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REVIEW ARTICLE

Stafne's bone cyst revisited and renamed: the benign mandibular concavity

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Objective: Stafne's bone cyst (SBC) is an asymptomatic, round or oval-shaped, well-defined, uniform radiolucent lesion, usually incidentally observed in the posterior aspects of the mandible. Radiographical appearance may be confusing though. This study aimed to review the literature on SBCs and to map the SBC as shown in their respective papers.
Methods: A Pubmed® search (1/9/2018 till 31/5/2019), mentioning SBC, was carried out. Included papers had to contain: patient's age, gender and radiographs.
Results: In total, 114 papers were found, but only 64 papers were retained, which contained 109 cases (95 males, 14 females). The patients' ages ranged between 14 and 89 years old (mean age being 52 years). Male patients' ages (N = 95) ranged from 14 to 89 years (mean age 52.3 years), whereas the females (N = 14) ages ranged between 22 and 68 (mean age 50.2 years). 28 combinations of locations of SBC were recorded. Only three lesions were located at the symphysis, six were bilateral, 55 appeared on the left hand side and 45 on the right hand side of the mandible.The literature mentioned that these cavities could contain salivary gland tissue, muscular tissue, lymphoid tissue, blood vessels, fat tissue or connective tissue.
Discussion/Conclusion: This study illustrates the wide variation of SBC locations across the mandible and leads us to conclude that the differential diagnosis of every asymptomatic, oval or round, well-defined, uniform radiolucent lesion on two-dimensional radiographs of the mandible should include "a benign mandibular concavity", formerly known as SBC.
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Introduction

A clinically justified diagnostic panoramic radiograph is common grounds in dentistry and provides an overview of the dentomaxillofacial complex and its adjacent anatomy. Dental clinicians are trained to recognize normal anatomical boundaries and landmarks, besides identifying pathology in the jaws. However, aberrant anatomy and its radiographical appearance can be confusing and may be misinterpreted as pathology.[1] The latter can lead to unnecessary additional imaging (*e.g.,* CBCT or multi-slice CT (MSCT)) which results in an increased cost and radiation burden for the patient. Moreover this can also result in unnecessary surgical interventions, which also have a cost, a potential risk of complications and it contradicts the Hippocratic oath (primum non-nocere). This can all be avoided if clinicians can be trained better by providing evidence, by publications, for these aberrant anatomical presentations appearing in radiographs.

Panoramic and intraoral radiographs of the mandible can reveal incidental findings, like a unilocular round or oval cystic appearing, uniform radiolucent, lesion with well-defined borders, which can be confusing for interpretation, especially if it occurs superior of or seems to be encroaching on the inferior alveolar nerve canal or on the inferior cortical border of the mandible, or if

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projected on a tooth's apex, or if observed in the ramus of the mandible.[2–4] It is important that clinicians think of a differential diagnosis when assessing their diagnostic images.

In 1942 Edward Stafne described for the first time, 35 asymptomatic, radiographically radiolucent cavities, unilaterally located in the posterior region of the mandible, between the mandibular angle and the third molar, and below the inferior dental canal and slightly above the inferior cortical border of the mandible. He described the following phenomenon as Stafne's bone cyst (SBC).[5–10] While at the time Stafne did not analyse the contents of these cavities on a microscopic level, what triggered his interest was fuelled by a multitude of factors. These included the similarity in the appearance of these cavities and a somewhat consistent location in oblique lateral and lateral radiographs. This congruence across the 35 cases that he observed suggested a common aetiological factor, which continued to be reported in the literature over time.[6] The aetiology remains unclear, but assumptions such as hypertrophy of a lobe of a salivary gland, bony erosion caused by vascular compression, or an incomplete Meckel cartilage calcification during ossification, have been proposed. According to Hisatomi et al. (2019),[6] the incidence of SBC is less than 0.5% for the posterior lingual variant, and differences in prevalence have been reported to vary between 0.10 and 0.48% and in a cadaver study a prevalence as high as 6.06% was reported.[11,12] The anterior lingual variant appears to be seven times less prevalent than the posterior one and can be expected between the incisors and the premolar area, superior to the insertion of the mylohyoid muscle.[13,14] According to the literature, the age range is quite wide, with a predilection for males over 50 years of age.[7,8,15] The size of the lesion was studied by Adisen et al. (2015) and they found that the mean volume of the defect was 361.7 mm^3.[16] This also explains the confusion it can cause when visible on radiographs.

Different definitions can be found in various radiology text books:

(1) a bone cavity or depression on the lingual aspect of the mandible near the lower border;[17]
(2) a well-defined round or ovoid or occasionally lobulated radiolucency that ranges in diameter from 1 to 3 cm, below the inferior alveolar canal and anterior to the angle of the mandible, in the region of the antegonial notch and the submandibular gland fossa;[18]
(3) a corticated unilocular radiolucency below the mandibular canal, between the first molar and a the angle of the mandible, with rare location in the ramus of the mandible (buccal or lingual);[19]
(4) a developmental depression along the mesial aspect of the mandible most commonly near the angle but sometimes along the body more anteriorly;[20]
(5) a not always corticated, well-defined round or oval defect or pseudocyst in the lingual surface of the mandible, usually at the mandibular angle caudally to the mandibular canal, seldom in the anterior part of the mandible.[21,22]

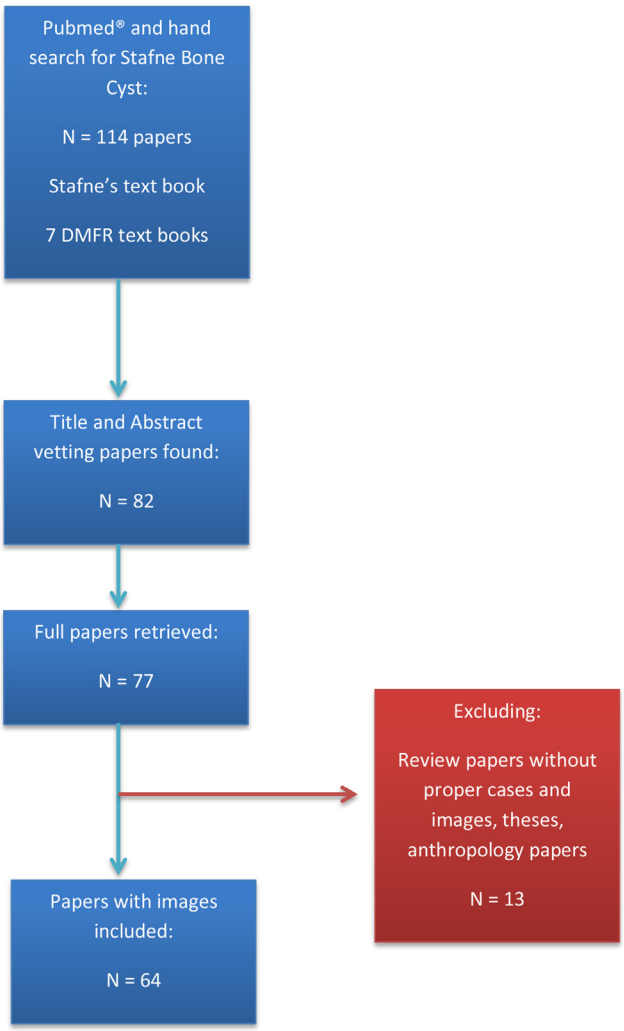

Figure 1 PRISMA flow chart for Stafne Bone Cyst narrative review (1 September 2018 – 31 May 2019)

The SBC is also known under a myriad of synonyms: Stafne bone cavity, lingual salivary gland depression, lingual mandibular bone depression, developmental salivary gland defect, Stafne defect, static bone cavity, latent bone cyst, mandibular salivary gland defect and idiopathic bone cavity.[1–22]

The aim of the present narrative review was to investigate, verify and map the different locations of a SBC from publications that actually showed the radiographs with the exact location of the lesion. Knowledge of the possible locations of this lesion helps clinicians establish a differential diagnosis when a round or oval, uniform radiolucent, well-defined, lesion is observed on a radiographic image of the mandible.

Methods and materials

A Pubmed® search for "SBC" was conducted between 1 September 2018 and 31 May 2019. Inclusion criteria for the review were: English, Dutch, French or German language, full papers available (some with the help of

Table 1 Publications on Stafne's Bone Cyst found on Pubmed® in the period 1980–2019, which contained images that were eligible for this narrative review

Year of Publication	*Authors*	*Country of Origin*
1980	Adra et al.[24]	Lebanon
1984	Chen and Ohba[25]	Japan
1985	Smith et al.[26]	United Kingdom
1986	Wolf et al.[27]	Finland
1988	Barker[28]	United Kingdom
1989	Tominaga et al.[29]	Japan
1990	Grellner et al.[30]	USA
1993	Ariji et al.[31]	Japan
	Shigematsu et al.[9]	Japan
1994	Tsui and Chan[32]	Hong Kong
1997	Graham et al.[33]	United Kingdom
1998	Reuter[34]	Germany
1999	Apruzzese and Longini[35]	Italy
	Branstetter et al.[36]	USA
	Junquera et al.[37]	Spain
2000	Boyle et al.[38]	United Kingdom
2001	Katz et al.[14]	Israel
2002	de Courten et al.[39]	Switzerland
	Dorman and Pierse[40]	United Kingdom
	Philipsen et al.[7]	Japan
2003	Longo et al.[41]	Italy
2004	Campos et al.[42]	Brazil
	Guedes Queiroz et al.[43]	Brazil
	Phillips and Yates[44]	United Kingdom
2006	Minowa et al.[45]	Japan and USA
	Quesada-Gomez et al.[8]	Spain
	Segev et al.[46]	Israel
2009	Bornstein et al.[47]	Switzerland
2010	Kao et al.[48]	Taiwan
	Kopp et al.[49]	Germany
	Krafft et al.[50]	Germany
	Sisman et al.[13]	Turkey
	Turkoglu and Orhan[51]	Turkey
	Flores Campos et al.[52]	Brazil
2011	Vieira Aguiar et al.[53]	Brazil
	Li et al.[54]	China
2012	Dereci and Duran[55]	Turkey
	Etoz et al.[56]	Turkey
	Friedrich et al.[57]	Germany
	Munevveroglu and Aydin[58]	Turkey
	Sisman et al.[12]	Turkey
2013	Boffano et al.[59]	Italy
	Prechtl et al.[60]	Germany
	Saglam et al.[61]	Turkey
2014	Assaf et al.[11]	Switzerland
	Herranz-Aparicio et al.[62]	Spain
	Kim et al.[63]	Korea
	Miloglu et al.[64]	Turkey
	Probst et al.[65]	Germany
	Schneider et al.[66]	Switzerland
	Taysi et al.[67]	Turkey
2015	Adisen et al.[16]	Turkey
	Lee et al.[68]	Australia and United Kingdom
	Mauprivez et al.[69]	France
	More et al.[70]	India
	Ozaki et al.[71]	Japan
	Venkatesh[72]	Kingdom of Saudi Arabia
2016	Atil et al.[73]	Turkey
	Chen et al.[74]	USA
	Deyhimi et al.[75]	Iran
	Lee et al.[76]	Korea
2017	Hisatomi et al.[77]	Japan and Brazil
2018	da Silva et al.[78]	Brazil
	Kaya et al.[79]	Turkey
	Nishimura et al.[80]	Japan
2019	Lee et al.[81]	USA
	Bayrak[82]	Turkey

the library of the University of Western Australia and by emailing authors), radiographic image of the lesion available in the publication, and finally patient gender and age clearly mentioned. A hand search was also performed through dental and medical handbooks available to the authors. A check was done using different search terms, such as aberrant salivary gland, Stafne bone cavity, Stafne's bone cavity and static bone cavity, in order to identify more papers, but that did not result in any additional manuscripts. Therefore, the authors considered their search as comprehensive and complete enough for the purpose of this narrative review.

Results

Figure 1 provides an overview of the search methodology in a typical PRISMA flow chart model.[23] A total of 114 papers were found on Pubmed®, of which 82 were deemed eligible for the purpose of this review. Only 77 papers were available as full paper. After thorough selection and vetting by the three authors, a final selection of 64 papers was deemed eligible for the final assessment, as they contained enough detailed imaging and patient information to be used for mapping the lesions. This resulted in 109 patient cases with SBC. The 13 papers that were excluded contained anthropomorphic studies, a PhD thesis, letters to the editor, general papers about incidental findings on radiographs and papers regarding other types of lesions mimicking SBC and finally papers covering classification of SBCs. The hand search resulted in retrieving Edward Stafne's 1963 text book and six other text books on dental and maxillofacial radiology which had a section dedicated to SBC.

Table 1 provides an overview of the publications per year, the authors and the country of origin of these papers. The period covers 1980 to 2019. There seems to be a trend, namely that the number of reports published per year has

Table 2 Distribution of imaging modalities used in the papers used for this narrative review about Stafne's bone cyst (N is the number of cases per imaging modality or combination of imaging modalities)

Type of Imaging	*Publications*	*N*	*%*
PANO	Chen and Ohba 1984[25] Sisman et al. 2012[12] Philipsen et al. 2002[7] Shigematsu et al. 1993[9] Adra et al. 1980[24] Assaf et al. 2014[11] Smith et al. 1985[26] de Courten et al. 2002[39] Apruzzese and Longoni 1999[35] Barker 1988[28] Prechtl et al. 2013[60] Friedrich et al. 2012[57] Kim et al. 2014[4] Miloglu et al. 2014[64]	40	36.70%
PANO + MSCT	da Silva et al. 2018[78] Minowa et al. 2006[45] Ariji et al. 1993[31] Boyle et al. 2000[38] Grellner et al. 1990[30] Phillips and Yates 2004[44] Kao et al. 2010[48] Katz et al. 2001[14] Bayrak 2019[82] Campos et al. 2004[42] Etoz et al. 2012[56] Reuter 1998[34] Junquera et al. 1999[37] Nishimura et al. 2018[80] Vieira Aguiar et al. 2011[53]	18	16.50%
PANO + CBCT	More et al. 2015[70] Ozaki et al. 2015[71] Atil et al. 2016[73] Bornstein et al. 2009[47] Sisman et al. 2010[13] Boffano et al. 2013[59] Muenevveroglu and Aydin 2012[58] Venkatesh 2015[72] Kopp et al. 2010[49] Taysi et al. 2014[67] Krafft et al. 2010[50] Lee et al. 2019[81]	19	17.40%
CBCT	Assaf et al. 2014[11]	1	0.90%
PANO + CBCT+MRI	Schneider et al. 2014[66] Probst et al. 2014[65] Bornstein et al. 2009[47]	3	2.80%
PANO + MSCT+MRI	Branstetter et al. 1999[36] Herranz-Aparicio et al. 2014[62] Segev et al. 2006[46] Saglam et al. 2013[61]	4	3.70%
MSCT	Dereci and Duran 2012[55]	1	0.90%
PANO + IO	Deyhimi et al. 2016[75] Dorman and Pierse 2002[40] Guedes Queiroz et al. 2004[43]	3	2.80%
PANO + MSCT+ SCINTIGRAPHY	Flores Campos et al. 2010[52]	1	0.90%
PANO + MRI	Graham et al. 1997[33] Lee et al. 2015[68] Hisatomi et al. 2017[77] Kaya et al. 2018[79] Mauprivez et al. 2015[69]	5	4.60%
PANO + IO+MSCT	Turkoglu and Orhan 2010[51] Grellner et al. 1990[30]	2	1.80%
IO + MSCT	Minowa et al. 2006[45]	1	0.90%
PANO + MSCT+ SIALOGRAPHY	Li et al. 2011[54] Tominaga et al. 1989[29]	2	1.80%
PANO + OCCL	Tsui and Chan 1994[32]	1	0.90%
PANO + SIALOGRAPHY	Wolf et al. 1986[27]	1	0.90%
CEPH + CBCT	Lee et al. 2016[76]	1	0.90%
PANO + CEPH+CBCT	Longo et al. 2003[41]	1	0.90%
PANO + CBCT+MSCT	Sisman et al. 2012[12]	5	4.60%
		109	100.00%

CBCT, cone beam computed tomography; CEPH, cephalometric radiography; IO, intraoral radiography; MRI, magnetic resonance imaging; MSCT, multi-slice computed tomography; OCCL, occlusal radiography; PANO, panoramic radiography.

increased after 2009, compared to the period before. The majority of papers came from Turkey, Japan and the UK.

In Table 2, one can find the distribution of imaging modalities that were used in the 64 manuscripts. Panoramic radiography, only, was used in 36.7% of the reports, with a combination of panoramic radiography and CBCT being used in 17.4% of cases and a combination of panoramic radiography and multi-slice CT being used in 16.5% of cases. The table shows the prevalence of imaging combinations used in the different papers. Panoramic imaging was involved in 14 of the 18 found imaging combinations in these papers (77.7%), as was expected as SBC is an incidental finding after all. Table 2 also shows which imaging modalities or combinations of modalities were used in the respective papers.

Table 3 summarises the different locations of SBC as recorded from the radiographic images found in the included papers of this review. The table also shows the anatomical locations per gender for the 109 cases in total. Only three lesions were located at the symphysis, six were bilateral, 55 appeared on the left hand side and 45 on the right hand side of the mandible.

With regard to patient age range, our analysis showed that patients' ages ranged from 14 to 89 years old, with the mean age being 52 years. Male patients' ages (N = 95) ranged from 14 to 89 years (mean age 52.3 years), whereas the females (N = 14) ages ranged from 22 to 68 (mean age 50.2 years).

In Figure 2, the different anatomical locations of SBCs are mapped on a panoramic radiograph of a mandible. This figure shows graphically what Table 3 summarised. In red are the ones that did not encroach on the mandibular canal, whereas the blue ovals are. The orange ovals involve

Table 3 Summary of the locations of Stafne's bone cysts, for the entire sample (N) and per gender (M,F), as checked in the radiographs included in the manuscripts used for this narrative review. The abbreviations used in this table are: RHS = right hand side, PM = premolar, *M* = molar, IANC = inferior alveolar nerve canal

Location of Stafne's Bone Cyst	*Publications*	*N*	*M*	*F*
RHS PM inferior to IANC	Shigematsu et al.1993[9]	1	1	
RHS PM superior to IANC	Phillips and Yates 2004[44]	1	1	
LHS PM superior to IANC	Krafft et al. 2010[50]	1	1	
Bilateral M to angle area, inferior to IANC but encroaching	Junquera et al. 1999[37]	1	1	
Bilateral angle of mandible	Mauprivez et al. 2015[69]	1	1	
LHS angle to M area inferior to IANC - double lesion	Miloglu et al. 2014[64]	1	1	
RHS ramus and LHS M area inferior to IANC and encroaching	Nishimura et al. 2018[80]	1	1	
RHS angle and RHS anterior mandible	Ozaki et al. 2015[71]	1	1	
RHS M inferior to IANC + RHS PM inferior IANC + LHS PM inferior IANC	Vieira Aguiar et al. 2011[53]	1	1	
LHS M superior to IANC	de Courten et al. 2002[39] Kopp et al. 2010[49]	2	2	
RHS anterior mandible	Tominaga et al. 1989[29] Turkoglu and Orhan 2010[51]	2	2	
RHS ramus	Campos et al. 2004[42] Lee et al. 2015[68]	2	2	
Bilateral PM area	Grellner et al. 1990[30] Kim et al. 2014[63]	2	1	1
RHS angle of mandible and encroaching IANC	Chen and Ohba 1984[25] Ozaki et al. 2015[71] Philipsen et al. 2002[7]	3	3	
LHS ramus	Barker 1988[28] Hisatomi et al. 2017[77] Lee et al. 2019[81]	3	3	
Midline mandible	Dorman and Pierse 2002[40] Grellner et al. 1990[30] Guedes Queiroz et al. 2004[43]	3	1	2
RHS M inferior to IANC	Minowa et al. 2006[45] Shigematsu et al. 1993[9] Sisman et al. 2012[12]	4	4	
RHS PM inferior to IANC and encroaching	Bornstein et al. 2009[47] Shigematsu et al. 1993[9] Sisman et al. 2010[13] Sisman et al. 2012[12]	4	2	2
LHS M inferior to IANC	Assaf et al. 2014[11] More et al. 2015[70] Sisman et al. 2012[12]	4	3	1
LHS PM inferior to IANC and encroaching	Apruzzese and Longoni 1999[35] Bornstein et al. 2009[47] Katz et al. 2001[14] Taysi et al. 2014[67]	4	3	1
LHS anterior mandible	Bayrak 2019[82] Dereci and Duran 2012[55] Deyhimi et al. 2016[75] Minowa et al. 2006[45]	4	4	
LHS angle to M area inferior to IANC and encroaching	Etoz et al. 2012[56] Flores Campos et al. 2010[52] Lee et al. 2016[76] Prechtl et al. 2013[60] Reuter 1998[34]	5	4	1
RHS angle to M area inferior to IANC and encroaching	Friedrich et al. 2012[57] Kaya et al. 2018[79] Longo et al. 2003[41] Muenevveroglu and Aydin 2012[58] Saglam et al. 2013[61]	5	5	

(Continued)

Table 3 (Continued)

Location of Stafne's Bone Cyst	*Publications*	*N*	*M*	*F*
LHS angle of mandible	Philipsen et al. 2002[7] Shigematsu et al. 1993[9] Sisman et al. 2012[12]	6	5	1
LHS angle of mandible and encroaching IANC	Adra et al. 1980[24] Assaf et al. 2014[11] Chen and Ohba 1984[25] da Silva et al. 2018[78] Herranz-Aparicio et al. 2014[62] Sisman et al. 2012[12]	9	9	
RHS angle of mandible	Branstetter et al. 1999[36] Chen and Ohba 1984[25] More et al. 2015[70] Sisman et al. 2012[12]	11	9	2
RHS M inferior to IANC and encroaching	Ariji et al. 1993[31] Atil et al. 2016[73] Boyle et al. 2000[38] Grellner et al. 1990[30] More et al. 2015[70] Schneider et al. 2014[66] Shigematsu et al. 1993[9] Smith et al. 1985[26] Tsui and Chan 1994[32]	11	10	1
LHS M inferior to IANC and encroaching	Adra et al. 1980[24] Ariji et al. 1993[31] Boffano et al. 2013[59] Graham et al. 1997[33] Kao et al. 2010[48] Li et al. 2011[54] More et al. 2015[70] Muenevveroglu and Aydin 2012[58] Probst et al. 2014[65] Segev et al. 2006[46] Sisman et al. 2012[12] Smith et al. 1985[26] Venkatesh 2015[72] Wolf et al. 1986[27]	16	14	2
Total		109	95	14

IANC, inferior alveolar nerve canal; LHS, left hand side; M, molar; PM, premolar; RHS, right hand side.

locations where the SBC was close to the apex of a tooth, mimicking a radicular lesion. In green are the lesions that covered a larger area of the jaw and in purple are the lesions located in the rami of the mandible. Eight cases were not graphically shown in this figure as these were patients in whom multiple SBCs were observed in different locations. The information about these can be retrieved from Table 3 (the white boxes in the prevalence column).

Discussion

The current study can be considered an inventory of published radiographically illustrated cases, which has resulted in an evidence-based map of SBC locations, which can now be used for teaching purposes. This study did not question and/or investigate the aetiology of the SBC or the age range in which one can expect this asymptomatic, incidental, radiographic finding to be observed. Therefore, the information regarding prevalence, age and gender is nothing new.

What is new, is however, what is illustrated in Figure 2: the mapping of possible locations of SBCs. The latter shows that radiographic interpretation of radiolucent lesions in the mandible can be complicated, as for instance SBCs can mimic other radiolucent lesions, as they can occur anywhere in the mandible. Occurrence of an SBC superior of the inferior alveolar canal and/or close to the apex of a tooth, complicates differential diagnosis of radiolucent lesions.[1–4,38,39,50,70,78] It is therefore the authors' opinion that all asymptomatic well-defined, round or oval, uniform radiolucent lesions, in the mandible should be considered as possible SBC. That being said, the definition of SBC as mentioned in text books and by Edward Stafne in 1942 is no longer accurate and we think these lesions should be called "benign mandibular concavities", as their content may be salivary gland tissue, but also lymphoid, muscular, adipose or vascular tissue, as mentioned in several publications.[5,10,17–22,31,33,36,46,47,54,57,63,69,70,80]

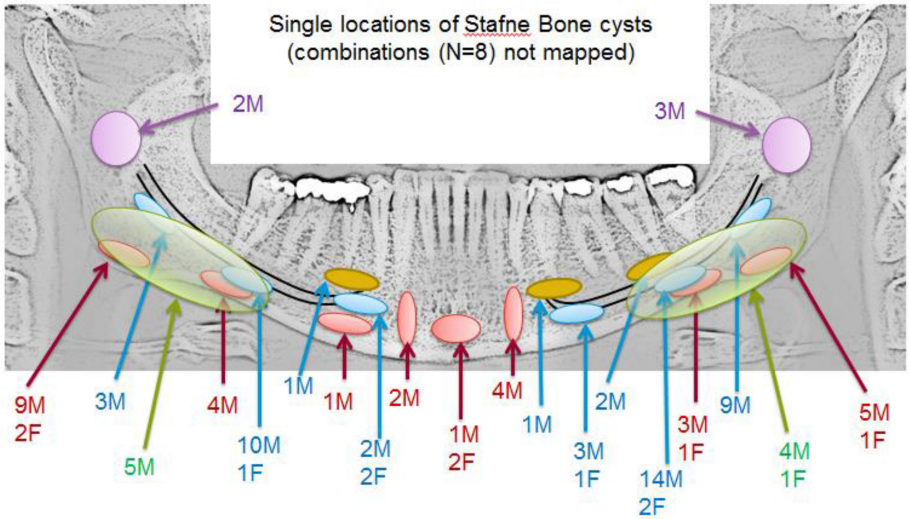

Figure 2 The mappings of the different locations of Stafne Bone Cysts as retrieved from the 64 papers which were included in this narrative review. M stands for male, F stands for female and the number indicates how many cases were identified. Eight cases in which multiple locations were present are not shown in this figure, as it would clutter the figure.

We are aware that several classifications have been proposed to identify the depth of the concavity and its actual content, but as this was not within the scope of the current review, we refer the interested reader to the relevant literature on this.[9,10,17–22,31,70] We were purely interested in the anatomical mapping of the lesions in order to illustrate to clinicians that a benign concavity in the mandible should always be considered with the incidental identification of an asymptomatic, round or oval-shaped, well-defined, uniform radiolucent lesion, anywhere in the mandible.

As mentioned above, the aetiology is unknown and it seems interesting to the authors that, despite the fact that millions of teenagers receive orthodontic treatment, and therefore undergo panoramic and other diagnostic imaging, there is no information available on the presence or emergence of these benign mandibular concavities in teenagers. Does that support the hypotheses that it is a developmental lesion? This calls for a longitudinal or retrospective study in which panoramic radiographs, taken for the right reasons, from patients are studied from childhood into adulthood.

It should be emphasized that this narrative review did not investigate the validity or accuracy of the imaging techniques to identify mandibular concavities. However, this could be an interesting topic for a future review.

Conclusion

SBC is an asymptomatic, incidental radiographic finding in the mandible. The current review study has mapped the locations of these lesions as mentioned in their respective publications. The conclusion is that these lesions can occur anywhere in the mandible and that they can mimic any asymptomatic round or oval-shaped, well-defined, uniform radiolucent lesion on a two-dimensional radiograph of the mandible. Therefore we suggest to leave the name SBC and to rename the entity as "benign mandibular concavity".

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