

# Branchial cleft or cervical lymphoepithelial cysts

## Etiology and management

JEFFREY W. GLOSSER, D.D.S.; CARLOS ALBERTO S. PIRES, B.D.S.; STEPHEN E. FEINBERG, D.D.S., M.S., Ph.D.

**T**he branchial cleft cyst, or cervical lymphoepithelial cyst, is a unilateral, soft-tissue swelling that typically appears in the lateral aspect of the neck, anterior to the sternocleidomastoid muscle. It is clinically apparent in late childhood or early adulthood. The cysts, by definition, do not physically connect with the skin or aerodigestive tract.

At least four theories have been proposed regarding the origin of the branchial cleft cyst.

**Surgical excision of the branchial cleft cyst is considered curative, with recurrence unlikely if all remnants are removed.**

These include incomplete obliteration of branchial mucosa, persistence of vestiges of the precervical sinus, thymopharyngeal ductal origin and cystic lymph node origin.<sup>1-3</sup>

### SOURCE OF BRANCHIAL CYSTS

The branchial apparatus that begins to form in the second week of fetal life and is completed by the sixth or seventh week<sup>4</sup> is probably the structure most widely believed to be the source of

branchial cysts; however, definitive data remain elusive.<sup>3</sup> Proponents of this theory point to incomplete closure or incomplete obliteration of the fetal branchial arches, pouches or both as the source of the anomaly.<sup>5</sup> Specifically, the lack of degeneration of the cervical sinus created by the growth of the second arch over the third and fourth arches is the proposed cause.<sup>6</sup> The third and fourth arches thus overlaid by the second arch persist as small pockets with their ectodermal epithelium. These pockets usually fill in during fetal development; however, when they do not, cysts, sinuses and fistulas

**Background.** The cervical lymphoepithelial or branchial cleft cyst is a developmental cyst that has a disputed pathogenesis. The objective of this article is to provide a brief review of the literature and to define diagnostic terms related to this anomaly, as well as to describe its etiology, clinical presentation and treatment.

**Case Description.** The cervical lymphoepithelial or branchial cleft cyst usually presents as a unilateral, soft-tissue fluctuant swelling that typically appears in the lateral aspect of the neck, anterior to the sternocleidomastoid muscle, and becomes clinically evident late in childhood or in early adulthood. Clinicians can diagnose the cyst with appropriate imaging to assess the extent of the lesion before definitive surgical treatment. The authors describe a patient who underwent excision of a well-encapsulated cystic structure that was diagnosed as a branchial cleft cyst.

**Clinical Implications.** The cervical lymphoepithelial or branchial cleft cyst can be easily misdiagnosed as a parotid swelling or odontogenic infection. It is imperative that clinicians make an accurate diagnosis so that appropriate treatment (that is, surgical excision) can be performed. If the cysts are treated properly, recurrences are rare.

may arise later.<sup>7</sup> These anomalies represent fusional lesions.<sup>8</sup>

In normal human development, the branchial arches rise with cranial nerves V, VII, IX, X and XI. The branchiomic muscles include muscles of mastication; anterior belly of the digastric muscle; mylohyoid, tensor tympani and tensor palatine muscles; muscles of facial expression; as well as muscles involved with the hyoid bone and pharynx. Skeletal derivatives from the branchial apparatus include the ossicles, sphenomandibular ligament, styloid process, hyoid bone and laryngeal cartilages. The middle ear and eustachian tubes, the palatine tonsil, thymus and parathyroid glands are derived from the pouches.<sup>9-11</sup> It is of note

that even in the presence of a branchial cyst, the above structures usually develop without notable deformity or loss of function.

Several authors have discarded the branchiogenic theory in favor of the cystic cervical lymph node or salivary gland inclusion theory.<sup>2,12,13</sup> In 1949, King<sup>2</sup> concluded that changes within the cervical lymph nodes caused an inflammatory response leading to a lateral neck swelling, which he termed “lateral lymphoepithelial cyst.”

In 1959, Bhaskar and Bernier<sup>13</sup> examined 468 specimens submitted as branchial cleft cysts and interpreted 96 percent of them to be cystic changes in regional lymph nodes. The cystic change was caused by epithelial entrapment within the node at the time of development. In addition, these

authors noted that the parotid gland develops at the same time as the regional lymph nodes and speculated that this was the origin of the entrapped epithelium. Furthermore, Bhaskar and Bernier<sup>13</sup> observed that the size of the lesion often varied with concurrent infections in the patient. In view of their findings, they suggested the term “benign cystic lymph nodes” or “benign lymphoepithelial cysts.”

The controversy over the etiology of the swellings has given rise to a multitude of terms used (presumably) to describe the same lesion. This makes the study of these lesions more difficult.

## DEFINITIONS

We use the term “branchial cleft cyst” to refer to these lesions, which can be considered synonymous with the cervical lymphoepithelial cyst. Howie and Proops<sup>14</sup> defined the branchial cysts (or lymphoepithelial cysts) as “lesions found behind the angle of the mandible in the anterior triangle of the neck at the junction of the upper third and lower two-thirds of the sternocleidomastoid muscle. The cysts have a lining of stratified squamous epithelium resting on a complete or incomplete band of lymphoid tissue with part of the cyst wall resembling a lymph node.”

Several authors of case reports, however, have reported these lesions in other places. Golledge and Ellis<sup>3</sup> defined these cysts according to two parameters: position and histology. With regard to position, the lesion must lie outside the midline

of the neck or within any position in the lateral aspect of the neck. With regard to histology, the cyst lining is squamous or columnar and is surrounded by lymphoid tissue. The lymphoid tissue generally has a follicular pattern with germinal centers or a diffuse bandlike pattern.<sup>12</sup> The lining epithelium has been described as stratified squamous or low columnar, and evidence of keratinization may be seen when stratified squamous epithelium is present. Some authors have noted the presence of hair follicles and sebaceous and sweat glands within the cyst.<sup>2</sup>

Regardless of etiology, it is essential to understand the differences between a cyst, fistula and sinus. Although this may seem somewhat basic, the literature is replete with erroneous use of these terms.<sup>10,11</sup> This is especially troublesome when speaking of branchial cleft cysts because some have suggested that cysts are wholly different from fistulas and sinuses, in regard to their time of development, clinical presentation, histology and familial tendencies.<sup>9-11</sup> Some investigators doubt a common etiology between the cysts and sinuses. Many surgeons believe that congenital lateral cervical sinuses and fistulas result from the branchial apparatus; however, it is possible that many mechanisms may lead to branchial anomalies.<sup>3,7,11</sup>

Regarding age at presentation, Telander and Deane<sup>7</sup> found in their survey that sinuses and fistulas typically arise in the first decade of life and to a lesser extent in the second decade, whereas cysts occur in the adolescent and adult. Congenitally presented sinus tracts usually are the result of spontaneous or surgical drainage from a cyst and frequently have a familial incidence.<sup>5</sup> Microscopically, cysts are lined by stratified squamous epithelium lying on top of lymphoid tissue, with part of the wall resembling a lymph node.<sup>14</sup>

In contrast, sinuses and fistulas usually have a persistent opening to the exterior and muscular tubes lined with respiratorylike epithelium.<sup>10,11</sup> The pattern of inheritance is consistent with an autosomal gene having incomplete penetrance.<sup>15</sup> As suggested by Howie and Proops<sup>14</sup> and Chandler and Mitchell,<sup>12</sup> tubelike lesions should be called fistulae if they are open on each end, and sinuses if they are open only on one end. The definition of a cyst in this context is a closed pouch without

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opening to the skin, alimentary canal or pharynx.<sup>12,13</sup>

## PRESENTATION AND DIAGNOSIS

The branchial cleft cyst typically is a slow-growing, fluctuant mass that becomes apparent in the second and third decades of life.<sup>3,7,16</sup> A close association with the external ear, angle of the mandible and upper lateral aspect of the neck usually is observed, which is consistent with the most commonly accepted etiology (that is, coincident with the fetal location of the branchial apparatus). The swelling typically develops in the junction between the upper one-third and lower two-thirds of the anterior sternocleidomastoid muscle, but it can occur at any level from the hyoid to the suprasternal notch.<sup>16</sup> Chandler and Mitchell<sup>12</sup> described the location as being between the tragus and clavicle.

The cyst generally is movable in all planes and is motionless during swallowing.<sup>16</sup> Twenty to forty percent of patients relate its appearance to a recent upper-respiratory-tract infection, odontogenic infection or even to pregnancy.<sup>4,6,16,17</sup> If large enough, the anomalies can cause asymmetry of the neck, as well as dyspnea, dysphagia and dysphonia.<sup>4</sup> The swelling usually is unilateral, but cases of bilateral cysts/sinuses have been reported, with an incidence of 2 to 3 percent.<sup>15,17</sup> When bilateral cysts/sinuses develop, there seems to be a familial tendency.<sup>4,5</sup> Some believe there is an overall familial tendency for the development of unilateral cysts as well, but these authors seem to be in the minority.<sup>4</sup>

There is no sex predilection according to most observers,<sup>16</sup> but some researchers have noted that the cyst appears more commonly in males than in females.<sup>7,16</sup> In addition, no right- or left-sided tendency exists. Inflamed cysts may become abscessed, which eventually can lead to rupture. A permanent sinus may form or recurrent cyst formation and infection may occur.<sup>12</sup>

**Differential diagnosis.** The clinical differential diagnosis of these lesions includes tuberculous lymphadenitis, lipoma, cystic hygroma, carotid body tumors, thyroglossal duct cysts, metastatic neoplasms, lymphomas, suppurative lymphadenitis, branchial fistulas/sinus dermoid cysts, neurofibroma, hemangioma, lymphan-

gioma, teratoma, parotid neoplasm, ectopic salivary tissue, laryngocele and plunging ranula.<sup>4,5,13</sup> Clinicians must consider malignancies involving the lymph nodes, either primarily or secondarily.<sup>5</sup>

**Diagnosis of branchial cleft cyst.** The diagnosis of branchial cleft cyst is made primarily by medical history, clinical manifestations and exclusion. Preoperative ancillary diagnostic procedures include computed tomography, or CT, sonography and fine-needle aspiration, or FNA. CT may be particularly useful not only to visualize the full extent of the lesion, but also to delineate its association with adjacent structures.

These lesions are deemed amenable to sonography because of their typically superficial

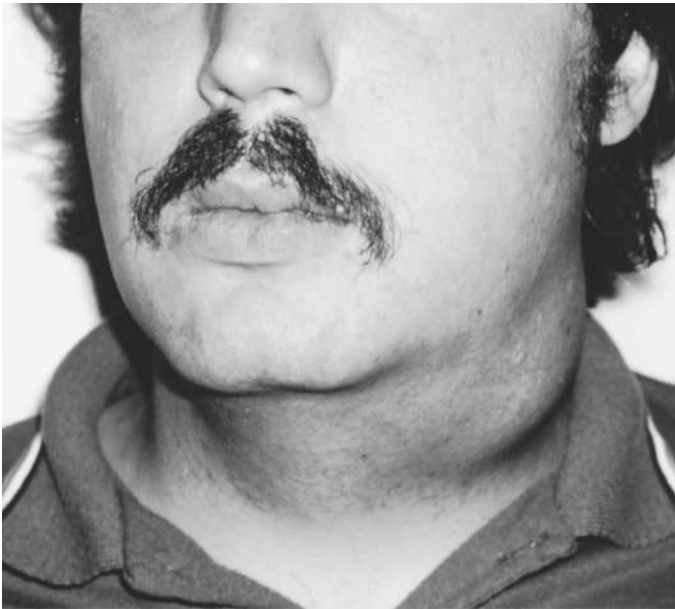
nature.<sup>18</sup> The sonomorphologic findings typically yield a rounded mass that has a uniform low echogenicity lacking internal septation, with no acoustic enhancement or motion.<sup>18,19</sup> This echogenicity probably is due to the accumulation of cellular material as well as cholesterol within the cyst lumen. Other sonographic reports have noted echogenicity in only the gravity-dependent portions of the cysts.<sup>18</sup>

The use of FNA has been mentioned in the literature as being effective in narrowing the diagnosis when a lateral neck lesion is present. Aspirate appears as a straw-colored fluid that microscopically may exhibit squamous cells, polymorphonuclear cells, lymphocytes and cholesterol crystals. Burgess and colleagues<sup>20</sup> conducted a study from which they determined that squamous-cell carcinoma could be recognized on FNA by observing an increased cellular nuclear/cytoplasmic ratio, irregularity of nuclear outline and nuclear hyperchromatism.

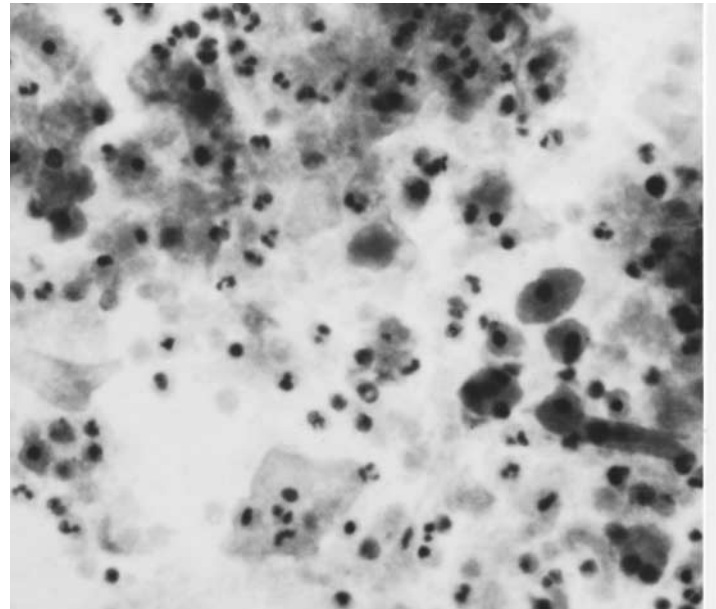
Branchial cleft cysts, on the other hand, exhibit benign squamous cells and mild nuclear atypia. Many researchers and clinicians believe that FNA can be an important adjunct to clinical diagnosis of lateral neck lesions, especially when attempting to categorize the swelling as benign or malignant. In addition, the procedure is quick and findings typically are available in a matter of hours rather than days. However, FNA is not a substitute for thorough, microscopic examination of the lesion.

Although FNA may result in a decrease in mass size, this improvement is temporary and is no substitute for excision of the cyst. Moreover, it

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**Figure 1. Clinical photograph of a fluctuant mass in the left aspect of the neck that had gradually increased in size during the previous five months. The lesion is 5 to 6 centimeters in diameter.**



**Figure 2. Cytopathologic examination findings of the fluid from the fine-needle aspirate, which showed polymorphonuclear cells, lymphocytes and squamous cells.**

can be difficult for a cytopathologist to distinguish a well-differentiated (that is, low grade) metastatic deposit of squamous cells from a benign lesion such as a branchial cleft cyst. This represents a potential pitfall for those who rely on cytologic results alone.<sup>20</sup> Branchial cleft cysts may have the ability to become malignant.<sup>21-23</sup> To date, though, there has been no report of such an occurrence. Because of their lymphoid nature, these cysts can be confused with a metastatic lymph node or a primary malignancy from the thyroid gland.<sup>21-23</sup>

#### TREATMENT

Surgical excision of the branchial cleft cyst is the treatment of choice and is considered definitive.<sup>4,24</sup> Some controversy exists about when to operate and whether every cyst requires removal. Many surgeons believe that presence of the lesion is reason enough for removal, primarily because of its propensity for infection.<sup>7</sup> Excision of an asymptomatic lesion precludes or minimizes the chance of infection complicating a later surgery.

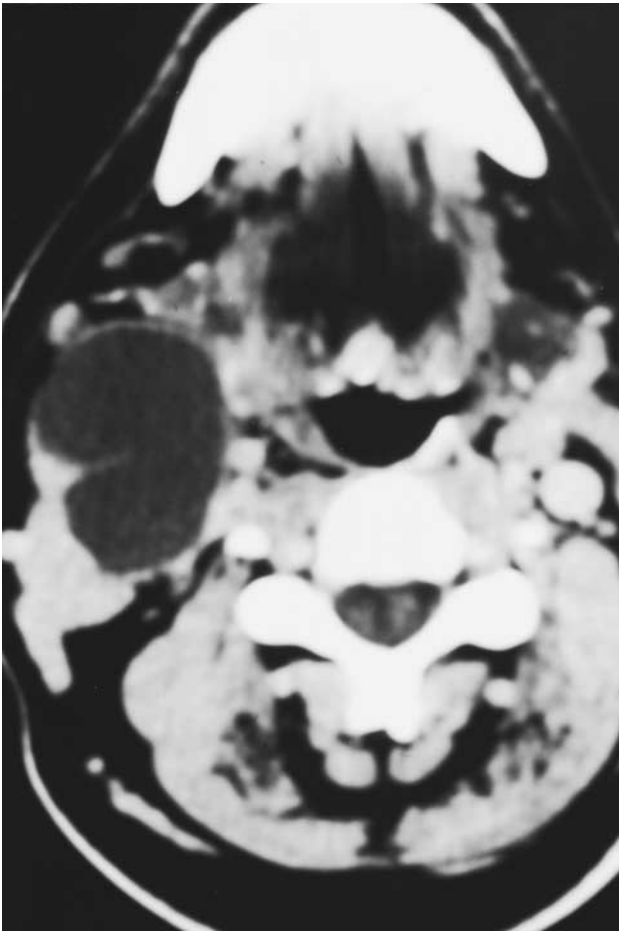
Any existing infection must be controlled before surgery can be performed, and can be achieved with antibiotics, with or without incision and drainage. Although incision and drainage may be

necessary to treat an infective episode before surgery, it is not recommended as definitive treatment.<sup>24</sup> Furthermore, some authors have suggested that complete drainage of lumenal contents before surgery is not desirable because this may make dissection more difficult.<sup>6,24</sup> Infection doubles the recurrence rate of these anomalies,<sup>12</sup> as do incomplete dissections and non-curative procedures.

Approximately 80 percent of branchial sinuses will open to the skin, and fewer will open to the pharynx. These sinuses initially may suggest a cyst, but on surgical exploration prove to be sinuses with a tract leading medially. Clinicians must take care to remove the entire tract to decrease the chance of recurrence. In particular, involvement of the deep aspect of the auditory canal has been attributed to an increased recurrence rate.<sup>24</sup>

Surgical complications include injury to surrounding structures such as the carotid sheath and the spinal accessory and hypoglossal nerves.<sup>16</sup> Other treatment modalities that have been reported are radiation therapy, repeated incision and drainage, and use of sclerosing agents.<sup>16,17,24</sup> These are considered noncurative, and if performed before surgery can increase the recurrence rate after surgical excision.<sup>12</sup>

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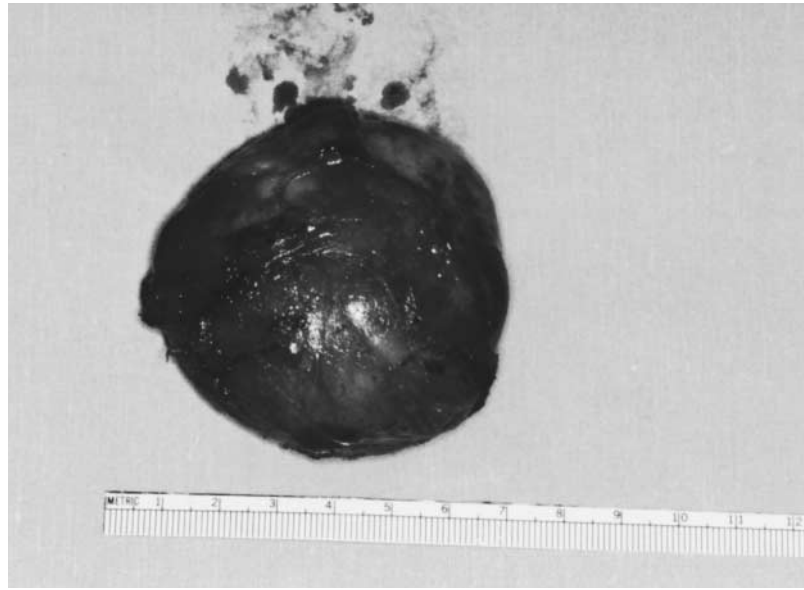
**Figure 3.** A computed tomographic scan shows a well-defined cystic lesion that was confined to the left aspect of the neck.

### CASE REPORT

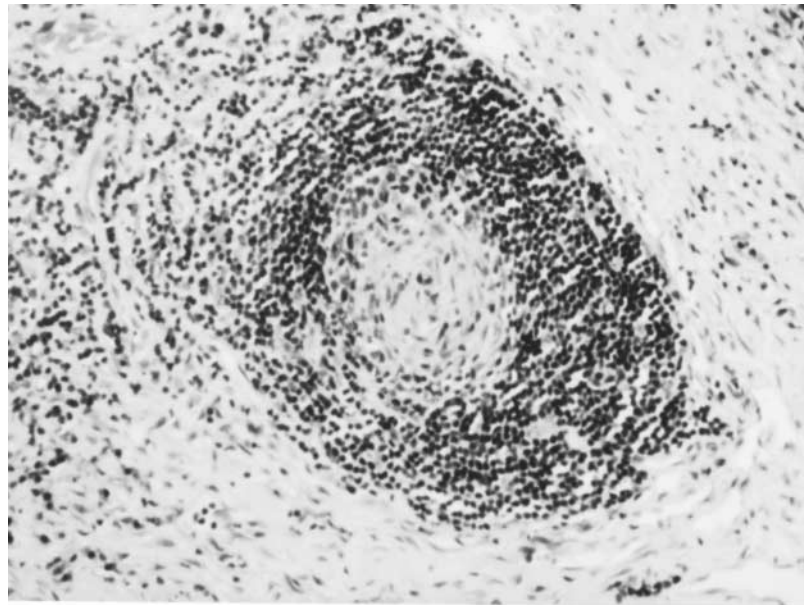
A 32-year-old man visited the oral and maxillofacial surgery clinic at the University of Michigan Medical Center, Ann Arbor, with a history of swelling that had gradually increased in size during the previous five months (Figure 1). The patient had no history of any previous swelling or infection within the head and neck region. His family's medical history was negative for developmental swellings.

Our clinical examination revealed a fluctuant mass within the left aspect of the neck that was approximately 5 to 6 centimeters in diameter. There was no clinical evidence of a sinus or fistulous tract.

A radiologist obtained a sialogram of the left parotid gland, which showed no abnormalities. We then performed FNA and removed 15 to 20 milliliters of straw-colored fluid. Histologic examination of the fluid revealed polymorphonuclear



**Figure 4.** The excised specimen shows an encapsulated cystic structure approximately 7 centimeters in diameter.



**Figure 5.** Photomicrograph of the wall of the cyst. The wall is composed of stratified squamous epithelium with evidence of underlying lymphoid aggregates.

cells, lymphocytes and squamous cells (Figure 2).

A CT scan of the neck (Figure 3) showed a well-defined cystic lesion that was confined to the left side of the neck. It appeared to be located deep to the investing fascia, as well as anterior to and as deep as the upper two-thirds of the sternocleidomastoid muscle.

The surgical procedure involved dissection and removal of a well-encapsulated cystic structure

(Figure 4). No fistulous tracts were noted to extend medially. Examination of the tissue resulted in a diagnosis of branchial cleft cyst or cervical lymphoepithelial cyst, which was consistent with the cytopathologic report findings. The wall of the cyst was composed of stratified squamous epithelium with underlying lymphoid aggregates (Figure 5). The patient's operative and postoperative course was uneventful. Long-term follow-up has shown no evidence of recurrence.

## CONCLUSION

The branchial cleft cyst, or cervical lymphoepithelial cyst, is a pathological entity whose etiology has yet to be delineated. It shares a clinical presentation with other pathological entities of the neck, making diagnosis difficult at times. Surgical excision of these lesions is considered curative, with recurrence unlikely if all remnants are removed. Dentists should be aware of this entity because it can be easily confused with an odontogenic infection or parotid pathology, especially if it develops high up in the neck. Clinicians should consider referring patients to a surgeon for diagnosis and treatment if no obvious dental or glandular pathology is seen as the cause of the swelling. ■

At the time this article was written, Dr. Glosser was a resident at the University of Michigan Medical Center, Ann Arbor. He now is in private practice in Berlin, Vt.

Dr. Pires is a postgraduate student, University of Michigan Medical Center, Department of Oral and Maxillofacial Surgery, Ann Arbor.

Dr. Feinberg is an associate professor, Department of Oral and Maxillofacial Surgery, University of Michigan Medical Center, 1500 E. Medical Center Dr., Ann Arbor, Mich. 48109-0018, e-mail "sefein@umich.edu". Address reprint requests to Dr. Feinberg.

The authors thank Carl M. Allen, D.D.S., The Ohio State University College of Dentistry, Columbus, for his review of the histopathologic specimens in this case.

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