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Case report

Enlargement of a Stafne cyst as an indication for surgical treatment – A case report

Christopher Prechtl*, Philipp Stockmann, Friedrich Wilhelm Neukam, Karl Andreas Schlegel

Department of Oral and Maxillofacial Surgery (Head: Prof. Dr. Dr. Dr. h. c. Friedrich Wilhelm Neukam), Friedrich-Alexander-University Erlangen-Nuremberg, Glückstraße 11, Erlangen, Germany

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ABSTRACT

Stafne cysts are often defined as static lesions located in the angle of the mandible. Consequently many authors have proposed that there is no need for surgical treatment of these bony defects on the lingual side of the mandible. This article describes the case of a 55-year-old patient, in whom a Stafne cyst showed a significant enlargement, reaching a size that necessitated surgical intervention because of the risk of pathological fracture. A literature search showed 5 additional similar cases, where progression in the size of a Stafne cyst could be radiographically documented. Consequently, the recommended management of these pseudocysts should be reconsidered.

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1. Introduction

Cysts are common pathologies of the jaw bone. Because of their capacity to enlarge steadily over time, in most cases the complete removal of the cyst is adequate treatment. It may be necessary to fill the remaining osseous defect with autogenic bone transplants or bone substitution materials after enucleation of the cyst, while primary closure has been recently reported to be state of the art in most cases (Ettl et al., 2011).

Radicular cysts and follicular cysts belong to the group of the odontogenic cysts and are the commonest, whilst other cysts and pseudocysts like keratocysts, aneurysmal bone cysts or solitary bone cysts represent only a small number of the cystic pathologies of the jaw bone (Pitak-Arnnop et al., 2010; Strabbing et al., 2011; Zadik et al., 2011; Triantafillidou et al., 2012). As the differential diagnoses for such cystic structures in panoramic tomograms are, for example, arterio-venous malformations, the ameloblastoma, the ameloblastic carcinoma or the squamous cell carcinoma arising in dentigerous cysts, these rare cystic changes to the jaw can be a diagnostic and therapeutic challenge (Siniscalchi et al., 2009; Colbert et al., 2012; Pirklbauer et al., 2012).

Stafne bone cysts are defined as pseudocysts of the jaw (Stafne, 1942, 1947; Uil, 1971; Collett, 1986; Davis, 1994). They do not contain an epithelial lining and usually occur in the jaw angle below the mandibular nerve. Less frequently, they are diagnosed in

other locations like in the anterior mandible (Malkin and Berg, 1974; Apruzzese and Longoni, 1999; Katz et al., 2001; de Courten et al., 2002; Queiroz et al., 2004; Krafft et al., 2010; Sisman et al., 2010; Turkoglu and Orhan, 2010). Sometimes the defect area includes the lower border of the jaw. The terms "static bone defect", "Stafne defect", "lingual mandibular bone cavity" and "idiopathic bone cavity" are commonly used as synonyms for this bony defect. In panoramic tomography this structural change of the bone appears to be a well-circumscribed radiolucency with a sclerotic border. Usually, there is no connection with dentoalveolar pathology. In most cases, the Stafne cavity is diagnosed as an incidental asymptomatic finding in routine radiological examinations. In the literature, Stafne cysts are described as structural changes of the mandible, which remain stable in size. In conclusion it has been stated that this cyst does not need to be removed (Richard and Ziskind, 1957).

Pathogenesis

The pathogenesis of this pseudocyst of the jaw still is obscure. There are two major theories for the development of these cysts:

1. Stafne cysts represent a developmental anomaly. They may be caused by a congenital inclusion of the submandibular gland during the development of the mandible (Choukas and Toto, 1960). Another possible reason is deficient bone deposition in an area which had originally filled by Meckel's cartilage (Stafne, 1942). This would explain the frequently reported static

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^{*} Corresponding author. Tel.: +49 9131 85 33627; fax: +49 9131 85 34106. *E-mail address*: Christopher.Prechtl@uk-erlangen.de (C. Prechtl).

properties of Stafne cysts, which are often seen in clinical and radiological examinations.

2. Stafne cysts are active and progressive processes. The hypothesis most frequently used is that the bone defect on the lingual side of the jaw is a result of pressure atrophy of the submandibular gland (Choukas and Toto, 1960). This theory is affirmed by the postoperative histologic findings, which often indicate the presence of seromucous salivary gland tissue, suggestive of the presence of the submandibular gland in the defect. Furthermore, the literature supports bone remodelling due to functional loading as a reason for the development of Stafne cavities (Menzel, 1968). There is also the hypothesis of local bone resorption by microtraumata caused by the pulsation of the facial artery (Kay, 1974). Other authors discuss aneurysms, remains of solitary bone cysts and eosinophilic granulomas as reasons for the development of this pseudocyst (Rushton and Cantab, 1946; Uemura et al., 1976).

2. Case report

In 2004, a well defined cystic lesion at the angle of the mandible, below the mandibular nerve, of a 55-year-old patient was seen on panoramic tomography (Fig. 2). In a previous radiological examination in 1994, there was only a discreet round translucency (Fig. 1). As these radiographic findings were consistent with a Stafne bone cyst, no operative treatment was conducted and the lesion was monitored. In January 2011, a panoramic tomogram showed a significant enlargement of the bony defect (Fig. 3), and the patient was consequently referred to our clinic.



Fig. 1. Panoramic tomography (09/1994): a discreet translucency can be seen in the left jaw angle below the canalis mandibularis (arrow).



Fig. 3. Panoramic tomography (01/2011): further enlargement of the bony lesion (arrow).

Examination did not reveal any dentoalveolar pathology associated with the radiographical findings. The patient reported that he had no complaints. Because of the enlargement of the lesion and the critical size that could have led to a pathological fracture, we decided on a biopsy, histopathological examination of the lesion and an augmentation and stabilization of the resultant defect.

In February 2011, the area was operated on under general anaesthesia. Preoperative three-dimensional imaging of the cyst was performed (digital volume tomography; Fig. 4). An extraoral incision was chosen and skin and periosteum were elevated from the lower jaw. The cystic lesion was identified at the angle of the mandible. Macroscopically, no epithelial lining could be seen. A space-occupying lesion was removed from the defect in the mandible. The tissue showed a connection to the inferior part of the submandibular gland. The excess tissue was removed and sent for histopathological examination. The inferior alveolar nerve was identified and not damaged in the superior part of the lesion. After adequate freshening of the wound edges, the defect was filled with autogenous bone from the anterior iliac crest. The bone block transplant was fixed in place using an osteosynthesis plate and covered by a resorbable BioGide[®]-membrane (Geistlich Biomaterials Vertriebsgesellschaft mbH, Baden-Baden, Germany) (Fig. 5). After the insertion of a drainage, the wound was closed using resorbable sutures. The postoperative period was uneventful.

The histopathological examination showed seromucous salivary gland tissue in the removed mass. There were no signs of inflammation or malignity. Therefore, the results were consistent with the diagnosis of a Stafne cavity.

In October 2011, the osteosynthesis plate was removed. Clinically, good bony regeneration of the autogenous bone graft was observed (Figs. 6 and 7). The wound healing was again uneventful.



Fig. 2. Panoramic tomography (11/2004): cystic translucency in the left jaw angle, matching the typical finding of a Stafne cyst (arrow).



Fig. 4. Digital volume tomography (02/2011): preoperative imaging.

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