Lymphangioma Mimicking Apical Periodontitis

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Abstract

Introduction: Lymphangiomas mimicking apical periodontitis, usually found in soft tissue of children and infants younger than 2 years, are extremely rare in aged patients, particularly in bone tissues. **Methods:** An 81-year-old woman was seen in a radiological service to undergo imaging studies for the placement of dental implants. A panoramic radiograph showed a radiolucent multilocular structure in the left mandibular molar region, where teeth #17 and #18 had previously been treated endodontically. A fracture in tooth #17 was detected. Because of the patient's clinical history and the characteristics of the image, which showed changes in trabecular bone structure, the patient was referred for cone beam computed tomography (CBCT) and magnetic resonance imaging (MRI) for further evaluation. CBCT scans demonstrated a well-circumscribed lesion immediately below the roots of teeth #17 and #18. MRI showed no involvement of the surrounding soft tissues, and the lesion affected only the mandibular bone. Incision biopsy was performed, and the tissue sample (histopathology specimen) was sent to microscopic evaluation. **Results:** Microscopically, there was a proliferation of vascular endothelium covered by long and mature endothelial cells. In the vessel lumens, there was eosinophilic material similar to lymph. The microscopic examination suggested the diagnosis of lymphangioma. **Conclusions:** Pathologies of non-endodontic origin such as lymphangioma, which might be in the area of the tooth apex, should be included in the differential diagnosis of apical periodontitis. Histopathologic examination is mandatory for their diagnosis and treatment. *(J Endod 2011;37:91–96)*

Key Words

Apical periodontitis, differential diagnosis, lymphangioma, oral disease, periapical diseases

Diagnostic accuracy is essential for treatment success, and the correct management of information obtained from the patient's history, clinical examinations, and complementary test results poses a great challenge (1). The definition of a diagnosis involves the establishment of a differential diagnosis (2), which should distinguish diseases of non-endodontic and endodontic origins. Radiolucent images in the mandibular or maxillary area surrounding the root apices might be a sign of non-endodontic disease and might lead to a misdiagnosis of apical periodontitis, particularly when the radiolucency is associated with an endodontically treated tooth. Several pathoses have been misdiagnosed as apical periodontitis (3–6).

Lymphangiomas are benign, slow growing lesions primarily characterized by the proliferation of lymphatic vessels in a part of the human body, usually the head and neck (7–12). It is still controversial whether such a lesion is neoplastic or hamartomatous. It usually appears at birth or in early childhood, before 2 years of age, and is classified as a focal proliferation of well-differentiated lymphatic tissue with a multicystic or sponge-like appearance. It is subdivided into 3 basic pathologic categories: lymphangioma simplex, described as thin-walled lymphatic channels that appear as small, well-circumscribed cutaneous lesions; cavernous lymphangioma, described as microscopic thin-walled lymphatic channels with an associated stroma; and cystic lymphangioma, described as large, well-circumscribed, multilocular cystic spaces lined by endothelium containing significant connective tissue components. Cavernous and cystic lymphangiomas might coexist within the same lesion (11, 12). They usually occur in the neck, skin, or other soft tissues of infants and children. Its clinical course is relatively benign (13). Although it usually occurs in early childhood and the beginning of adulthood, it might be a rare condition in elderly patients. This report describes the case of an 81-year-old woman who presented with a lymphangioma mimicking a periapical lesion around the roots of teeth #17 and #18.

Case Report

An asymptomatic 81-year-old woman was seen in an imaging service (RIO Radiological Institute, Brasília, DF, Brazil) in July 2008 to undergo radiographic assessment for the placement of dental implants. Intraoral examination showed that the mucosa was normal (Fig. 1). The panoramic radiograph showed a radiolucent multilocular image in the left mandibular molar region, where teeth #17 and #18 had undergone endodontic treatment, and a fracture in tooth #17 (Figs. 2–4).

The patient's history revealed that in 2004, tooth #18 had been treated endodontically (Fig. 3*A*) because of pulp inflammation. In 2005, also because of inflammation, a periapical radiograph was taken after the endodontic treatment of tooth #17 (Fig. 3*B*).

The patient did not complain of any pain, swelling, or any other symptom associated with the lesion. The analysis of the periapical radiographs revealed that the lesion

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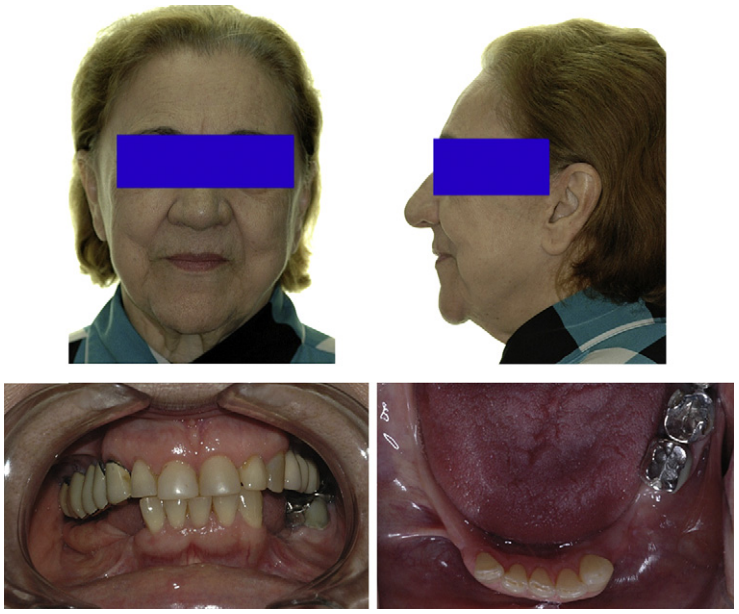

Figure 1. Intraoral and extraoral clinical photographs.

was already there at the time of the first endodontic treatment in tooth #18, but it was misdiagnosed as periapical inflammation as a result of endodontic problem and as a variation of trabecular bone structure. The panoramic radiograph requested before the placement of dental implants revealed the lesion's size and location. Cone beam computed tomography (CBCT) (I-Cat; Imaging Sciences International, Hatfield, PA) and magnetic resonance imaging (MRI) (Gyroscan T-5-II; Phillips Medical Systems International, Best, The Netherlands) were used to examine the lesion. CBCT scans showed a well-circumscribed lesion immediately below the roots of tooth #18 that extended from below tooth #17 to the mental foramen (Fig. 4). MRI showed no involvement of the surrounding soft tissues; the lesion had affected only the mandibular bone (Fig. 5).

A considerable hypodense area in the left side of the mandibular body affected the mental foramen area and extended back to the apex of the mesial root of tooth #17. The lesion approached the alveolar border, particularly in the region of tooth #19, and the lower cortex of the mandible. The multilocular aspect was seen only in regions surrounding the mental foramen (sections 108 and 110) and below the apex of the third molar (sections 134 and 136). There were remarkable irregular resorptions in the internal buccal and lingual areas, without expansions. A small rupture of the buccal cortex or

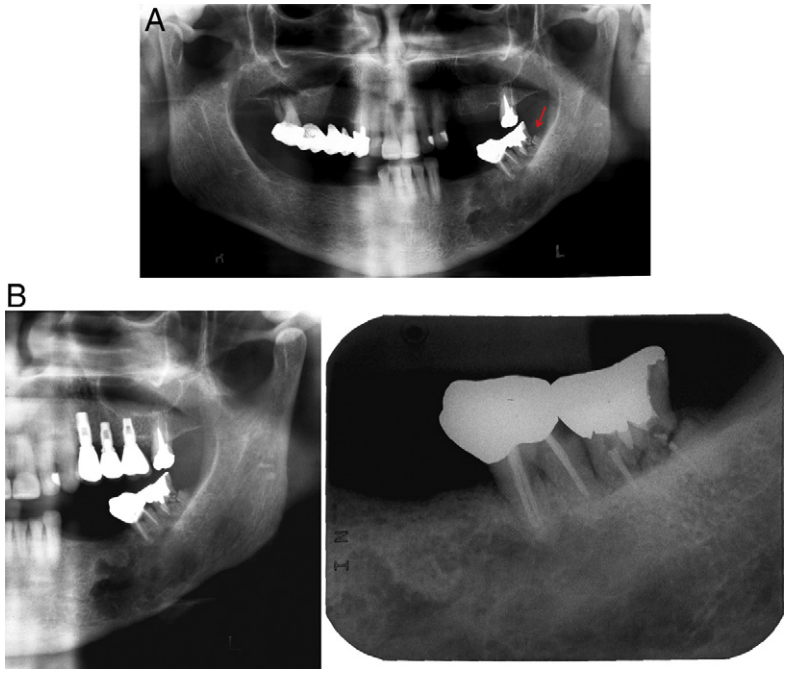

Figure 2. (*A*) Panoramic radiograph for placement of dental implants. A fracture is seen in the distal root of tooth #17 (arrow) in July 2008. (*B*) CBCT scan and periapical radiograph show fracture in distal root of tooth #17 in July 2008.

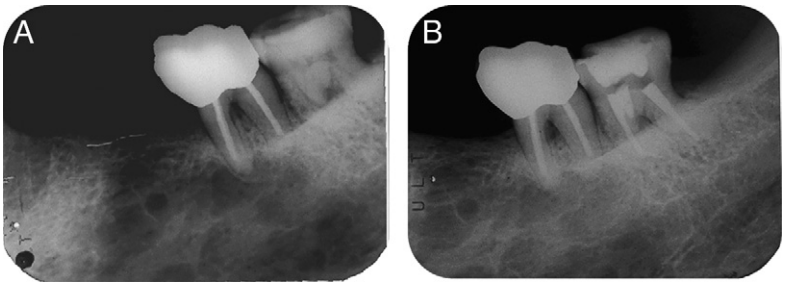

Figure 3. Periapical radiographs of tooth #18 at end of endodontic treatment (*A*) in 2004 and of tooth #17 (*B*) in 2005. Multiple radiolucencies in left mandibular molar region around tooth apices are present.

enlargement of the mental foramen was visible (sections 106 and 108). The cortex of the mandible canal was preserved and seen in several parts of the lesion. The images suggested the diagnosis of neoplasia.

The histopathologic exam is mandatory in these cases. By means of a small incision, a little flap was laid on the surface of the edentulous region's alveolar ridge (#19), and with a bur a thin bone layer was removed to obtain a sample of the lesion. Microscopic analysis of the specimen revealed proliferation of the vascular endothelium covered by long and mature endothelial cells. In the vessel lumina, there was eosinophilic material similar to lymph. The diagnosis was lymphangioma (Fig. 6). Because of the patient's age and her health condition, the clinical and radiographic control of the lesion was recommended and also the removal of tooth #17, which presented the root fractured. However, because of health issues, the patient came back to the clinic only in January 2010, 18 months later (Fig. 7). In preparation for surgery, an additional CBCT was taken to compare with the initial diagnostic image, so that the progression of the lesion could be evaluated. Although the surgical removal of tooth #17 was planned, it did not occur because the patient died in March 2010 of cardiac complications and iliac thrombosis.

Discussion

The lymphatic vascular system develops during the sixth week of life as an outgrowth of the venous system within mesenchymal tissues. It plays an important role in human circulation and organ perfusion homeostasis and is also relevant for protection against microorganisms and cells in the body itself. Lymphocytes are the most representative cells of functions and differences that are part of a complex system responsible, among other things, for immunity (14, 15).

Lymphangiomas are thought to result from congenital errors of lymphatic development (16, 17) and are classified as vascular lesions composed of dilated lymphatic channels. They are lymphatic cysts that became isolated during embryological development and failed to drain into the venous system. They might occur in cases of

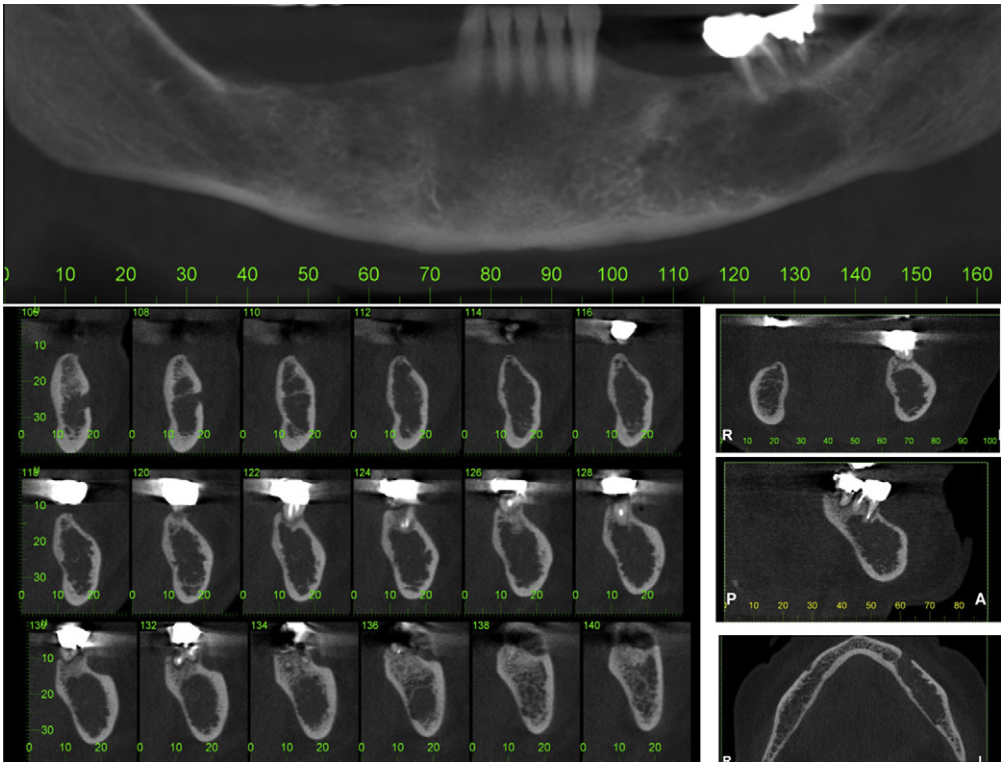

Figure 4. CBCT scan shows well-circumscribed lesion (panoramic, coronal, sagittal, and axial reconstructions) in July 2008.

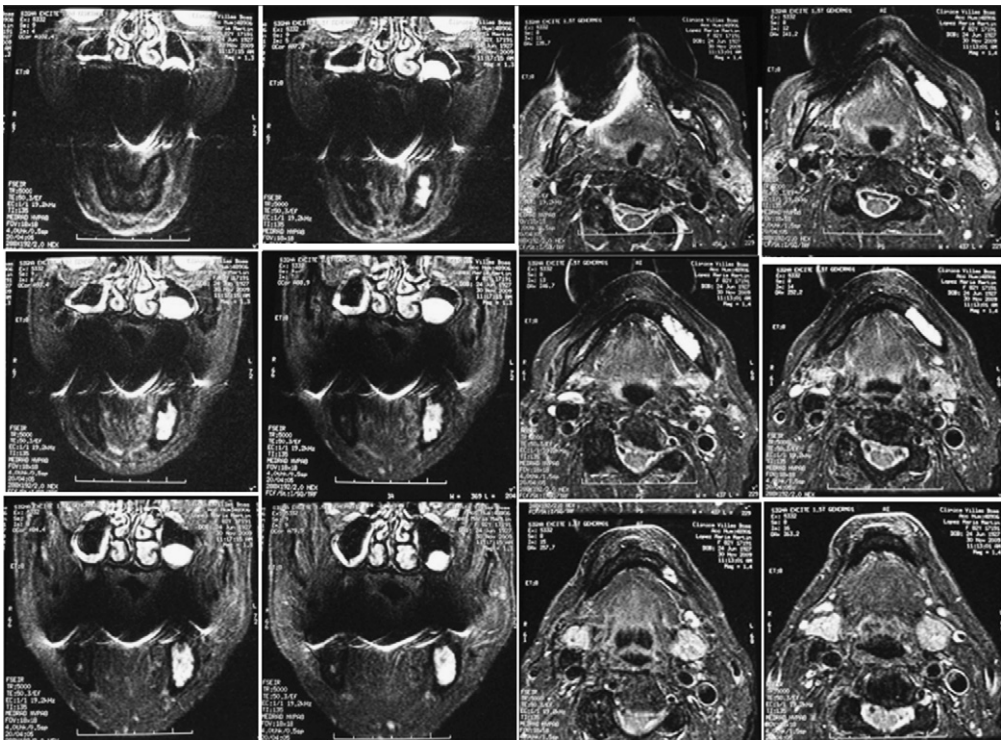

Figure 5. MRI confirms no involvement of soft tissues; lesion affected only mandibular bone.

Turner syndrome and in other malformation syndromes; their existence in abortuses has also been recorded (18).

This disease is sometimes fatal in infancy or early childhood, especially when associated with other complex pulmonary and cardiac anomalies. The onset of its clinical manifestations might be delayed into adolescence or early adulthood in patients with a mild form of the disease (19). However, it is a rare finding in adult patients, especially among the elderly (20). According to some authors, occurrence in adult life is significantly lower than in infancy (21), and the most common type in adult life is superficial cutaneous lymphangioma (lymphangioma circumscriptum) (22, 23). Although they might affect almost any part of the body served by the lymphatic system, they have a particular predilection for the head, neck, and axilla, sites that account for nearly three fourths of all lymphangiomas. There have also been reports of their occurrence in various parenchymal organs such as lungs, spleen, liver, and bone (24).

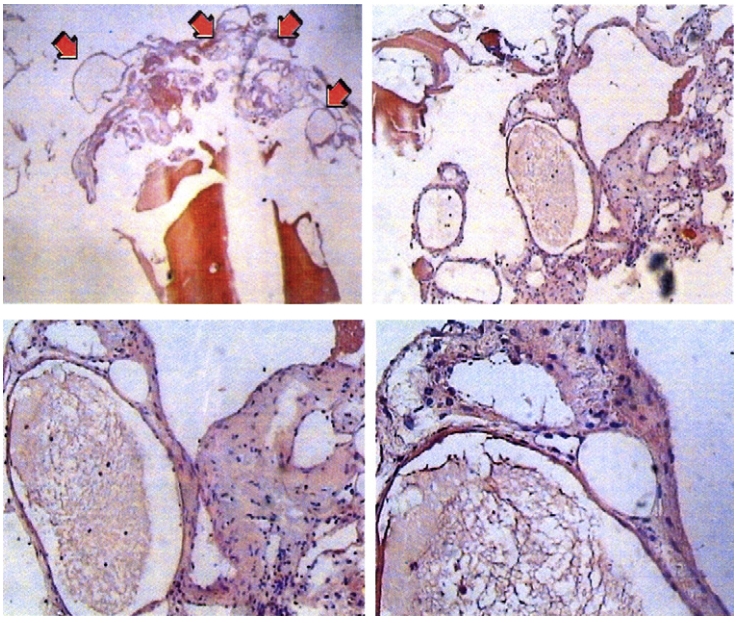

Figure 6. Histopathologic findings show proliferation of vascular endothelium covered by long and mature endothelial cells. Eosinophilic material similar to lymph is seen in vessel lumens. Diagnosis was lymphangioma.

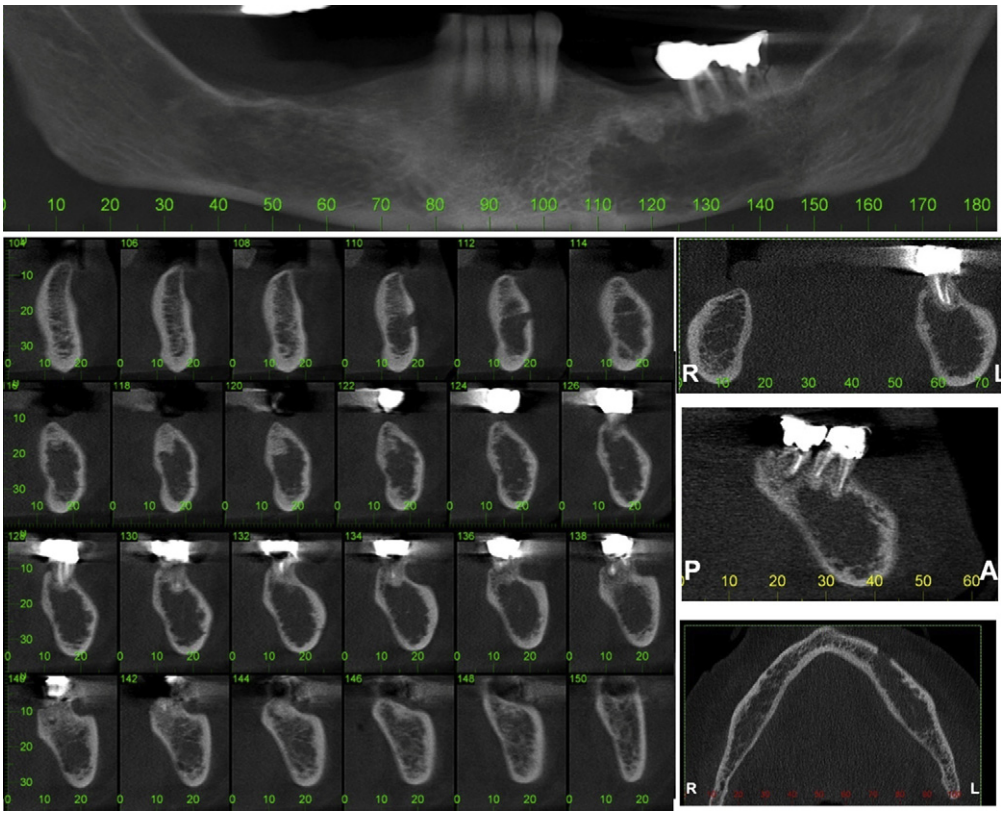

Figure 7. CBCT scan: panoramic, coronal, sagittal, and axial reconstructions show slow progression of lesion in January 2010.

The intraosseous lymphangioma is extremely rare, and when it affects the periapical area, it can be easily misdiagnosed as a periapical lesion such as a granuloma or an odontogenic cyst. Its progression is usually slow, particularly when it affects elderly patients whose cellular activity is lower. Patient history, clinical examination, pulp vitality tests, and follow-up of endodontically treated teeth are essential for an endodontic diagnosis. Endodontists should remember that periapical lesions might be of endodontic or non-endodontic origin. Many osteolytic lesions have radiographic features that are similar to those seen in cases of intraosseous lymphangiomas, such as cherubism, simple bone cyst, aneurysmal bone cyst, early stages of periapical cemento-osseous dysplasia, and desmoplastic fibroma of bone (7, 8, 25). The diagnosis of the periapical lesion associated with endodontic infection does not seem to be difficult, especially because periapical lesions are very common findings in endodontic routine. Periapical lesions appear as a result of root canal infection associated with inflammation and gradual destruction of the alveolar bone and are very often diagnosed by endodontists.

Diseases of non-endodontic origin such as lymphangioma, which might be associated with the tooth apex, should be included in the differential diagnosis of apical periodontitis. All resources that aid in the definition of the diagnosis should be used. This case could be followed up because of the lesion site and the patient's health condition and age. However, other treatment options have been described in the literature for each particular case (7, 9, 26).

Lymphangiomas are rarely associated with tooth apex diseases, and their diagnoses are not often made in endodontic practice. CBCT and MRI are important imaging exams, and histopathologic examination is mandatory to establish a correct diagnosis and treatment.

Acknowledgments

The authors deny any conflict of interest.

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