PEDIATRIC DENTISTRY/Copyright © 1986 by
The American Academy of Pediatric Dentistry
Volume 8 Number 3

CASE reports

Multiple lymphangiomas of the alveolar ridge in a neonate: case study

Stephen Wilson, DMD, MA, PhD Alan R. Gould, DDS, MS
Carolyn Wolff, DMD

Abstract

A case report of bilateral lymphangiomas of the mandibular alveolar ridge in the neonate is presented. This case is of interest in that it constitutes the first report of multiple lymphangiomas occurring within a single dental quadrant.

Lymphangiomas generally are regarded as hamartomas and as such represent a developmental malformation of lymphatic vessels. The most common intraoral site of lymphangiomas is the tongue, which when so affected may be enlarged and referred to as macroglossia.[1,2] Lymphangiomas are classified as simple, cavernous, cellular, diffuse systemic, and cystic lymphangiomas or hygroma.[2] Lymphangioma histologically consists of numerous enlarged lymphatic vessels lined with endothelium. These channels usually are fitted with protein-rich fluid with only a few blood cells.[1]

Lymphangiomas of the alveolar ridges in neonates were first described by Levin et al. in 1976.[3] In their study of 2092 infants, it was noted that only 55 (3.7%) manifested lymphangiomas of the alveolar ridges. The lesions were observed to occur only in blacks with a definite predilection for males. The lymphangiomas were limited anatomically to the posterior aspects of the maxillary ridge and to the posterior lingual surface of the mandibular ridge. The authors indicated that a majority of affected infants had multiple lesions, with the most common distribution being bilateral on the mandibular alveolar mucosa. In a similar series of cases, Jorgenson et al.[4] noted that 4% of black neonates had alveolar lymphangiomas ranging in size from 1 to 9 mm in diameter. No cases were noted in white infants and there was a 2:1 male-to-female distribution. Most of the neonates had 1 or 2 lesions, the latter being more common.

Interestingly, no cases have been described in any series in which there were multiple lesions confined to any single quadrant. The present report is, to the authors' knowledge, the first in which multiple lesions in a single quadrant of the mandibular alveolar ridge are observed. This also represents the first report in the dental scientific literature of lymphangioma of the alveolar ridge of a neonate.

Case Report

The male patient was born after 37½ weeks of gestation to a 16-year-old black female via vaginal delivery. The birth was without incident, and the child was the mother's first born. At birth the child weighed 6 pounds, 10 ounces and the only congenital anomaly noted was a small "mucinous" cyst on the right mandibular alveolar ridge. Following a normal neonatal course in the hospital, the patient was released with the mother.

Five days following release from the hospital, the patient was seen in the hospital emergency room with a complaint of diarrhea and formula intolerance. In the subsequent 3-week period, the patient was seen several times in a private office setting with continuing difficulties secondary to formula intolerance. It was noted that the lesion of the right side of the mandible had persisted, and that a new lesion apparently had developed on the inner aspect of the left mandibular alveolar ridge. The mother of the patient had indicated that the lesions appeared to fluctuate in size. Referral to the pediatric dentistry advanced education program was made by the attending physician who registered concern regarding the relative rapid enlargement of the lesions. Following consultation with the pediatrician, it was decided to take the patient to the operating room for the purposes of excisional biopsy. This decision was based on the pediatrician's physical evaluation of the pa-

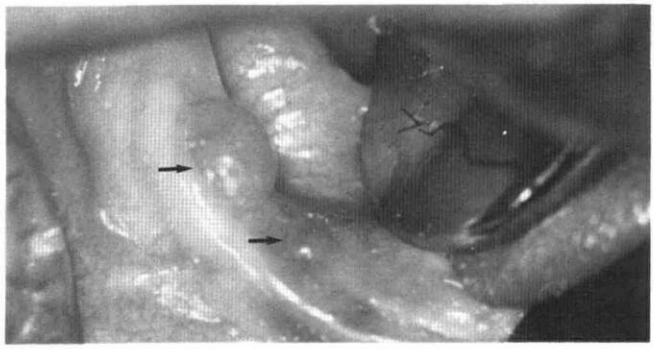

FIG 1. Photograph of right mandibular alveolar ridge demonstrating 2 lesions (arrows) on the lingual aspect of the ridge. The tongue has been retracted to the right. Photograph taken prior to removal of larger, more distal lesion in the operating room.

tient for undergoing general anesthesia and the potential patient risk/benefit associated with local anesthesia vs. general anesthesia, especially in light of the small operative field, and the need to obtain a definitive histological diagnosis of the lesion.

General anesthesia was induced in the operating room and a nasotracheal tube was inserted. At this time 2 smaller lesions were noted, each occurring mesial to the lesions noted earlier. It is not known whether these lesions had developed recently, or instead were overlooked initially due to their small size. All of the lesions exhibited a cystic, fluid-filled, bluish translucent appearance, with the largest measuring approximately 1 cm in diameter (Fig 1). The overlying mucosa appeared normal, and there was no clinical evidence of inflammation. It was decided to remove the posterior lesion of the left mandibular ridge for immediate microscopic examination. The excision extended to the depth of the alveolar bone. Frozen section differential diagnosis indicated fibroma or lymphangioma. The lesion located most posterior on the right side of the mandibular alveolar ridge then was excised and placed in 10% neutral buffered formalin. The apparently newer, more mesially-placed lesions were not removed because of potential interference with the extensive surgical excisions and feeding. It was decided instead to observe the fate of the new lesions, as similar lesions had been noted to resolve spontaneously in previous instances. The patient recovered from general anesthesia without incident and was released into the care of the parent. The patient will be followed periodically.

Microscopic Findings

Excised formalin-fixed tissue was dehydrated in ascending grades of alcohol, infiltrated with paraffin, sectioned at 4 μm, and stained with hematoxylin and eosin. Light microscopic examination of the excised tissue revealed a benign proliferation of delicate, thin-walled lymphatic structures exhibiting a discontinuous endothelium lining (Fig 2). The endothelial cells were flattened with a uniform, hematoxylinophilic nucleus (Fig 3). The endothelium was supported by thin, fibrous connective tissue cores, with the tissue demonstrating an alveolar pattern. The spaces enclosed by lymphatic structures generally appeared empty, containing only scattered lymphocytes along with a sparse, finely fibrillar eosinophilic material (Fig 3). Occasional rests of odontogenic epithelium were observed and were interpreted as dental lamina rests (Fig 4). The tissue was covered by a well-formed oral surface epithelium with an underlying thin lamina propria (Fig 2). The diagnosis was neonatal lymphangioma of the alveolar ridge.

Discussion

Lymphangiomas of the alveolar ridge of neonates first were reported in 1976 in a study involving more

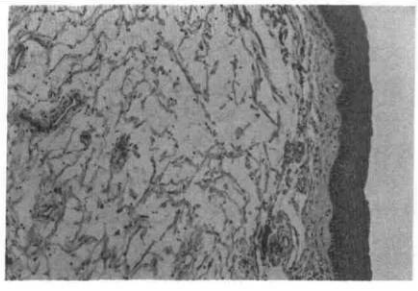

FIG 2. Photomicrograph demonstrating numerous thin-walled lymphatic structures arranged in an alveolar pattern. The overlying surface epithelium and lamina propria are well formed (H&E stain, original magnification, 63×).

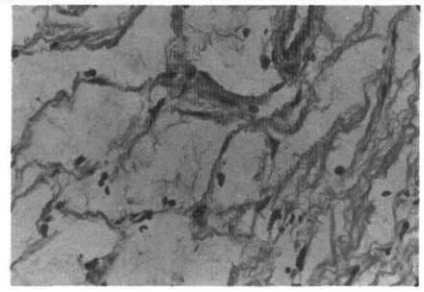

FIG 3. A discontinuous layer of flattened endothelium lines thin, connective tissue cores. Scattered lymphocytes and sparse, lightly fibrillar material are apparent within the lymphatic spaces (H&E stain, original magnification, 250×).

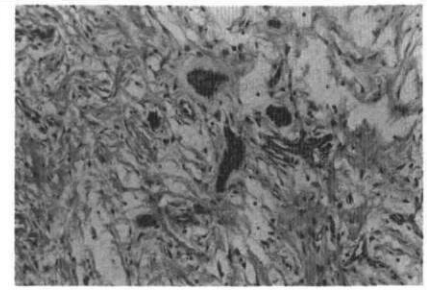

FIG 4. A cluster of odontogenic epithelium rests is present within the lymphatic proliferation (H&E stain, original magnification, 100×).

than 2000 infants.[3] A follow-up to the original article appeared in 1982,[4] reinforcing the recognition, concerning this lesion, of several distinctive characteristics. Lymphangiomas of the alveolar ridge of neonates have not been reported as occurring in the Caucasian population, and in the black population the male-to-female distribution was approximately 2:1. The lesions often are found on the posterior lingual aspect of the mandibular alveolar ridge and multiple lesions per patient are not uncommon.[3] However, this case report is the first to describe multiple lesions in a single quadrant. Levin et al.[3] indicated that their population consisted of neonates from birth to 5 days of age. Although the age of those in the second report was indicated to be well-baby neonates,[4] it may be that secondary lesions of alveolar lymphangiomas form in affected individuals at a slightly older age, as the patient in this case was noted to have additional lesions present at least 5 weeks postpartum. Some cases reportedly have resolved within the first 6½ months of life.[3] It is most likely that the majority of these lesions resolve spontaneously, as there are no reports of lymphangioma of the alveolar ridge in young children. There has been a recent report describing lymphangiomas of the gingiva in a teenage patient.[5] However, the duration of the lesions was unknown. Further, the described histologic features were quite dissimilar from those associated with lymphangioma of the alveolar ridge of the neonate.[3]

The etiology of the lymphangioma of the alveolar ridge of the neonate is unknown, but probably involves developmental factors. Histologically the lesion seen in this report was similar to those described by Levin et al.[3] in that there was a benign proliferation of endothelium-lined lymphatic structures loosely supported by thin, fibrous connective tissue cores. In addition, the lymphatic channels were observed to contain scattered lymphocytes and a finely fibrillar material. Rests of odontogenic epithelium also were noted in the presently reported case and have not been described previously in connection with this condition. Such rests in oral soft tissue from the alveolar ridges are a relatively common finding and, in itself, of minor significance.

The clinical presentation of lymphangioma of the alveolar ridge of the neonate, though distinctive, nonetheless shows some similarity to several other conditions—dental lamina cyst of the newborn, eruption cyst in the newborn, mucous retention phenomenon (mucocele), and congenital epulis of the newborn. It is not surprising then that examples of lymphangioma of the alveolar ridge in the neonate probably have been misreported as dental lamina cyst,[7] eruption cyst,[8,9] or mucous retention phenomenon.[10] Clinical photographs describing each of the above-listed cases are strongly suggestive of lymphangioma of the alveolar ridge. In no instance are the histologic features of these cases described, thus precluding precise confirmation of the diagnosis. This notwithstanding, the clinical features of lymphangioma of the alveolar ridge of the neonate are sufficiently distinctive as to generally permit accurate clinical separation of this lesion from dental lamina cyst, eruption cyst, mucocele, and congenital epulis of the newborn. Coupled with microscopic assessment, a precise diagnosis is always possible.

Dental lamina cyst of the newborn typically presents as multiple, pale nodules affecting buccal and lingual aspects of the alveolar mucosa in both anterior and posterior quadrants.[2] Microscopic features include a thin squamous epithelium lining with desquamated keratin occupying the cyst lumen.

Eruption cysts have been reported in the newborn,[11,12] presenting as fluctuant, bluish swelling of the alveolar mucosa. Significantly, these lesions always are associated with erupting teeth, and have been found only in the mandibular incisor region in this age group.

Though bearing some clinical resemblance to the alveolar ridge lymphangioma, the mucous retention phenomenon rarely, if ever, is found arising from attached alveolar mucosa or gingiva. This observation is no doubt related to the absence in this region of accessory salivary gland tissue.[13] The mucocele histologically is composed of extravasated mucus, granulation tissue, and inflammatory cell infiltrates. In many cases, portions of accessory salivary gland are observed histologically in the surgical specimen.[14]

The congenital epulis of the newborn is characterized by an 8:1 female-to-male distribution with a marked predilection for the anterior maxillary gingiva.[15] In contrast to the soft, cystic bluish appearance of lymphangioma of the alveolar ridge, congenital epulis of the newborn has been described as a pink, firm, and often lobulated mass, ranging from a few millimeters to 4 cm in diameter. The histologic findings are equally distinctive, consisting of sheets of finely granular, plump eosinophilic cells.[15]

Comparison of lymphangioma of the alveolar ridge to "typical" lymphangioma discloses some interesting comparisons. Both conditions may occur congenitally and present as bluish, cystic swellings. However, typical lymphangioma shows no sex or racial predilection, and is capable of reaching considerable size, producing significant enlargement or distortion of the tongue, neck, or other soft tissue structures, necessitating appropriate treatment.[2] Recurrence following attempted removal is common, and spontaneous regression is rare.[2] In contrast, several examples of lymphangioma of the alveolar ridge in the

neonate apparently have resolved without treatment.[3]

Dr. Wilson is an assistant professor of pediatric dentistry, The Ohio State University; Dr. Gould is an associate professor in diagnostic services, University of Louisville; and Dr. Wolff is a pediatric dental resident, University of Louisville and Kosair Children's Hospital. Reprint requests should be sent to: Dr. Stephen Wilson, Pediatric Dentistry, College of Dentistry, The Ohio State University, 305 W. 12th Ave., Columbus, OH 43210.

1. Brightman VJ: Benign tumors of the oral cavity, including gingival enlargements, in Burket's Oral Medicine, 8th ed, Lynch MA, Brightman VJ, Greenberg MS, eds. Philadelphia; JP Lippincott Co, 1984 p 323.
2. Shafer WG, Hine MK, Levy BM: A Textbook of Oral Pathology, 4th ed. Philadelphia; WB Saunders Co, 1983 pp 159-60, 268-69.
3. Levin LS, Jorgenson RJ, Jarvey BA: Lymphangiomas of the alveolar ridges in neonates. Pediatrics 58:881-84, 1976.
4. Jorgenson RJ, Shapiro SD, Salinas CF, Levin LS: Intraoral findings and anomalies in neonates. Pediatrics 69:577-82, 1982.
5. Josephson P, van Wyk CW: Bilateral symmetrical lymphangiomas of the gingiva. J Periodontol 55:47-48, 1984.
6. George DI Jr, Gould AR, Behr MM: Intraneural epithelial islands associated with a periapical cyst. Oral Surg 57:58-62, 1984.
7. Peters RA, Schock RK: Oral cysts in newborn infants. Oral Surg 32:10-14, 1971.
8. Robinson HBG: Cysts of the oral cavity, in A Manual of Oral Surgery, 2nd ed, Archer WH, ed. Philadelphia; WB Saunders Co, 1956 p 297.
9. Gorlin RJ: Cysts of the jaws, oral floor, and neck, in Thoma's Oral Pathology, Vol 1, 6th ed, Gorlin RJ, Goldman HM, eds. St. Louis; CV Mosby Co, 1970 p 449.
10. Einhorn AH: The mouth, in Pediatrics, 17th ed, Rudolph AM, ed. Norwalk, Connecticut; Appleton-Century-Crofts, 1982 p 860-61.
11. Clark CA: A survey of eruption cysts in the newborn. Oral Surg 15:917, 1962.
12. Rushton MA: A malformed tooth associated with an eruption cyst at birth. Br Dent J 94:254-56, 1953.
13. Dale AC: Salivary glands, in Oral Histology: Development, Structure, and Function, ten Cate AR, ed. St. Louis; CV Mosby Co, 1980 p 305.
14. Standish SM, Shafer WG: The mucous retention phenomenon. J Oral Surg 17:15-22, 1959.
15. Fuhr AH, Krogh PHJ: Congenital epulis of the newborn: centennial review of the literature and a report of case. J Oral Surg 30:30-35, 1972.

Academy Changes Printer

Due to continuing difficulties with the former printer of the *Journal,* the Academy has made a change to Allen Press, Incorporated of Lawrence, Kansas.

The Academy already has joined a cooperative subscription catalog of scholarly and scientific journals which is published by Allen and distributed to libraries and subscription agencies around the world.

Because of the many stylistic requirements, reprint details, back order inventories, and transfer of fulfillment lists, this September issue of *Pediatric Dentistry* is late in delivery. The editorial staff apologizes for any inconvenience to readers. The December issue should be nearly on time, and by the March, 1987, issue we expect to be back to our normal delivery schedule.