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An oral hemangioma in a three-month-old child: clinical report

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Abstract

A rare presentation of an oral hemangioma in a 3-month-old child is reported. The lesion was gray, began central in bone, and expanded rapidly into the oral cavity.

The hemangioma is a tumor of mesenchymal origin. It is found occasionally in the oral cavity.

The cellular hemangioma is one of a family of tumors characterized by the formation of vascular tubes of endothelial cells. The tumors are differentiated from one another by the predominant cell type found on histologic examination.

Borrmann[1] first used the term hemangioendothelioma in 1899. Later, Mallory[2] used the term to describe a malignant vascular tumor. Stout[3] described the histologic features of hemangioma in 1944. A review of the literature reveals a variety of names which were used for tumors which exhibited vascular channels of endothelial origin. These include hemangioma, lymphangioma, angiosarcoma, hemangiosarcoma, angioendothelioma, and angioblastoma.[4-7] This varied terminology has led to a confusion in diagnosis and treatment of these tumors. Several authors have attempted to clarify this situation.[8,9] Shklar and Meyer[10] differentiate between an "extremely cellular benign hemangioma" and the hemangioendothelioma, which they prefer to refer to as an angiosarcoma. Others, however, feel that the term angiosarcoma is too nonspecific, referring simply to a sarcoma of blood vessels without reference to cellular origin.[9] They have advocated the term hemangioendothelioma prefaced by a term identifying the clinical behavior of the tumor, for example, benign or malignant.[8] In addition, a distinction has been made between the malignant hemangioendothelioma and the infantile form of hemangioendothelioma. This type is felt to be benign but can be fatal due to local extension from one or several of the tumors.[9] The malignant form also has been documented in infants and children.[7,11] Shafer[12] points out that the currently preferred term for the lesion previously designated juvenile hemangioendothelioma is cellular hemangioma. The authors subscribe to this latter view. The following is a clinical report of a rare presentation of an oral hemangioma which began central in bone and expanded rapidly into the oral cavity.

Clinical Report

A 3-month-old boy was referred to the Department of Oral and Maxillofacial Surgery, University of Florida, College of Dentistry, for evaluation of a soft tissue mass located on the left mandibular ridge. The lesion which first was noticed by the parents when the child was approximately 10 days old, was described as being a small bluish-appearing nodule, about a centimeter in diameter. The family pediatrician observed the mass for about 2 months, but due to its increasing size, and obvious discomfort and feeding difficulties, the child was referred for further evaluation and treatment. The child had a normal medical history with no unusual findings in relation to the mother's pregnancy, labor, or delivery. His family history was noncontributory and on physical examination he appeared to be healthy and of normal size and weight. Physical findings included multiple cutaneous, exophytic hemangiomas on the right upper eyelid, neck, abdomen, groin and feet, ranging in size from 1 mm to 3 cm (Fig 1). The patient had an additional hemangioma in the right subaxillary region which was removed at age 5½ weeks. An enlarged liver was noted on physical examination and

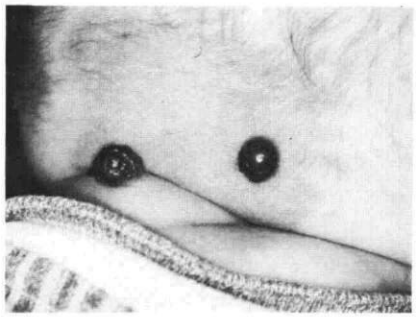
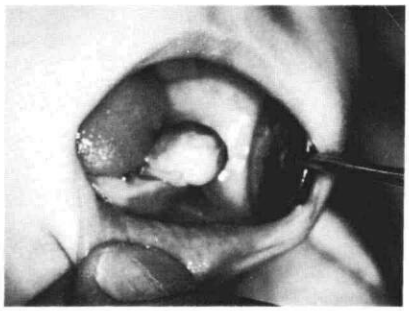


FIG 1. (*left*) Hemangiomas on child's neck.
FIG 2. (*right*) Appearance of pedunculated soft tissue, mandibular lesion on initial exam.



CAT scan and ultrasound studies demonstrated a multinodular lesion of the liver's right lobe which was suspected to represent hemangiomatous involvement of this organ also. The remainder of the general physical examination was within normal limits.

Oral examination demonstrated a 2.5 × 2.0 cm pedunculated soft tissue mass on the left mandibular alveolar ridge (Fig 2). The surface appeared to be smooth and hyperkeratotic with a gray hue that blanched slightly under pressure. The lesion was firm to palpation and fixed to the underlying bone.

The remainder of the oral examination was essentially normal. No positive cervical lymphadenopathy could be detected.

A CAT scan of the area demonstrated an osteolytic defect in the left mandible (Fig 3). Results of thorough laboratory studies did not reveal any abnormal values.

The patient was taken to the operating room twice, initially for an incisional biopsy and then for complete excision of the lesion (Fig 4). Due to the vascular nature of this lesion and the potential for severe hemorrhage with a central hemangioma of bone, the initial incisional biopsy was immediately followed by application of a cryosurgical probe to control bleeding. This particular lesion, however, was cellular and the bleeding was minimal. At the time of the complete excision of the lesion, local anesthetic with a vasoconstrictor was infiltrated around the lesion (adequate transfusable blood was available) but as was

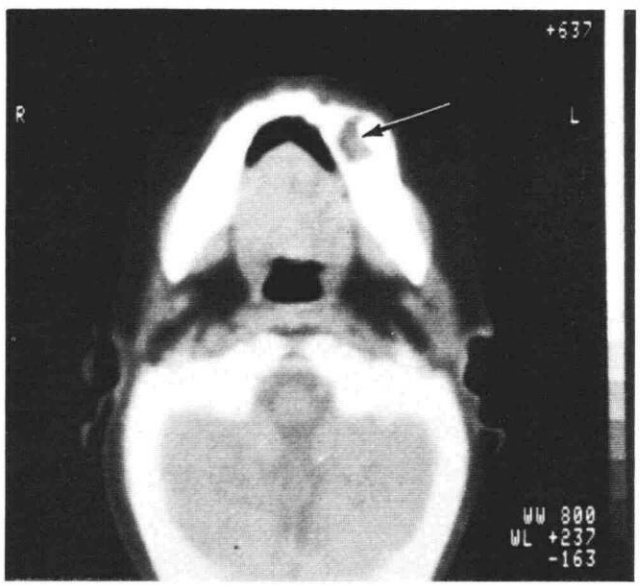


FIG 3. CAT scan showing osteolytic defect.

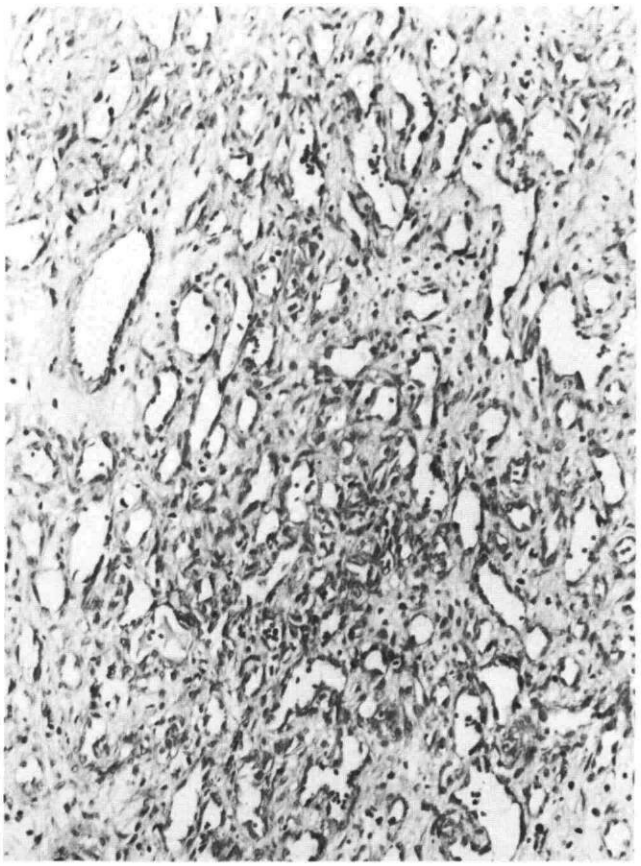


FIG 4. Surgical specimen *en toto*.

expected due to the minimal bleeding during the biopsy, there was little hemorrhage during lesion removal. Careful curettage was sufficient to separate the tumor from the mandible and the lesion was removed *en toto* (Fig 4). The minimal oozing which occurred after the removal of the specimen was controlled easily with pressure. The residual bony defect was not felt to be large enough to justify a grafting procedure. Postoperative care consisted of gentle irrigation of the bony defect to keep it free from debris. Histopathologic examination disclosed the lesion to be a cellular hemangioma (Fig 5). Continued follow up since the procedure has demonstrated normal healing of the area. A short course of prednisone therapy at a dose of 2 mg/kg/day, and low-dose radiation have been suggested for the liver involvement.

Discussion

The clinical appearance of this tumor is similar to that of many other vascular tumors. The hemangioma typically presents as a red or bluish red, slightly raised lesion, moderately firm to palpation. It will ulcerate and possibly hemorrhage if traumatized. They may rarely enlarge rapidly, causing local deformity and loss of function. This patient's lesion fell into this category and also involved bone which made clinical diagnosis more difficult.

Though it is usually painless, there are several reports of pain associated with vascular tumors.[14-16] Infants with oral lesions often develop difficulty in feeding due to the enlarging size of the tumor.[4,7,17-19]

Surgery is the most widely advocated treatment of hemangioma. However, surgery, radiation, and chemotherapy have been employed alone and in combination to treat vascular neoplasms with varying degrees of success.[8]

The extraoral hemangiomas were of the classical strawberry type due to their having less connective tissue than the more mature variety — thus their very red color. Ninety-five per cent of these lesions spontaneously regress with age. If they are located in areas of chronic irritation or need to be removed for cosmetic reasons, simple excision, cautery or cryosurgery can be used.[13] There are 3 types of hemangiomas which appear most frequently in children.[13] The small, isolated, focal hemangiomas which are unrelated to any apparent genetic transmission. This patient's extraoral hemangiomas appear to be of this type. A second variety are the multiple, focal, petechia-sized hemangioma associated with Rendu-Osler-Weber disease. The third type is the large, grossly disfiguring, unilateral hemangiomas associated with Sturge-Weber syndrome.

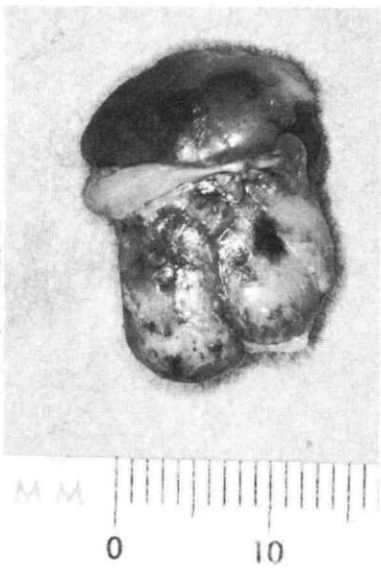


FIG 5. Photomicrograph showing proliferating endothelial cells and associated vascular channels typical of cellular hemangioma.

Summary

The cellular hemangioma is a rare tumor of the oral cavity. A clinical report of this lesion occurring in a 3-month-old child has been presented. The lesion was gray in color, central in bone, and expanded rapidly into the oral cavity. This unusual appearance and clinical behavior made the initial diagnosis a difficult challenge.

Dr. Laskin is an associate professor and Dr. Lawrence is a resident, oral and maxillofacial surgery, University of Florida, College of Dentistry, Gainsville, FL 32610. Reprint requests should be sent to Dr. Laskin.

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Pioneers in pediatric dentistry: Lyle Smith Pettit

Dr. Lyle Smith Pettit was born in Wisconsin, February 18, 1905. He attended The Ohio State University and in 1929 received the DDS degree from the College of Dentistry.

Following graduation he began a general practice of dentistry in Columbus, Ohio. His love for treating children, however, began to take preference and his practice evolved into a specialty practice of dentistry for children. He participated in postgraduate courses at the University of Michigan and University of Pennsylvania.

In 1936 he organized and developed a program of dentistry for children at The Ohio State University and at the Children's Hospital in Columbus. He also developed a dental program for the handicapped in the Department of Physical Medicine at Ohio State and a program of dental services for the United Cerebral Palsy Organization.

He was one of 44 dentists who attended the 1944 organizational meeting of the American Academy of Pediatric Dentistry in Ann Arbor, Michigan, and served the academy as a member of the Board of Directors and as its fifth president.

Dr. Pettit is a member of the Ohio State Dental Association, a life member of the ADA, Honorary member of the Ohio Society of Dentistry for Children, member of ASDC, American Association of Dental Schools, Fellow of the American College of Dentistry, Omicron Kappa Upsilon, and since 1969 has been a Kentucky Colonel.

His civic involvement includes membership in the Upper Arlington Civic Association and a volunteer to the Senior Center of Upper Arlington where he received two awards for faithful services.

He is coauthor of a *History of the Ohio Dental Association*, and has been asked to write a history of the dental programs at Children's Hospital. Since 1972 he has been Professor Emeritus at The Ohio State University.

Dr. Pettit lives at 2004 Berkshire Rd., Columbus, Ohio. He and his wife Anita have three children.