CASE REPORT

An Unusual Presentation of Stafne Bone Cyst

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Abstract Stafne bone cyst is a rare mandibular defect. It is a developmental abnormality that commonly presents as a small, well demarcated, and asymptomatic radiolucency seen at the angle of the mandible below the mandibular canal. There are usually no clinical signs nor symptoms. Diagnosis is commonly by plain X-ray, but use of more accurate imaging such as MRI is required in atypical cases. This case study reports an unusual presentation of Stafne bone cyst in the ramus of the mandible in a young man and reviews the relevant literature.

Keywords Stafne bone cyst · Mandibular · Defect

Introduction

In 1942, Stafne reported 35 asymptomatic, well-circumscribed, round or ovoid radiolucency near the angle of the mandible based on radiographic findings [1]. He followed literature of five of the defects for 5–11 years and did not notice any change in the size or characteristics of the cavities. The cavity was found to be a depression in the lingual cortical surface of the mandible, indented by normal salivary gland tissue. Such defects had since been given a clinical entity. Other terms that have been used to describe this entity include static bone defect, lingual mandibular bone cavity, latent bone cavity, idiopathic bone

cavity, ectopic submandibular or sublingual gland in the mandible, and lingual cortical mandibular defects [2].

Epidemiological data has shown an increased incidence of Stafne bone cyst in the middle aged males [2]. The prevalence of Stafne bone cyst in published series has been reported to be from 0.10 [3] to 0.48 % [2]. However the incidence is much lower for bone cavity in the ramus of the mandible.

No treatment is required for Stafne bone cyst, although there has been report of surgical exploration of the cavity [4]. Confirmatory tests including sialography, computed tomography scan (CT), or magnetic resonance imaging (MRI) have been used. However, periodic clinical examination and plain radiograph are considered suffice to monitor vast majority of the lesions.

Stafne bone cyst can present with different anatomical variations. There are reports of bony cavity with extra-osseous course of the mandibular neurovascular bundle [5], with buccal expansion [6], in young persons [7], above the neurovascular bundle, in the anterior region of the mandible [8] and as bilocular radiolucency [4].

Case Report

A 30 year old man was referred by his local medical officer to the Oral and Maxillofacial Surgery Unit at Barnet Hospital for his right sided facial swelling (Figs. 1, 2). The swelling had persisted over last 6 months, with no change in size and no symptoms of pain, purulent discharge nor systemic features of infection and malignancy. On clinical examination, the swelling was not consistent with lymphadenopathy nor a parotid gland swelling.

Incidentally, the patient presented with facial asymmetry which was queried to be congenital. The facial asymmetry consisted of hypoplasia of the upper left face and

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Fig. 1 Clinical photo demonstrating asymmetry

Fig. 2 Frontal view of the patient showing facial asymmetry

hypoplasia of the lower right face and a chin deviation to the right side. The patient did not express any complaint, apart from the obvious deformity of his mandible. Medically, the patient had mild growth delay with resultant short stature. There is a family history of neurofibromatosis but this was not expressed in the patient.

Plain radiograph showed a radiolucent lesion in the posterior area of the right angle of the mandible and in the ramus (Fig. 3). The condyle on the right side appeared to be rudimentary. Ultrasound did not reveal evidence of focal lesion and no obvious lymphadenopathy.

Further investigations were undertaken. On the CT scan, the radiolucency was represented as a depression and lack of cortical surface in the angle and ramus of the mandible (Fig. 4). The parotid gland was enlarged but the MRI and ultrasound did not show any evidence of focal lesion in the gland. The MRI did show a 1 cm lymph node anterior to the superficial lobe of the right parotid and also extension of the

Fig. 3 Plain radiograph showing a radiolucent lesion in the posterior area of the right angle of the mandible and in the ramus

superficial lobe of the right parotid along the ramus of the mandible with enhancement and high signal on T2 (Fig. 5). The deformity was dramatically illustrated in the three-dimensional reconstruction (Fig. 6). Ultrasound guided fine needle aspiration confirmed normal glandular tissue.

The diagnosis of the facial deformity was mild hemifacial microsomia (type 1) due to hypoplasia of the right side of the mandible and TMJ. The bone resorption and atrophy was a direct result of Stafne bone cavity. The patient has been under long term review with no surgical intervention deemed necessary.

Discussion

Stafne bone cyst is very rarely encountered at ramus of the mandible. In one study, which looked at 2,693 orthopantomographs, there was a total of 13 lingual cortical mandibular defects, but none were located in the ramus [2]. Reports of ramus bone cavity in the literature is rare: there are currently two case reports from literature search [9, 10]. A large review of literature of mandibular bone depression showed that the incidence of Stafne bone cyst in mandibular ramus was <2.5 % of all bone depressions (14 cases out of 583) [11].

There has been debate as to whether the defect is congenital or developmental in nature. Stafne suggested that the bone cavities are congenital and are caused by a failure of fusion in the areas formerly occupied by Meckel's cartilage [1]. However, the location of the lesion relative to the major salivary gland, the findings of histologic content and the absence of lesion in new born point to a close relationship between the defect and the salivary glands. The general consensus on the pathogenesis of Stafne bone cyst is the "glandular" hypothesis where a hyperplastic or hypertrophic lobe of the submandibular, sublingual or

Fig. 4 CT scan showing the radiolucency represented as a depression and lack of cortical surface in the angle and ramus of the mandible

Fig. 5 MRI showing lymph node anterior to the superficial lobe of the right parotid and also extension of the superficial lobe of the right parotid along the ramus of the mandible

parotid salivary gland exerts pressure on the lingual or buccal cortex of the mandibular body or ramus to create a focal bony resorption with resultant bone cavity [11].

Similar to bone cavity in other regions of the mandible, ramus cavity is usually diagnosed after middle age in men. In a literature survey of six clinical cases, all males, the age

Fig. 6 Three-dimensional reconstruction of the deformity

of diagnosis ranged from 36 to 60 years, with a mean age of 48 years [11]. Another study involved five men between the age of 36–57 years, with a mean of 46 years [10]. There is report of Stafne bone cavity in young persons [7], but none reported in the ramus.

Investigation usually involves plain radiograph as the lesion is typically an ovoid, unilocular radiolucency. The margin is generally well demarcated and sclerotic. Sialography had been used in the past to detect extend of the parotid gland into the defect. Seward found the presence of salivary gland inclusion in a bone cavity in 1960 [12]. However, subsequent sialograph use has given conflicting results. Wolf reported five cases of ramus defects in which one case had changes suggestive of chronic sialoadenitis and the other four cases had normal sialogram of the parotid gland [13].

No treatment is required for this condition. Surgical exploration to establish diagnosis is possible. However, as the lesion is asymptomatic and non-progressive, surgical intervention is not indicated in majority of cases. Periodic radiographic follow up is generally agreed to be the best long term option. The use of computed tomography (CT) scanning and magnetic resonance imaging (MRI) is important to investigate the atypical lesion and to differentiate the defect from a centrally occurring lesion [14, 15]. Buccal expansion is rarely present, which is another clinical clue to differentiate the defect from a central lesion. However, there has been study of lingual mandibular bone defect presenting with buccal expansion [6]. In such case, an incisional biopsy may be indicated to prevent unnecessary radical treatment. When surgical exploration is undertaken, salivary tissue is rarely found. However, there has been report of salivary gland inclusion in the bone cavity [16].

Stafne bone cavity at the ramus is a rare clinical finding. It needs to be differentiated by clinical and radiographic

examination from lesions of the bone such as traumatic bone cyst and ameloblastoma, lesions of the salivary gland such as pleomorphic adenoma, or lesions of the adjacent structures such as lymphadenopathy of different aetiologies. When there are associated symptoms, surgical exploration and biopsy may be undertaken. Asymptomatic bone cavity should be monitored clinically and radiographically on regular basis. Any abnormal change or abnormal initial presentation warrant more advanced imaging techniques such as CT or MRI.

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