Bone exostosis is defined as a benign overgrowth of bone tissue of unclear origin. Rarely, bone exostosis might develop following soft tissue graft procedures like mucogingival surgical interventions (eg, FGG or subepithelial CTG). This aberration has been mainly associated with surgical trauma or fenestration of the periosteum but is still a matter of debate. The present paper (1) presents a clinical case with clinical, radiographic, and histologic findings at 30 years following application of an FGG to increase the gingival width and (2) provides a short literature review on this particular clinical condition. At the clinical examination, the FGG was firm to palpation, and the 3D images showed an area of increased radiopacity. Histologic analysis revealed localized thickening of the bone with an overlaying connective tissue covered by keratinized epithelium. The bony tissue was vital, had a convex shape, and contained many osteocytes and resting lines, demonstrating some moderate signs of bone remodeling. The connective tissue and keratinized epithelium displayed a regular thickness without any signs of inflammation. Taken together, the histologic findings failed to reveal any pathologic signs except for the presence of vital bone formed outside the bony envelope. It can be concluded that: (1) the development of a bone exostosis following a mucogingival procedure is a rare clinical sequela of uncertain etiology, and (2) surgical removal of the exostosis may be indicated accordingly with patient symptoms.

First described by Björn in 1963 and later modified by Sullivan and Atkins and Gordon et al in 1968, the use of an autogenous free gingival graft (FGG) harvested from the palate has been widely demonstrated to be an effective mucogingival procedure for increasing the width of the attached mucosa. However, some postoperative early (eg, hemorrhage and bone exposure from the donor site) and late (eg, graft dimension changes, color, and texture alteration of the recipient area) complications have been reported in the literature. Among these, the development of a bone exostosis as a sequela to such a surgical procedure has been detected only in a few cases. Moreover, exostosis formation after other transplantation procedures, such as connective tissue grafts (CTG) or skin grafts, have also been observed. However, at present, very few reports are available presenting clinical, radiographic, and histologic findings related to the use of an FGG.

Therefore, the aim of the present report is to summarize the available evidence on the development of bone exostosis following transplantation of an FGG or CTG in the mobile alveolar mucosa and to present the clinical, radiographic, and histologic features of a patient presenting formation of a bone exostosis after placement of an FGG.
Case Report

In May 2019, a 48-year-old woman in good general health not affected by periodontal disease and not presenting any bone exostosis in the oral cavity presented for a clinical examination. An increased volume of a previously placed FGG on the buccal aspect of the maxillary canine and first premolar was observed (Fig 1). The patient reported to have been treated 30 years before with an FGG harvested from the palate in order to increase the width of the attached gingiva and to prevent the development of gingival recessions. She could not recall any complications related to the donor or recipient sites. Over time, she noticed a gradually slight increase in tissue thickness in the grafted area, accompanied by an irritation of her lips. Therefore, she wanted the excess tissue to be removed.

At the clinical examination, the FGG was very firm to palpation, and the 3D images revealed an area of increased radiopacity (Figs 2a and 2b).

All of the procedures followed the revised Declaration of Helsinki. The patient received verbal information and signed informed consent prior to surgery.

To remove the grafted part, a full-thickness mucoperiosteal flap was raised, and the FGG was gently excised from the underlying periosteum (Fig 3a). In order to avoid tissue damage, a piezoelectric device was used. The removed biopsy specimen (Fig 3b) had a hard consistency and was about 15 mm wide and 4 mm deep. The operated area was then covered with a collagen matrix (Mucograft, Geistlich) to reduce the patient’s postoperative discomfort and improve soft tissue healing (Figs 4 and 5).

Histologic Processing

Immediately after surgery, the excised biopsy specimen was fixed in 4% formaldehyde, decalcified in 4.13% ethylenediaminetetraacetic acid (EDTA), and sliced into several sections. All sections were dehydrated in ascending concentrations of ethanol, and some were processed for embedding in LR white acrylic resin or paraffin. The specimens were sectioned in a bucco-oral plane and stained with...
toluidine blue (resin sections) or hematoxylin and eosin (paraffin). Microphotography was performed with a digital camera (Axiocam MRc, ZEISS) combined with a light microscope (Axio Imager M2, ZEISS).

**Histologic Findings**

A localized thickening of the bone with overlaying connective tissue covered by a keratinized epithelium was observed (Figs 6a and 7a). The bony tissue was vital, had a convex shape, and contained many osteocytes and resting lines (Figs 6b and 7b). Furthermore, signs of bone remodeling were visible, but moderate. The connective tissue and epithelium showed a regular thickness without any signs of inflammation (Figs 6b, 6c, 7b, and 7c). Moreover, the epithelium was keratinized. Altogether, no pathologic findings were seen, except for the exostosis.

**Literature Review and Discussion**

Oral bone exostosis is a congenital benign overgrowth mainly located on the lingual aspects of the mandible (mandibular tori), palate (palatal tori), and on the buccal aspect of the gingiva (buccal bone exostoses). Their etiology has been...
related to several factors. In a study in mono- and dizygotic twins, it was shown that oral bony outgrowths were dominantly influenced by genetic factors. However, the heritability did not explain all the cases in that study, and furthermore, it was impossible to answer the question on the continuing lifetime growth and occurrence of exostoses. Therefore, environmental factors such as occlusal stress, well-developed muscles, and eating habits (eg, vitamin deficiency, supplements rich in calcium, fish with omega-3 fatty acids, and vitamin D), have been suggested to be associated with oral bone exostosis. In a more recent study, there was a clear connection between the prevalence of mandibular tori and dental attrition or occlusal contact area. Furthermore, mandibular anatomy (eg, square-shaped), which may favor higher forces in specific areas, might be related to mandibular tori.

In the literature, 27 cases of bone exostosis formation following mucogingival surgery have been reported. Details of these findings are summarized in Table 1. Most of the cases were treated with an FGG, while only 3 developed after a skin graft and 2 following a connective tissue graft. The reason for the development of exostoses after mucogingival surgery is still not well understood, and the number of reported cases is limited. It is hypothesized that an accidental or intentional trauma of the periosteum could lead to inflammatory reactions with subsequent liberation and differentiation of osteoprogenitor cells, inducing new bone formation. Furthermore, a combination of the surgical trauma, occlusal stress, and genetic factors was also suggested. The association with extra- and intraoral exostosis and its development following mucogingival surgery has been described by Pack et al, who reported two cases, one with a mandibular torus and one with exostosis of the arms and feet. They suggested that these patients were more susceptible to exostosis development. Nonetheless, these speculations could not be confirmed by other authors, who did not detect any association between these findings. These results are in agreement with
the present case report, as the current patient presented only this new bone overgrowth following an FGG procedure.

Interestingly, almost all cases reported in the literature were in the same intraoral location, namely the premolars and canines in both the maxilla and mandible. Most of the mucogingival surgeries presented in the published case reports were performed between 17 and 30 years before tissue excision in a group of patients aged from 18 to 29 years. Most gingival recessions develop at premolar, canine, and maxillary first molar sites. Therefore, the location of exostosis occurrence after transplantations may be mainly due to the area of treatment (ie, premolars and molars).

After development of exostosis, patients are most frequently free of any symptoms. Nevertheless, some patients complain of mild increasing pain or discomfort, food retention, inability to wear protheses, and inability to wear dentures.

### Table 1 Summary of the Cases Reported in the Literature

<table>
<thead>
<tr>
<th>Study, y</th>
<th>Age at graft, y</th>
<th>Follow-up, y</th>
<th>Surgery (y performed)</th>
<th>Gender (F/M)</th>
<th>Sites</th>
<th>Symptoms</th>
<th>Histologic report</th>
<th>Tori</th>
</tr>
</thead>
<tbody>
<tr>
<td>Siegel and Pappas, 1986&lt;sup&gt;12&lt;/sup&gt;</td>
<td>46</td>
<td>10</td>
<td>SG</td>
<td>M</td>
<td>Mandible&lt;sup&gt;b&lt;/sup&gt;</td>
<td>Inability to wear denture</td>
<td>Sclerotic mature lamellar bone and loose CT</td>
<td>NR</td>
</tr>
<tr>
<td>Pack et al, 1991&lt;sup&gt;23&lt;/sup&gt;</td>
<td>36 26</td>
<td>9 11</td>
<td>FGG (1977) FGG (1977)</td>
<td>F F</td>
<td>34&lt;sup&gt;b&lt;/sup&gt;</td>
<td>Disturbing</td>
<td>Very dense lamellar bone NA</td>
<td>Yes No</td>
</tr>
<tr>
<td>Efeoglu and Demirel, 1994&lt;sup&gt;20&lt;/sup&gt;</td>
<td>17 23</td>
<td>6 1</td>
<td>FGG FGG</td>
<td>F F</td>
<td>43&lt;sup&gt;b&lt;/sup&gt;</td>
<td>Concerned</td>
<td>Very dense lamellar bone, no evidence of surface activity NA</td>
<td>No No</td>
</tr>
<tr>
<td>Czuszak et al, 1996&lt;sup&gt;18&lt;/sup&gt;</td>
<td>17 17</td>
<td>FGG (1977)</td>
<td>F</td>
<td>34, 35&lt;sup&gt;b&lt;/sup&gt;</td>
<td>Food retention</td>
<td>Dense, viable cortical bone</td>
<td>NA</td>
<td></td>
</tr>
<tr>
<td>Otero-Cagide et al, 1996&lt;sup&gt;22&lt;/sup&gt;</td>
<td>23 22 18 30 20 19 19 24</td>
<td>5 16 7 2 10 5 15 16</td>
<td>FGG (1980) FGG (1976) FGG FGG FGG FGG FGG</td>
<td>F F F F</td>
<td>14&lt;sup&gt;b&lt;/sup&gt; 24 31, 41 33&lt;sup&gt;b&lt;/sup&gt; 34&lt;sup&gt;b&lt;/sup&gt; 43 33 33&lt;sup&gt;b&lt;/sup&gt; 34&lt;sup&gt;b&lt;/sup&gt; 13, 23, 41 13, 23, 33, 43 13, 23, 24 13, 23, 33, 43 13, 23, 24 13, 23, 33, 43 23 43 34 23 14</td>
<td>No symptoms</td>
<td>No symptoms</td>
<td>No symptoms</td>
</tr>
</tbody>
</table>

CT = connective tissue; CTG = connective tissue graft; EMD = enamel matrix derivative; FGG = free gingival graft; NA = not applicable; NR = not reported; SG = skin graft.

<sup>a</sup>FDI numbering system.

<sup>b</sup>Surgical removal.
<table>
<thead>
<tr>
<th>Study, y</th>
<th>Age at graft, y</th>
<th>Follow-up, y</th>
<th>Surgery (y performed)</th>
<th>Gender (F/M)</th>
<th>Sites&lt;sup&gt;a&lt;/sup&gt;</th>
<th>Symptoms</th>
<th>Histologic report</th>
<th>Tori</th>
</tr>
</thead>
<tbody>
<tr>
<td>Corsair et al, 2001&lt;sup&gt;9&lt;/sup&gt;</td>
<td>NR</td>
<td>5</td>
<td>CTG</td>
<td>NR</td>
<td>15, b 13&lt;sup&gt;b&lt;/sup&gt;</td>
<td>NR</td>
<td>Viable bone and marrow</td>
<td>NR</td>
</tr>
<tr>
<td>Echeverria et al, 2002&lt;sup&gt;19&lt;/sup&gt;</td>
<td>23</td>
<td>19</td>
<td>FGG (1980)</td>
<td>F</td>
<td>33, b 34&lt;sup&gt;b&lt;/sup&gt;</td>
<td>Progressively uncomfortable</td>
<td>Woven bone with osteocytes, dense CT, no inflammatory infiltrate</td>
<td>No</td>
</tr>
<tr>
<td>Lang and Barritt, 2016&lt;sup&gt;11&lt;/sup&gt;</td>
<td>27, 3</td>
<td></td>
<td>CTG + EMD (2011)</td>
<td>F</td>
<td>24&lt;sup&gt;b&lt;/sup&gt;</td>
<td>NR</td>
<td>Mature, cortical bone with scattered osteocytes in lacunae</td>
<td>No</td>
</tr>
<tr>
<td>Francetti et al, 2019&lt;sup&gt;21&lt;/sup&gt;</td>
<td>26, 8</td>
<td></td>
<td>FGG (2008)</td>
<td>F</td>
<td>23, b 32–34&lt;sup&gt;b&lt;/sup&gt;, 43–45&lt;sup&gt;b&lt;/sup&gt;</td>
<td>Esthetic problem</td>
<td>Orthokeratinized epithelium, dense CT, no inflammatory infiltrates</td>
<td>NR</td>
</tr>
<tr>
<td>Sturque et al, 2019&lt;sup&gt;24&lt;/sup&gt;</td>
<td>48, 20</td>
<td></td>
<td>FGG (1998)</td>
<td>F</td>
<td>44&lt;sup&gt;b&lt;/sup&gt;</td>
<td>Pain, discomfort</td>
<td>Benign osteoadipogenic nodule</td>
<td>NR</td>
</tr>
<tr>
<td>Present report</td>
<td>48, 30</td>
<td></td>
<td>FGG (1989)</td>
<td>F</td>
<td>23, b 24&lt;sup&gt;b&lt;/sup&gt;</td>
<td>Discomfort</td>
<td>Mature, cortical bone with scattered osteocytes in lacunae, no signs of inflammation</td>
<td>No</td>
</tr>
</tbody>
</table>

CT = connective tissue; CTG = connective tissue graft; EMD = enamel matrix derivative; FGG = free gingival graft; NA = not applicable; NR = not reported; SG = skin graft.
<sup>a</sup>FDI numbering system.
<sup>b</sup>Surgical removal.

and/or esthetic concerns.<sup>10,12,18,19,21,24</sup>

Consequently, the removal of the tissue excess may be indicated in certain cases and in accordance with patient symptoms.

When focusing on histologic reports, these results corroborate several similar previous findings documenting healthy vital bone marrow with no signs of inflammation.<sup>10,18,19,21</sup>

The many resting lines in the bone matrix suggest a steady growth of bone with intermittent periods of rest. However, whether the bone exostosis continues to grow during life...
or whether this process may stop after a certain time should be further investigated.

Conclusions

The development of a bone exostosis following a mucogingival procedure is a rare clinical sequela of uncertain etiology, even though the most frequently discussed hypothesis is related to surgical periostral trauma (e.g., fenestration).

Acknowledgments

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