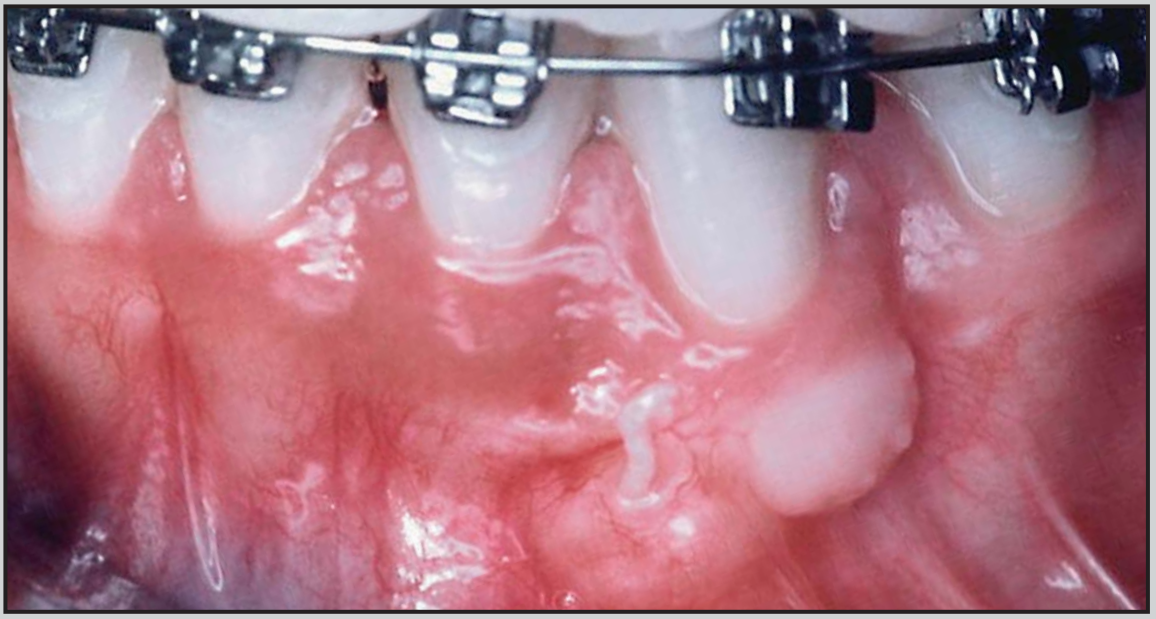
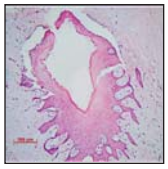


Epithelial Inclusion Cyst After Free Gingival Graft: A Case Report



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There have been few case reports of cystic lesions occurring after gingival grafting. There are three reports of this type of sequela after connective tissue grafting, but this is the first known case of a cystlike lesion developing secondarily to a free gingival graft procedure. The bulky tissue, which had developed under the previously grafted area, was properly excised under local anesthesia. The small specimen removed was sent for histologic analysis. A new gingival graft was performed immediately after the lesion enucleation, as the cystic lesion had dislocated the earlier graft. The surgical wound healed uneventfully, and no recurrence was seen 18 months later. The microscopic sections showed a cystic cavity lined with an orthokeratinized, hyperplastic, stratified squamous epithelium covered with fibrous connective tissue. The development of a cystlike lesion following a free gingival graft is, to date, an unpublished event. The fact that most cystic lesions appear in the mandibular lateral incisor–canine–first premolar area deserves further consideration. (Int J Periodontics Restorative Dent 2007;27:465–469.)

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The free gingival grafting technique is commonly used to predictably increase the width of the keratinized gingiva.[1,2] The guidelines, indications, and techniques for successful free autogenous gingival grafting were established by Sullivan and Atkins[3] nearly 40 years ago.

Although the surgical procedures for free gingival grafting are simple and usually well tolerated, some undesirable occurrences, both during and after surgery, have been reported in the literature. Complications such as excessive hemorrhaging from the donor site, problems related to the size or thickness of the graft, stability, color differences, bone exposure at the host site, problems with graft bonding, prolonged healing, and recurrent herpes lesions have been reported.[4] The appearance of bony exostosis in areas that earlier had been treated with free gingival grafts has also been reported.[5,6]

The occurrence of epithelial inclusion cysts or cystlike lesions is rare. In the literature, there have been three reports of this type of sequela after connective tissue grafting.[7–9] This is the first known case of a cystlike lesion

Fig 1 (left) *Preoperative view of mandibular left canine area demonstrating a lack of keratinized gingiva in the buccal aspect.*

Fig 2 (right) *Bulky tissue appearance at 11-month postoperative visit. Yellowish discharge, which was characterized as keratin, drained from the lesion.*

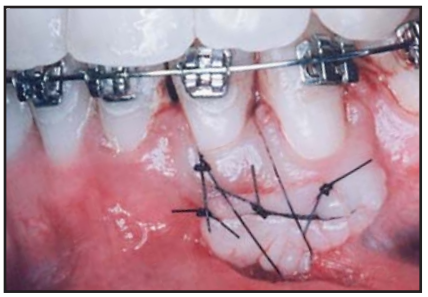

Fig 3 (left) *A new free gingival graft has been sutured into place with stretching and vertical stabilizing sutures.*

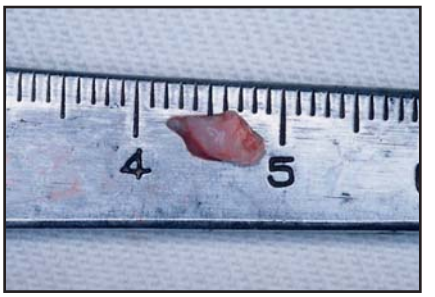

Fig 4 (right) *The small operative specimen.*

developing secondarily to a free gingival graft procedure, although in 1968, in a histologic analysis of five cases of free gingival grafting, Gordon et al[10] had mentioned the possibility of such an event.

Case report

The patient was a 22-year-old, healthy, nonsmoking woman who had been referred to the periodontal department by her orthodontist to increase the keratinized mucosa on the buccal aspect of the mandibular canines (Fig 1). Her medical history was noncontributory, and there were no contraindications for periodontal surgery.

A clinical examination showed that the patient used a fixed appliance; she had signs of gingivitis, and bleeding occurred on probing. She also had a substantial accumulation of plaque and calculus, although no pockets were deeper than 3 mm and there were no radiographic signs of bone loss. In the mandibular canine areas, a total lack of keratinized mucosa was observed as a result of orthodontic movement.

Therefore, basic periodontal therapy was carried out. This included oral hygiene instructions, ultrasound instrumentation, and scaling and root planing. In addition, to halt the recession and make further labial movement of the canines feasible, a free gingival grafting procedure was planned. This was considered because the technique is highly predictable and does not interfere with esthetics.

Initially, a free gingival graft was performed in the mandibular left canine area, and 5 months later, the mandibular right canine site underwent the same procedure. No complications such as hemorrhaging or infection were reported in either operation. Healing in both areas was normal.

During the follow-up visit, 11 months after the first surgery, a notable increase in the width of the gingiva on the right side was observed. However, there was a subtle tumefaction at the mesial aspect of the graft on the left site. On palpation, the tiny lesion was painless and a yellowish pasty secretion was drained. A clinical diagnosis of epithelial inclusion cyst was made (Fig 2).

A new gingival graft was performed immediately after the lesion enucleation, because the cystic lesion had dislocated the earlier graft (Fig 3). The material removed was sent for histopathologic examination (Fig 4). Microscopic sections showed a cystic cavity lined with an orthokeratinized, hyperplastic, stratified squamous

Fig 5 (left) *Photomicrograph of the cyst showing a cavity lined with an orthokeratinized, hyperplastic, stratified squamous epithelium covered with fibrous connective tissue.*

Fig 6 (right) *Site at 18 months after the second surgery.*

epithelium covered with fibrous connective tissue (Fig 5). These data confirmed the clinical hypothesis of an inclusion cyst occurring as a sequela of the surgical procedure. The repaired site healed normally, and no recurrence was seen 18 months later (Fig 6).

Discussion

Free autogenous gingival grafts typically are used to improve the width of keratinized gingiva, especially outside of the esthetic zone. This technique is simple, and the results are highly predictable.[11] Orthodontic labial movement is sometimes followed by bone dehiscence and loss of keratinized mucosa at the buccal aspect of the teeth.[12,13] This is an indication for the increase or reestablishment of the thickness of the gingiva because these areas become more susceptible to recession. In addition, the presence of a fixed appliance increases the accumulation of deposits and may affect periodontal health, especially in sites with thin gingiva.[14] Therefore, the present case was considered a good candidate for a free gingival graft because of its high predictability and because it was not situated in an area that could be affected esthetically.

Complications may occur after any surgical procedure and have been amply reported in the literature. Some cases of cysts or cystlike lesions developing after connective tissue grafts have been found over the last 10 years.[7–9] The occurrence of cystic spaces after autogenous masticatory mucosal grafting has not been noted in the literature. However, a microscopic study of grafted sites described epithelial invagination in one of the five cases studied.[10] According to Gordon et al, "the deep invagination of epithelium might have provided the basis for development of a cyst. It is impossible to know whether surgical procedures forced a strand of epithelium between the grafted tissue and the host site or whether an auto-marsupialization occurred."[10p133]

The presence of epithelial invaginations following graft procedures has also been reported by other authors. In the study by Ouhayoun,[15] it was shown that even connective tissue chemically separated from epithelium could develop epithelial invaginations at the graft-host interface, some of which could be so large that they could exhibit cystlike cavities.

These articles confirm that the physical presence of epithelial cells deep in the tissue is not necessary for the development of a cystic lesion. In fact, even if there is no deep epithelial island, such stimulation of the newly grafted connective tissue on the superjacent epithelium might occur, and this might invaginate into the graft. The worrisome question is: What stimulus would be necessary to cause the epithelial proliferation and development of a cystic lesion—surgical stimulation or self-stimulation?

Hence, a comparative analysis between the clinical and histologic aspects of gingival cysts in adults and these postsurgical cystic lesions would be appropriate.[16] Gingival cysts in adults are real developmental cysts that form from the remains of the dental lamina. They are considered rare and are noticed most often between the fifth and sixth decades of life, usually in incisive, canine, and premolar areas.[17,18]

Some reports present these lesions as entirely different from the histologic viewpoint, since gingival cysts in adults are lined with a thin epithelium of cuboidal or flattened cells with hyperchromatic nuclei, plaque formation, and isolated nests of clear cells. Furthermore, the histologic aspects of the "surgical" cysts would be more similar to the gingival cysts of the newborn, because of their acantholytic stratified squamous epithelium lining, with inflammatory infiltrate foci and the presence of keratin in their interior.[7]

However, other authors have reported epithelial linings of gingival cysts in adults of various types.[18] The most common type was a thin, flattened lining with or without localized thickenings (buds). Other types included nonkeratinized stratified squamous epithelium, keratinized stratified squamous epithelium, and parakeratinized epithelium with palisading basal cells. These reports confirm that the histologic aspect of gingival cysts in adults imitates those described as the "surgical cyst" by Breault et al.[7]

According to Ritchey and Orban,[19] possible sources of cystic formation are: *(1)* heterotropic glandular tissue; *(2)* degenerative changes in a proliferating epithelial peg; *(3)* remnants of dental lamina, enamel organ, or epithelial islands of the periodontal membrane; and *(4)* traumatic implantation of the epithelium. This should be considered as a reference on cyst development related to trauma or epithelial implantation, exactly like the gingival cyst that develops after a surgical procedure.

Epithelial tissue can be detected in 80% of connective tissue grafts. Nevertheless, the occurrence of cystic spaces from these epithelial islands is rare.[8] Thus, the presence of epithelial tissue alone does not cause cyst development; additional stimuli could be required.

Another interesting aspect to be considered is the common anatomic localization of both lesions. All the cysts that developed in grafted areas were in the mandibular incisive, canine, and premolar regions. This coincides with the most frequent location of gingival cysts in adults, although the relationship between these lesions cannot be explained.

Recently, Wei and Geivelis[9] described a new case of cystic formation after a connective tissue graft. The authors defended an anatomic correlation between gingival cysts in adults and cystic lesions that developed after grafting procedures. According to these authors, the inflammatory reaction during healing is able to induce cystic transformation of the preexisting remnants of dental lamina in the mandibular lateral incisor–canine regions. However, further studies are imperative to elucidate whether there is a true relationship between the pathogenesis of gingival cysts in adults and cystlike lesions.

Conclusion

The development of cystlike lesions as a sequela to free gingival grafts is rare. To detect this or other complications following a grafting procedure, an accurate examination is essential. This article described a cystlike lesion following a free gingival grafting procedure. This was excised surgically, and no recurrence was observed within an 18-month follow-up period.

Acknowledgments

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