

## A CASE REPORT

# INTRABONY LYMPHOMA SIMULATING PERIRADICULAR INFLAMMATORY DISEASE

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**M**alignant lymphomas are a heterogeneous group of immune system malignancies, classified as either Hodgkin's disease or non-Hodgkin's lymphoma. They account for nearly 5 percent of all malignancies in the United States and are the seventh most common cause of death from cancer.<sup>1</sup>

Lymphomas tend to affect lymphoid tissues and often spread to multiple sites, with a more predictable pattern of dissemination associated with Hodgkin's disease than with NHL. NHLs have a significantly greater propensity to affect non-lymphoid (extranodal) tissues, including oral tissues.

The NHLs originally were classified by their morphological features, but immunologic classifications were subsequently developed. An international multi-institutional study was conducted by the National Cancer Institute to compare the morphological classifications with biological behavior and in 1982, a Working Formulation for Clinical Usage was proposed that grouped the NHLs into low-grade, intermediate-grade and high-grade malignancies.<sup>2</sup> Today many malignancies are

### A B S T R A C T

**Intrabony lymphoma can mimic periradicular inflammatory disease of pulpal origin. Mismanagement of such cases can result if the clinician presumes the condition is inflammatory and fails to submit a tissue specimen for evaluation. This article reviews the features of jaw lymphoma and documents the importance of early diagnosis and histopathologic examination.**

classified as NHL, and the Working Formulation provides a relatively accurate method of predicting prognosis.

Treatment usually consists of radiation therapy, and/or chemotherapy, and prognosis is highly variable, depending on the type of lymphoma and the clinical extent of involvement (clinical stage).<sup>2</sup>

In a series of 1,467 cases of extranodal NHL, Freeman and colleagues reported that 28 percent affected the head and neck and 2 percent affected the oral cavity.<sup>3</sup> Takahashi and colleagues reported oral cavity

involvement in 8 percent of a series of patients with extranodal NHL.<sup>4</sup> Orally, NHLs tend to affect components of Waldeyer's ring, including the tonsils and the base of tongue, although all intraoral sites may be involved, including bone.<sup>5</sup> Eisenbud and colleagues reported that in 45 percent of lymphoma patients with oral involvement, the lymphoma originated in bone.<sup>6</sup>

As an intrabony disease, the NHLs can often simulate inflammatory disease and therefore could be confused with periapical disease of pulpal origin. We present a case of intrabony lymphoma which simulated periapical inflammatory disease and review its clinicopathologic features.

Because of the clinical, radiographic and occasionally histologic similarities between intrabony lymphomas and periradicular inflammatory disease, it is essential that an accurate diagnosis be established before endodontic therapy is instituted. If endodontic therapy is not successful, surgery would become necessary to obtain a biopsy and establish an accurate histopathologic diagnosis.

## CASE REPORT

A 37-year-old Caucasian male sought endodontic evaluation following root canal therapy of his mandibular right lateral incisor (no. 26) three months previously by his general dentist. The patient said he had been comfortable for about two months after the procedure; then, following what he perceived to be trauma from popped corn, no. 26 and no. 28 became tender. He was taking his second prescription of penicillin from his general dentist at the time of his initial visit.

Examination revealed that no. 26 and no. 28 were mobile and tender to percussion. Tooth no. 28 responded within normal limits to an ice test. Tooth no. 27 was within normal limits in all tests. The radiograph showed a diffuse radiolucency in the periapex of no. 26. Based

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on the radiograph, history and examination, a provisional diagnosis of periapical inflammation from either a second untreated canal or a root fracture was made. The past medical history was completely negative.

A follow-up was scheduled, but the patient returned before that appointment (about two weeks after the initial visit) because of increasing discomfort. Teeth nos. 25, 26, 27 and 28 were mobile and, with the exception of tooth no. 26, remained responsive to heat and

cold tests. There was swelling on the labial aspect of no. 26. There was no lymphadenopathy. The patient was placed on Augmentin (Smith-Kline Beecham Pharmaceuticals) 500 milligrams three times daily for 10 days and was scheduled for surgery.

At the time of surgery, a panoramic radiograph disclosed a relatively

well-circumscribed periapical radiolucency around tooth no. 26. The margins of the lesion tended to blend into the surrounding bone. Under local anesthesia, a full flap was reflected, exposing massive destruction of the labial plate of bone over nos. 25, 26 and 27 with some loss over no. 28. All interdental bone and some lingual bone was destroyed, and necrotic bone was present within the tissue.

Extensive curettage of the lesion from the area surrounding nos. 25 through 28 was done as well as a resection of the apex of no. 26, which seemed to be well-sealed. All tissue was submitted for histopathologic examination. The flap was

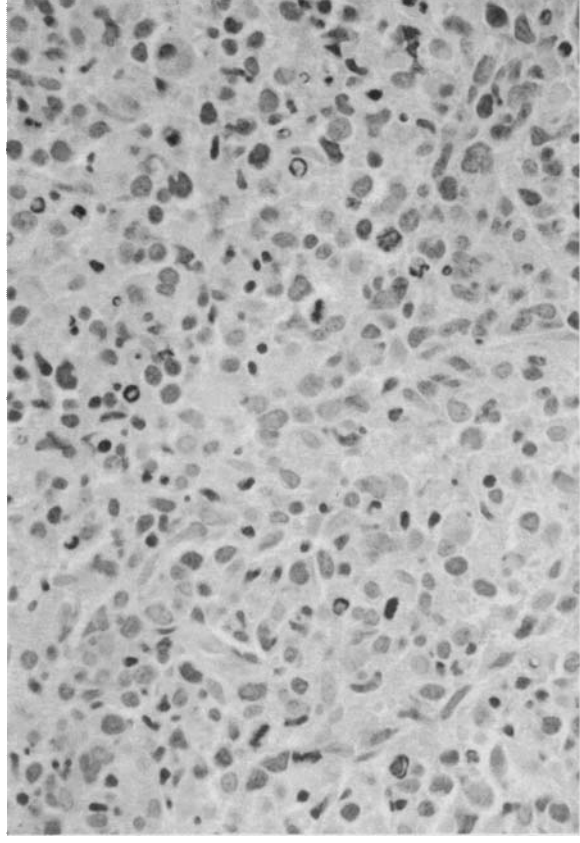


**Figure 1. Periradicular lucency on a radiograph of a patient complaining of pain following endodontic therapy.**

closed and a drain inserted into the defect. Flagyl (G.D. Searle & Co.) 500mg three times a day for 10 days was added to the Augmentin (SmithKline Beecham Pharmaceuticals).

The patient experienced a satisfactory postoperative course. The lesion was evaluated histologically by three oral pathologists, and a diagnosis of "atypical lymphoid infiltrate, highly suggestive of lymphoma" was rendered.

The patient was referred to an oncologist, and further testing of biopsy tissue confirmed a large cell, intermediate-grade lymphoma with a B cell phenotype. After the diagnosis was confirmed, the patient was evaluated for clinical staging.



**Figure 2.** Photomicrograph showing large, malignant lymphoid cells characterized by pleomorphism and mitoses (H&E stain, original magnification x 66.6).

Chest film, computed tomography scans, sonograms, bone marrow biopsy and meticulous physical examination did not detect additional involvement. HIV testing was negative and the patient was considered to have localized, Stage IE disease. The patient was started on a course of systemic chemotherapy, followed by local radiation therapy.

Six months after the surgery, on completion of chemotherapy and during the first week of radiation therapy, the patient was re-examined and a panoramic radiograph was taken. Clinical findings were completely negative. The soft tissues appeared normal, there was no probeable area around any of the involved teeth and no mobility of the teeth, and the patient was comfortable.

The panoramic radiograph showed no pathology in the mandible and complete resolution of the former lesion. Follow-up has continued for nearly 14 months with no

evidence of recurrent disease.

## DISCUSSION

Bone involvement by NHL is relatively rare and can lead to initial difficulties in diagnosis. In a review of 422 patients with malignant lymphoma of bone at the Mayo Clinic, Ostrowski and

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colleagues reported that 13 percent of those cancers affected the maxilla or mandible.<sup>7</sup> However, when one of the NHLs affects the oral cavity, it does have a propensity to involve bone: the jaws are affected 15 percent<sup>8</sup> to 45 percent<sup>9</sup> of the time in patients with oral NHL.

Jawbone involvement in NHL is well-documented, mostly in the form of case reports, although a few small series reports also are available.<sup>6,27</sup>

Patients commonly complain of pain and swelling,<sup>6,9,13</sup> features that may also suggest odontogenic infection. Less frequently, patients complain of loose teeth<sup>6,12</sup> and paresthesia or anesthesia.<sup>6,12,14</sup> Despite their malignant nature, these lesions also can be painless.<sup>6</sup>

The most common radiographic presentation of intrabony lymphoma is an ill-defined radiolucency that tends to blend into surrounding bone.<sup>6,9,15</sup> Steg and colleagues reported a series of 39 patients with jaw lymphomas: 22 of these cases demonstrated a destructive process radiographically.<sup>16</sup> The margins of the lesion are usually poorly circumscribed but on occasion may be well-circumscribed, a feature more suggestive of benign or inflammatory disease.<sup>8</sup> Occasionally the margins show sclerosis,<sup>17,18</sup> a feature indicative of a slowly enlarging process. However, it is also important to note that jaw lymphoma may show no radiographic features.<sup>12,19</sup> As a result, NHL may present at recent extraction sites in the absence of radiographic disease.<sup>20,21</sup>

It is not unusual for intrabony lymphomas to be mistaken for odontogenic infection, including periradicular pulpal disease.<sup>9,11,13,15,22,23</sup> Root canal therapy is often instituted but frequently fails. Vitality testing may be useful, but because many of the lesions produce pain, accurate vitality assessment is difficult.

Ellinger and Kelly reported a case of maxillary sinus lymphoma in which a tooth overlying the lucency tested nonvital in thermal and electric pulp testing.<sup>10</sup> The pulp was found to be necrotic, and the authors speculated that the

pulp may have become devitalized by the lymphoma. Although the most common features of intrabony lymphoma are also those of periradicular infection (pain and swelling) features of lymphoma that might alert the clinician to the possibility of malignancy include paresthesia, anesthesia and root resorption.

Mismanagement of patients with intrabony lymphoma results from a presumption that the pathologic changes are inflammatory. In such cases, the clinician often fails to submit a specimen for biopsy. The diagnosis of lymphoma can be established definitively only by biopsy and histologic examination. The neoplastic nature of the lymphoid infiltrate is usually apparent microscopically. On rare occasions, however, the initial biopsy can be misdiagnosed<sup>9,11</sup> because the cells of well-differentiated lymphoma are remarkably similar to normal inflammatory cells. Lymphomas also may become secondarily infected and inflamed, obscuring the neoplastic nature of the infiltrate. If a diagnosis of inflammation is rendered on a periapical

biopsy, the clinician should be alert to a lack of response to conventional root canal therapy. If the clinical disease progresses and symptoms persist, rebiopsy is mandatory.

Once the diagnosis of lymphoma is established by an oral biopsy, the patient must be referred for further medical evaluation and clinical staging (determining the clinical extent of the disease, with Stage I

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representing localized involvement and Stage IV disseminated disease). Lymphomas tend to spread and affect multiple sites. Jaw lymphomas, however, are typically limited to the jaw.<sup>6,8,24</sup> (Lymphomas confined to a single anatomic area and affecting an extranodal tissue are designated Stage IE, as in the case presented).<sup>25</sup> Some patients with Stage IE jaw lymphomas will develop more typical, multisite lymphoma with time while others never develop progressive disease.

The term *primary lymphoma of bone* has been applied to lymphoma confined to a single bone. This was originally described by Parker and Jackson in 1939.<sup>26</sup> It is now recognized that many NHLs may present as primary lymphoma of bone. When the lymphoma affects the jaws, the neoplasm is usually a high-grade B cell malignancy.<sup>7,23,27</sup> Although prognosis depends on the histologic type

of NHL, most patients with primary jaw lymphomas have a more favorable prognosis than patients with more conventional multifocal lymphoma. Five-year survival rates are around 50 percent.<sup>7</sup>

Any dental patient diagnosed with a high-grade or intermediate-grade lymphoma should be evaluated for HIV status because of the known association of NHL in AIDS patients.<sup>28-30</sup> Our patient tested negative, and his malignancy is apparently unrelated to immunosuppression.

**CONCLUSION**

This case report describes a dental patient who experienced a life-threatening intrabony lymphoma. Clinical staging indicated that the malignancy was confined to the jaw. Systemic chemotherapy and radiation therapy have resulted in a complete resolution of the malignancy. Although the patient is free of disease currently, additional time is needed to determine if this represents a cure. Early and accurate diagnosis by the dental profession has provided him with the best opportunity for long-term survival. ■

The opinions expressed or implied are strictly those of the authors and do not necessarily reflect the opinions or official policies of the American Dental Association or its subsidiaries.

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