in almost all locations of the mandible (11, 13). Bicortical lesions (15), lesions where the lower edge of the jaw is dissolved (14), as well as multilocular manifestations, are counted among the variants of SBC (12). The lesion can also be detected in patients with tumor predisposition syndrome (18).

The shape of the lesion can differ significantly from the common oval lesion running below the canal (13). Unilateral and lingual depression is by far the most common variant (3), but vestibular depressions in the cortical bone are also counted among the variants of SBC by some authors (4, 6). On the other hand, the cortical impression on one side can be associated with expansion on the other side and thus, dictate the skeletal finding of suspected pressure atrophy and opposite hypertrophy of the bone (19). However, a constitutive feature of SBC is the impression of the bone from the outside. Strictly intra-osseous lesions are not included in the category 'SBC'. The case presented here can therefore be ruled out as a case of SBC by the definition referring to X-ray imaging, but only after plain radiography has been supplemented by cross sectional imaging.

Complementary to the identification of a very variable SBC phenotype, the demands are increasing on the reliability of its initial radiological diagnosis (20, 21). The present case shows that SBC cannot be reliably diagnosed using PV. In the individual case of radiological diagnostics of jaw lesions as well as in the systematic examination for SBC in radiological studies, the findings should refer to cross sectional images. Adequate diagnosis of radiotranslucency is crucial because neoplasms can take the shape of SBC and even develop at typical mandibular locations (20, 21). The presented case exemplifies the importance of this requirement for the current diagnosis of jaw lesions. Information on the prevalence of SBC with reference to the analysis of PV is worth exploring according to the presented case (5, 17).

Conclusion

SBC is a radiological diagnosis made by exclusion, for which the representation of the region of interest on cross-sectional images is necessary.

Conflicts of Interest

The Authors have no conflicts of interest regarding the work presented.

Authors' Contributions

Diagnosis of patient: all Authors; treatment of patient: REF, SJC; drafting the manuscript: REF; final approval of the manuscript: all Authors.

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