Systematic Review

Sinus Floor Augmentation—Associated Surgical Ciliated Cysts: Case Series and a Systematic Review of the Literature

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Abstract: This study aimed to characterize the demographic and clinical features of underreported surgical ciliated cysts developing after sinus floor augmentation, based on a series of cases from our files and a systematic review of the literature. A series of five cases (four patients) of microscopically confirmed surgical ciliated cysts following sinus floor augmentation procedures from our files are described. A systematic literature search (1991–2020) with strict clinical-, radiological- and microscopic-based exclusion and inclusion criteria was performed to detect additional similar cases. The systematic review revealed only five cases that fulfilled the inclusion criteria. Altogether, surgical ciliated cysts associated with sinus floor augmentation have been rarely reported in the literature, and have not been characterized either demographically or clinically. Graft materials were diverse, implants were placed simultaneously, or up to two years post-augmentation. The associated surgical ciliated cysts developed between 0.5 and 10 years post-augmentation. Although limited in its extent, this study is the first series to characterize possible underreported sequelae of surgical ciliated cysts associated with sinus floor augmentation. It emphasizes the need for long post-operative follow-up and confirmation of lesion by microscopic examination.

Keywords: sinus floor augmentation; surgical ciliated cyst; dental implant

1. Introduction

Surgical ciliated cysts (SuCCs), or postoperative maxillary cysts, are benign cystic lesions induced after a surgical procedure in the maxillofacial area. These lesions are associated with a history of radical sinus surgery, orthognathic surgery, and maxillofacial trauma [1–3]. The first report of postoperative maxillary cyst after sinus augmentation was made in 1991 [4]. In general, SuCCs are diagnosed in the 4th–5th decades of life, with an inconsistent gender distribution [5]. The mean period of cyst development after the surgical procedure is estimated to be 18 years (range 4 to 49 years), and might be accompanied by pain, discomfort, or swelling, either in the intra- or extra-oral adjacent areas. The radiological features are usually of well-defined radiolucent, unilocular lesions [1–5]. It was proposed that the entrapment of maxillary sinus mucosa during the surgical procedure, followed by an inflammatory process that induces cystic changes of
the entrapped respiratory mucosa and expansion of the cyst by the osmotic difference from the surrounding tissue, are the pathogenesis for SuCC formation [6].

Evidence-based medicine is an approach to healthcare that combines the best available clinical evidence to support a practitioner’s clinical expertise for each patient’s preferences and treatment needs [7–9]. It is founded on the process of systematically detecting, appraising, and exhausting scientific findings as the foundation for clinical decision-making. Systematic reviews institute the foundation for practicing evidence-based medicine. The solicitation of evidence-based practice in dentistry should lead to a decline of mistakes during the clinical decision-making process [7–10]. Thus, the evidence-based systematic review of the current literature concerning the sequelae of SuCCs associated with sinus floor augmentation is important in order to increase awareness among dental practitioners about this possible complication in the setting of a frequently performed surgical procedure.

It can be assumed that the incidence of SuCCs following sinus floor augmentation procedures is under-reported. In the current study, a series of five new cases of SuCCs associated with sinus floor augmentation is presented and analyzed. Furthermore, a systematic review of the literature was undertaken, aiming to detect and analyze the available scientific evidence concerning this specific type of lesion.

2. Materials and Methods

A series of four patients referred for sinus floor augmentation between 2016 and 2019, with a total of five cases of post-procedure, microscopically confirmed SuCCs, were retrospectively pooled and analyzed. The following variables were recorded for each of the patients based on their clinical and radiographic examinations and medical records: age and gender, graft material, number of implants placed after sinus floor augmentation, timing of implant placement related to the augmentation procedure, location of the cyst, and greatest dimension and timing of histopathological diagnosis of the SuCC in relation to the augmentation procedure. The study was performed in accordance with ethical requirements (both Israeli and the International Conference on Harmonisation Guideline for Good Clinical Practice standards) of clinical trials [11]. Institutional Review Board approval is not required for case series.

Data collection from the systematic review of the literature PRISMA guidelines were followed for the systematic review, which included the definition of a specific research issue, pre-specified eligibility criteria to fulfill that issue, explicit, reproducible searching methodology for the identification of all published studies that met the eligibility criteria, and a systematic presentation of the findings of the finally included studies [12].

2.1. Inclusion Criteria for Selecting Studies and Cases in the Systematic Review

The following inclusion criteria were used:

1. SuCC was diagnosed after the sinus floor augmentation procedure, based on a clinical and radiographic evaluation;
2. Absence of clinical and/or radiographic evidence of any antral pathology at the time of the sinus floor augmentation procedure;
3. SuCC was confirmed by microscopic evaluation.

2.2. Exclusion Criteria of Studies from the Systematic Literature Search

Studies not relevant to the topic of this study, expert opinions, and reviews were excluded. Literature sources that were not in English were also excluded.

2.3. Search Methods for Detection of Studies for the Systematic Review

These electronic databases were searched: MEDLINE database using PubMed (http://www.ncbi.nlm.nih.gov/sites/pubmed, accessed on 2 November 2020), Scopus (http://www.scopus.com, accessed on 2 November 2020), and Embase (https://www.embase.com, accessed on 1 November 2020) search engines from 1991 up to September 2020.
The following keywords were used for an initial search through MEDLINE: (((Sinus Floor Augmentation) OR Sinus Floor elevation) OR Sinus lift)) AND (((surgical ciliated cyst) OR paranasal cyst) OR postoperative maxillary cyst) OR post-operative maxillary cyst) OR respiratory epithelium implantation cyst) OR respiratory implantation cyst). Additional searches were then performed through the Scopus and Embase databases with the same keywords.

Furthermore, the reference lists of literature reviews that were detected in MEDLINE were manually screened to detect any additional eligible articles that were not yet detected during the electronic databases search.

2.4. Data Collection and Analysis for the Systematic Review

Articles that were detected in the literature search were initially screened by two independent observers (A.K. and R.H.) for relevance on the basis of their abstracts and titles. We planned to subject potentially eligible studies to a full-text assessment based on the inclusion and exclusion criteria for selected studies and cases in the systematic review. Finally, we planned to submit the detected eligible articles to data extraction and analysis.

We planned to analyze the identified eligible cases for the patients’ demographics, parameters of the sinus floor augmentation procedure, and of associated SuCCs. We also planned to assess the eligible studies concerning their methodological quality and heterogeneity, with the possibility of performing a meta-analysis of their data.

3. Results

3.1. Case Description

3.1.1. Case 1

A 52-year-old woman presented with bilateral edentulous posterior maxilla for fixed dental rehabilitation. Her medical history was non-contributory. The sub-antral space was grafted with allograft (cortical granules, DIZG, Berlin, Germany) and inorganic bovine bone mineral (XBM, 4BONE™, MIS Implants, Misgav, Israel). The access windows were covered with collagen membranes (Ossix Plus, Datum Dental Biotech, Lod, Israel). Three implants were bilaterally inserted in the posterior maxillary alveolar ridge 9 months after the augmentation. Intra-operatively, there was no visible damage to the Schneiderian membrane. The postoperative course was uneventful and the implants were rehabilitated with porcelain fused to metal bridges. Two years later, she presented with a swelling of the left buccal alveolar ridge. Cone beam computerized tomography (CBCT) revealed a hypo-dense lesion of the left posterior maxillary alveolar ridge involving the posterior implant (Figure 1A). The implant was removed and the cyst was enucleated. The histopathologic result was a SuCC (Figure 1B,C). During 2 years follow-up, no recurrent cyst was identified.

3.1.2. Case 2

A 68-year-old male presented with posterior partial edentulism of the right maxilla. The medical history was positive only for simvastatin-treated dyslipidemia. He underwent a right lateral window sinus floor augmentation using bovine bone mineral as the bone substitute (Bio Oss, Geistlich Biomaterials, Baden-Baden, Germany) and the lateral access window was covered with a collagen membrane (Bio Gide, Geistlich Biomaterials, Baden-Baden, Germany). He underwent an intraoperative procedure without any complications. Due to relocation, he came back for implant placement two years later, and the CBCT showed a hypodense lesion in the posterior part of the alveolar ridge. Otherwise, satisfactory bone formation was observed and three implants were inserted. During that setting, the hypodense lesion was enucleated and the histopathologic diagnosis was SuCC. The patient has come for follow-up visits for 2 years with no recurrent cyst.
Figure 1. Cone-beam computerized tomography (CBCT) of the surgical ciliated cyst in present case #1, as seen in reconstructed slices in three planes: axial, coronal, and sagittal (left to right, A); intra-operative view (B). The microscopic examination revealed a collapsed cystic structure (C); lined pseudostratified ciliated columnar epithelium typical of the maxillary sinus (D). (C,D)—hematoxylin and eosin stain; (C)—X20 original magnification; (D)—X400 original magnification).

3.1.3. Case 3

A 60-year-old male patient with bilateral posterior partial maxillary edentulism was referred for a fixed rehabilitation of the missing teeth. A bilateral lateral window sinus floor augmentation was performed using an allograft (Oragraft mineralized cortical particulate, LifeNet Health, Virginia Beach, VA, USA) as the bone substitute. The access windows at the lateral sinus walls were covered with a resorbable collagen membrane (Ossix). Six months later, two implants were inserted in the right posterior alveolar arch and three in the left posterior maxillary alveolar arch. The entire surgical procedure occurred with no complications. The CBCT obtained three months after the implant placement showed a hypodense lesion involving the implant located in the area of the first right molar. The lesion was enucleated, the cavity was filled with an allograft (Oragraft, LifeNet Health, Virginia Beach, VA, USA), and the adjacent implant was explanted. Microscopic examination revealed a SuCC. Follow-up for 3 years revealed no recurrence of the cyst.

3.1.4. Case 4

A 44-year-old healthy female with total maxillary edentulism and maxillary alveolar arch atrophy presented to the clinic for fixed maxillary dental reconstruction. She underwent bilateral sinus floor augmentation, and eight implants were inserted for a full arch maxillary dental rehabilitation. The intraoperative procedure was uneventful. Two years later, the patient returned to the clinic with bilateral swelling on the buccal aspect of the maxillary alveolar ridges. Radiologically, bilateral well-circumscribed hypodense lesions were observed on a maxillary CBCT (Figure 2A,B). The lesions were enucleated and grafted with bovine bone mineral. The histopathologic diagnosis for both lesions was SuCC. The patient was lost to further follow-up.
Figure 2. Cone-beam computerized tomography (CBCT) of the surgical ciliated cysts in present case #4-5—partial pseudo-panoramic reconstruction and appropriate transaxial slices, right maxilla, (A) and left maxilla (B).

3.2. Results of the Systematic Review

In the MEDLINE search, 560 studies were detected. The Scopus and EMBASE databases search detected an additional four and nine articles, respectively, and the manual search identified ten additional relevant articles. After excluding duplicates, the systematic review included a total of 581 items, which were screened based on their abstracts and titles. Nevertheless, according to the prespecified exclusion and inclusion criteria, 567 were excluded since they were either irrelevant or reviews, nine were excluded because of inadequate location, cyst type not specified, surgery procedure not aimed for sinus floor augmentation, or lack of microscopic evaluation of the cystic lesion. Only five case report articles were finally included in the review (Figure 3). Consequently, the systematic literature search found that to date, there are only a few case reports that report SuCCs associated with sinus floor augmentation, with no possibility of performing any meta-analysis.
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3.3. Results of the Case Series—Our New Cases and Cases from the Literature Review

A total of new five cases of SuCCs after maxillary sinus augmentation were collected from our files, which represented 0.15% from the odontogenic cysts accessioned between 2016 and 2019 (Table 1). We added five cases from the literature to these [4,13–16] (Table 2).

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**Table 1.** Compilation of cases of surgical ciliated cysts (SuCCs) from our files.

| Case #     | Age at Diagnosis (Years) | Sex | Side of Maxillary Sinus | Graft Material                  | No. of Implants Placed/mo. after Augmentation | Cyst Diagnosis (Years after Augmentation) | Cyst Dimension (Largest Diameter) |
|------------|--------------------------|-----|--------------------------|---------------------------------|-----------------------------------------------|------------------------------------------|----------------------------------|
| Present case #1 | 54                        | F   | Left                     | Allograft, bovine bone mineral | 3 implants/9 mo.                            | 2                                       | 1 cm                             |
| Present case #2 | 70                        | M   | Right                    | Bovine bone mineral            | 2 implants/24 mo.                           | 2                                       | 1.5 cm                           |
| Present case #3 | 61                        | M   | Right                    | Allograft                      | 2 implants/6 mo.                            | 0.9                                     | 1.5 cm                           |
| Present cases #4–5 | 46                        | F   | Bilateral                | Bovine bone mineral            | 8 implants/simultaneous                     | 2                                       | 1 cm, 1.5 cm                     |

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**Figure 3.** Flow chart of the systematic search process of the literature.
Table 2. Compilation of cases of SuCC from the literature review.

| Case # | Authors                  | Country    | Age at Diagnosis (Years) | Sex | Side of Maxillary Sinus | Graft Material                                    | No. of Implants Placed/mo. after Augmentation | Diagnosis (Years after Augmentation) | Cyst Dimension (Largest Diameter) |
|--------|--------------------------|------------|--------------------------|-----|--------------------------|---------------------------------------------------|--------------------------------------------|-------------------------------------|----------------------------------|
| 1      | Misch et al., 1991 [4]   | USA        | 76                       | F   | Right                    | No information                                    | 1 blade implant/simultaneous                | 3                                   | 1 cm                             |
| 2      | Lockhart et al., 2000 [16] | France      | 41                       | F   | Left                     | Autogenous bone                                   | No information                             | 0.5                                | 1 cm                             |
| 3      | Kim et al., 2013 [14]    | South Korea | 60                       | M   | Right                    | Autogenous bone                                   | 5 implants/5 mo.                            | 10                                 | No information                   |
| 4      | Yamamoto et al., 2017 [15] | Japan      | 64                       | F   | Left                     | Tricalcium phosphate, hydroxyapatite, autogenous bone | 4 implants/6 mo.                            | 9                                  | No information                   |
| 5      | Han, 2018 [13]           | South Korea | 63                       | M   | Left                     | Allograft, autogenous bone, collagen                | 2 implants/simultaneous                     | 5                                  | 4.5 cm                           |

In total, cases were reported from North America (N = 1), Europe (N = 3), and Asia (N = 6). The group was comprised of five females and four males (ratio 1:1.25), with a mean age of 58 ± 10.65, and an age range of 41–76 years. The augmentation was performed by various types of graft materials, as detailed in Table 1. One to eight dental implants were placed in the augmented area in a period of time that ranged between the time of augmentation (simultaneous implant placement) and up to 2 years after. The SuCCs were diagnosed from 0.5 years after the augmentation up to 10 years post-operatively, the mean time to their diagnosis being 3.6 ± 3.2 years. Swelling was present in six cases (our cases #1, 4–5 and in those reported by Kim et al. [14], Yamamoto et al. [15], and Han [16]). Difficulty in mastication, tenderness on palpation, and pain were reported in three cases (Kim et al. [14], Yamamoto et al. [15], Han [16]).

4. Discussion

The foundations of evidence-based dentistry (EBD) rely on using the best available clinical evidence to support the practitioner’s daily practice. It relies on the practice of systematically finding, appraising, and exploiting research findings as the foundation for clinical decision-making [7–10,12]. This systematic process requires an extensive and comprehensive literature search aiming to detect as much of the relevant literature as possible. In the current study, a comprehensive literature search of three different electronic databases and a manual search of related literature reviews resulted in the detection of 581 potential titles. To prevent heterogeneity of data, rigorous exclusion and inclusion criteria were applied in order to evaluate studies for the systematic review, including surgical procedure for sinus floor augmentation, absence of any clinical/radiographic sinus pathology prior to this procedure, and microscopic confirmation of the diagnosis of SuCC [5]. These same criteria were adopted in order to include cases from our files. As SuCC has been mostly associated with reports from the Japanese literature in connection with the Caldwell-Luc procedure for treating maxillary sinusitis, most of the titles from the initial screening had to be excluded for the purposes of the present search, so much so that only 14 possibly relevant full-text articles remained. Of these, nine articles were also excluded since they did not fulfill all inclusion criteria, yielding only five eligible articles, each presenting one case of SuCC as a sequela of sinus floor augmentation. Thus, the present systematic review of the literature revealed that to date, the current scientific knowledge regarding the possible complication of the development of SuCC following sinus floor augmentation relies primarily on case reports, which is considered to be a
low level of evidence [7–10]. Thus, although limited in its extent, this case series is the first systematic report of this possible complication in the setting of a common surgical pre-prosthetic procedure.

Maxillary sinus augmentation is a successful and frequently used pre-prosthetic technique for augmentation of the edentulous posterior maxilla [17]. However, there are well-known possible complications, including sinus membrane perforation, bleeding, infection, and resorption of the graft [18,19]. Perforation of the Schneiderian membrane is the most common operative complication during sinus floor augmentation, and the incidence varies between 20% and 44% during the lateral window approach [18]. Another potential complication of sinus floor augmentation is SuCC.

Initially, postoperative maxillary cysts were not related to sinus floor surgery. In general, this type of cyst was first reported by Kubo in 1927 [20] and emerged as a common lesion in Japan, accounting for 19.5% of all oral cystic lesions. Subsequently, these cystic lesions were defined as “surgical ciliated cysts of the maxilla” [21], with only a few cases being reported in the English-language literature [2,22]. They can arise as a late complication of Caldwell-Luc operation for the treatment of maxillary sinusitis; however, the frequency of SuCC in this setting has seemed to decline since the introduction of endoscopy as an alternative conservative procedure. Other cases have occurred following mid-face orthognathic surgery [23] or genioplasty [24].

The SuCCs from our files occurred in the fifth to seventh decade of life, which is similar to the range reported in previous studies [4,13–16]. All our cysts were diagnosed up to three years following sinus floor augmentation, while three of the five cysts from the literature being diagnosed 5, 9, and 10 years following sinus floor surgery [13–15]. The dimension of the SuCCs reported by us were 1–1.5 cm in their greatest diameter. Misch et al. [4] and Lockhart et al. [16] reported similar cyst dimensions, diagnosed 6 months and 3 years after sinus floor augmentation, respectively. In contrast, Han [13] described a larger lesion, exceeding 4 cm in diameter, diagnosed five years following sinus floor augmentation. SuCCs, in general, can enlarge and expand with time, resulting in bone defects and thinning and perforation of the sinus walls to the point that they might mimic a malignant process [22,25]. This emphasizes the importance of the follow-up, which can aid in identifying smaller lesions and curb progressive bone destruction caused by cyst expansion. In two of our cases (#1 and #3), the two implants involved in the cystic lesion were explanted, although it was reported that involved implants may not need to be explanted during the SuCC enucleation [14].

Despite the very few reports of SuCC following sinus floor augmentation, the possibility of occurrence may not be negligible, especially in view of the proposed formation process related to Schneiderian membrane entrapment. Mucosal entrapment can be caused by Schneiderian membrane tear, which is the most common complication of sinus floor augmentation [26,27]. Perforations can occur due to inadequate surgical technique, the presence of septae, thin membranes, sinus pathology, sinus augmentation redo after failure, and overfilling at the time of graft placement [28]. In addition, an overestimation of approximately 2.5-fold in the assessment of the thickness of the Schneiderian membrane on CBCT images compared to the histological value has been reported [29]—a finding that could also increase the intraoperative risk of damaging the membrane. There is conflicting information in the literature regarding the effect of Schneiderian membrane perforation on postoperative infection and graft failure. Several studies have shown correlations between membrane perforation and acute sinusitis or graft infection [30,31], whereas others have shown no association between membrane integrity and infection [32]. Thus, knowledge of the range of these complications, and especially the risk of SuCC formation, emphasizes the need for adequate surgical techniques, with the need for careful augmentation of the sinus membrane and long-term clinical and radiologic follow-up, even in cases where the clinical procedure of sinus floor augmentation did not reveal macroscopic breach of the Schneiderian membrane.
Irrespective of the etiological factors, the frequent radiological features of SuCCs were of well-defined, radiolucent unilocular lesions adjacent to the maxillary sinus, albeit a few cases of multilocular appearance and less defined margins were reported [33]. Depending on the age of the patients, the common presentation of SuCC can raise the possibility of several differential diagnoses, including odontogenic cysts (e.g., residual cyst, odontogenic keratocyst), odontogenic tumors (e.g., ameloblastoma, odontogenic fibroma) and non-odontogenic tumors (e.g., central giant cell granuloma), as well as fibro-osseous lesions. In general, the diameter of SuCC, as measured on 2D-images, was most commonly found to be about 20 mm in diameter, however lesions over 31 mm in diameter were also reported [32], which is also the range found in our study in regards to SuCCs associated with augmentation of the sinus floor. Small lesions are expected to remain asymptomatic; however, as SuCCs slowly enlarge, perforation of the sinus walls might result, and even expansion beyond the original walls of the sinus has been described [34–36].

We may raise the assumption that the apparent rarity of SuCCs in conjunction with sinus floor augmentation is linked to their being underreported for a number of reasons—both lesion- and clinician-related. The commonly small dimensions of the cysts and their slow development do not elicit clinical symptoms; therefore, these areas might not be regularly evaluated radiologically. In addition, many of the surgical procedures related to dental implants are not necessarily performed at present by specialist dental surgeons (e.g., oral and maxillofacial surgeons, periodontologists). Thus, their degree of awareness of the development of this type of cyst, differential diagnosis, clinical sequelae, and the need to submit lesions for microscopic examination might not be as high as that of specialist surgeons. Variations in dental practice patterns are similar to those encountered in other fields of medicine, and are usually dependent on the individual practitioner’s knowledge, skills, attitudes, health system accessibility, time to receive biopsy results, and insurance issues [37].

Treatment of SuCCs, regardless of the etiologic factors, consists of enucleation; however, a more aggressive approach might be needed in the case of large lesions, bony perforation, or recurrence [34].

5. Conclusions

The present case series based on a systematic review of the literature and our cases, although limited in its extent, is the first clinical and systematic report of a possible complication of sinus floor augmentation procedure in relation to the ensuing development of SuCCs. Since we assume that this complication is underreported in the literature, this study aims to increase awareness among dental practitioners about SuCCs as a result of a frequently performed surgical procedure, and to emphasize the need for regular, long-time follow-up procedures. Additional clinical studies are warranted in order to shed light on this potential complication.

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