Central Hemangioma: An Overview and Case Report
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Abstract
Central hemangioma of the mandible and maxilla is extremely rare. Although the mucosal and soft tissue lesions are readily suspected by their clinical appearance, the intrabony lesions may be difficult to distinguish visually. The clinical and radiographic presentation is relatively nonspecific; therefore, a proper diagnosis has to be made. The management of central hemangioma is difficult because of the abundant vascular network in this region. Various therapies have been considered, but surgery has most frequently been used. A case report of an 8-year old boy with central hemangioma of the left mandibular body with vague clinical features but a characteristic radiographic and histological picture of central hemangioma is presented here. Also presented is a literature review that includes: (1) clinical features; (2) radiographic appearance; (3) histology; and (4) possible treatment modalities. Enbloc resection of the mandible was performed, followed by the insertion of a reconstruction plate, which will later be replaced by an autogenous graft. (Pediatr Dent 2006;28:460-466)

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Central hemangiomas of bone arise from vessels within the marrow spaces and may comprise arterial and venous vessels. Microscopically, engorged vascular sinuses are present with an endothelial lining supported by a connective tissue stroma interspersed by bony trabaculae, which are usually arranged at right angles to the surface. This characteristic appearance is called the “sunburst” or “soap bubble appearance.” These tumors may also sometimes exhibit elements of: (1) fibrous connective tissue; (2) osseous tissue; and (3) cavernous blood vessels.

Central hemangioma is a great mimicker, as it resembles: (1) osteosarcoma; (2) fibrous dysplasia; (3) central giant cell granuloma; (4) ameloblastoma; (5) multiple myeloma; (6) dentigerous cyst; and (7) odontogenic cyst radiographically. It also may clinically mimic: (1) a central arteriovenous fistula; (2) aneurysms; or (3) a shunt. It is a relatively rare condition, but may pose a lethal risk to the patient. Therefore, the patient’s history, radiographs, and other investigations are all important in fabrication of a final diagnosis and treatment plan.

Clinical findings
A patient with a central hemangioma may show little or no evidence of any intrabony lesion. Duration of symptoms before patients seek care can vary from 1 week to 10 months. It has been reported that there is no characteristic clinical sign, but the most common finding was a firm, nonpainful, bony swelling sometimes associated with a subjective pulsating sensation or throbbing discomfort.1,2,4 The swelling may...
be minimal or may cause gross facial asymmetry. Pain is an uncommon complaint, though on occasions it has been a factor in causing the patient to seek treatment. There have been cases when the pain associated with hemangioma radiates to the: (1) ear; (2) temporomandibular joint; or (3) mandibular condyle. Parasthesia of involved nerves is also reported, but it is not a regular finding. When the lesion involves a dentulous segment of bone, mobility of the adjacent teeth may occur. Some other dental findings include: (1) derangement of occlusion; (2) displacement of teeth; (3) suprareruption; (4) premature exfoliation of primary teeth; and (5) early eruption of permanent teeth as a result of the pressure of expanding lesion.3,4

**Radiographic features**

The radiographic appearance is definitely not pathognomonic, and only a working diagnosis of central hemangioma of the bone can be made from radiographs as it simulates other numerous bony lesions. Langland et al9 reported that the presence of a parallel or tube-like arrangement of radiopaque striae is an important indicator of hemangioma. The lesion is seen as an area of altered radiodensity, usually osteolytic and occasionally with central radio-opaque areas and an altered trabecular pattern. Worth10 described a pattern whereby the trabeculae are arranged in a manner resembling the spokes of a wheel, radiating from the lesion’s center towards the periphery. Patchy, multicycic osteolytic areas may give the lesion a soap bubble or honeycomb appearance. Many authors have also described the classical sunburst or sunray appearance, which is caused by coarse trabeculae perpendicular to the bony surface.11

Nearly any combination of lesion shape, location, or pattern can develop. There are, however, a greater number of variations and combinations of changes that may lead the clinician to suspect a vascular lesion. Careful evaluation of radiographs is therefore emphasized for detection of such lesions. Nagpal et al reported a case of central hemangioma of the mandible with numerous variations in the radiographic appearances in various areas of the lesion in different projections.12 In some cases, the lesion produces an alteration in a trabecular pattern, which may be present in some areas and lost in others.

The periphery can either show a well-defined or ill-defined corticated area with scalloped margins. Unilocularity, multilocularity, and heterogeneous degree of radiolucency are commonly reported radiographic variations that are associated with a: (1) honeycomb; (2) sunburst; (3) soap bubble; or (4) tennis racket appearance.13 Included in the radiographic differential diagnosis are: (1) ameloblastoma; (2) giant cell lesion; (3) myxoma; (4) dentigerous cyst; (5) sarcoma; (6) fibrous dysplasia; (7) multiple myeloma; (8) aneurysmal bone cyst; and (9) simple dental granuloma or cyst.3,11-13

Osteosarcomas produce a similar sunburst appearance. The lesion may be: (1) entirely radiolucent; (2) mixed radiolucent-radiopaque; or (3) radiopaque. There may be asymmetric broadening of the periodontal ligament space, however, and onionskin growth of the periosteal bone.

Central hemangiomas with areas of increased radiopacity can be mistaken for a ground glass appearance of fibrous dysplasia. Other radiolucent patterns of central hemangioma can be misdiagnosed as intermediate stages of fibrous dysplasia.

Multilocular regions of rarefaction that accompany expansion and thinning of the cortex may mimic giant cell granuloma of the mandible and maxilla. The loculations produced by central hemangioma, however, are accompanied by a fine fibrillar network.

Aneurysmal bone cyst may be suspected, but radiographically this lesion does not resorb the adjacent teeth and lacks the multiple noncorticated foraminae that are sometimes seen in central hemangioma.

Central hemangioma, like multiple myeloma, may also present with punched-out skull lesions highly characteristic of the latter. But mandibular lesions of multiple myeloma are well delineated without a cortical outline, and perforation of the cortex is more common than expansion as seen in hemangioma.

Since hemangioma of bone occurs early in life, the lesion can be in close proximity to the erupting teeth and, hence, can be mistaken for a dentigerous cyst. But in contrast to hemangioma, a dentigerous cyst will be associated with a corticated pericoronal radiolucency attached to the cementoenamel junction of the impacted tooth.

The central hemangioma of bone may be clinically indistinguishable from other vascular conditions, such as shunts or aneurysms. Thus, the ambiguity of the radiographic picture makes a diagnosis by this means alone virtually impossible.12

**Histology**

The histology of hemangioma is diagnostic. The microscopic picture is that of a proliferating mass of endothelial cells forming a plexiform arrangement of vascular spaces, which are either: (1) capillary; (2) cavernous; or (3) mixed.14 The thin-walled cavernous spaces are lined by a single layer of endothelial cells interspersed among bony trabaculae. The capillary type demonstrates fine capillary loops that tend to radiate outwards in a sunburst pattern.

Hirzot15 has suggested that the clinical and histological features may be a function of 3 different stages seen during the development of hemangiomas:

1. an early stage in which the lesion is highly vascular;
2. an intermediate stage that exhibits a blood clot present in cystic areas; and
3. a terminal stage in which various stages of ossification are demonstrated.

**Diagnosis**

A definitive diagnosis from the clinical and radiographic features may not be possible without a biopsy. Removal of tissue for microscopic examination, however, carries with
it the risk of uncontrollable hemorrhage and should be avoided. Angiography has proved to be a useful diagnostic tool when the features are prompting a diagnosis of hemangioma. This will be helpful in demonstrating the pressure of the vascular lesion as well as delineating the boundaries and arterial connections. Aspiration and analysis of the lesion’s contents can provide significant diagnostic information without the risk of hemorrhage.

**Treatment modalities**

Since hemangioma of the mandible and maxilla is a benign tumor, the greatest hazard is exsanguinating hemorrhage.\(^{14,16}\) These lesions may show remarkable growth potential and may become locally invasive. When deciding upon a treatment plan for central hemangioma, the practitioner’s primary concerns are: (1) control of hemorrhage; (2) eradication of the lesion; and (3) prevention of reoccurrence.

Treatment methods mentioned in the literature include: (1) noninvasive radiotherapy; (2) injection of sclerosing and embolizing agents\(^ {17}\); and (3) surgical intervention by curettage and radical resection with immediate osseous reconstruction.\(^ {4,7}\) Although most options have met with moderate success statistically, surgery has been the most favored mode of treatment in spite of the hazards of hemorrhage and a certain degree of deformity through loss of dental and bony tissues.\(^ {7}\)

Consideration must also be given to the position, extent, and clinical presentation of the lesion as well as the patient’s age and medical history before treatment is initiated.

Radiation therapy is often chosen for treatment when it is decided that the lesion is inaccessible or that surgical intervention would be too mutilating. Jaffe pointed out that although the growth of lesion may be checked with radiotherapy, any osseous deformity that is present would remain until surgical intervention is carried out.\(^ {18}\) Some authors, including Macnash and Owen, have reported numerous cases successfully treated by radiotherapy alone (500R to 3300R).\(^ {19}\) Clinical improvement, however, does not always indicate complete resolution of the lesion; the possibility of recurrence remains. Wilde et al expressed the opinion that radiotherapy cannot be curative, since hemangioma is a lesion of mature cells that are considered highly resistant to radiation.\(^ {20}\) In addition, the presence of the lesion within the mandible and maxilla lessens the actual exposure of the tumor to radiation. Possible complications, however—such as damage to the condylar growth center, developing teeth, and salivary glands—are associated with its use.

Large extensive lesions have also been treated solely and successfully with intralesional injections of sclerosing agents.\(^ {3}\) Various materials, including boiling water, sodium morrhuate, and sodium tetradecyl sulfate, have achieved widespread use because they are both tissue irritants and thrombogenic agents. These agents provoke an inflammatory response with subsequent fibrosis and obliteration of vascular channels. The success of sclerosing agents is, however, restricted to superficial soft tissues, and their value in treating intraosseous lesions is doubtful.

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**Figure 1.** Extraoral presentation of the patient with a central hemangioma of the left side of the mandible causing facial asymmetry.

**Figure 2.** Mandibular occlusal radiograph of the involved area showing an expansile lytic lesion of the body of the mandible and displaced teeth.

**Figure 3.** Panoramic view exhibiting coarse trabeculation and increased radiopacity extending from the mandibular lateral incisor to the permanent mandibular first molar region.
Embolization of major afferent vessels feeding a central hemangioma is also a possible treatment. This technique has also been used as the sole choice of treatment when the patient is not considered an ideal surgical candidate or in lesions that were so extensive that other forms of therapy would have been considered too radical.

Surgery, either alone or in combination with embolization, remains the treatment of choice for central hemangioma. The exact nature and extent of the surgical procedure employed depends upon the: (1) age; (2) medical history of the patient; and (2) lesion’s clinical aspect. The most conservative surgical approach has been simple aspiration of the intraosseous lesion. Reduction of such lesions is presumably secondary to: (1) reduction of vascularity; (2) organization fibrosis; and finally (3) reossification.

Presurgical preparation is the most important aspect of surgical treatment. Loss of blood during surgery must be anticipated, and, consequently, adequate replacement must be available. If surgical intervention is planned, unilateral or bilateral ligation to decrease the vascular supply of the external carotid artery or superficially feeding vessels is indicated.

Surgical removal of the intraosseous lesion can be accomplished by either conservative curettage or radical excision of a part of the jaw with immediate bone graft reconstruction. Curettage involves removal of the buccal plate and exposure of soft hemorrhagic tissue, which is conservatively removed while preserving the continuity of the jaw. La Dow et al considered block resection with immediate bone graft reconstruction (from the iliac crest) the most effective and safest form of treatment. They suggested that the borders

Figure 4. Contrast CT scan showing intense enhancement, thus suggesting a vascular pathology.

Figure 5. Sagittal section CT showing the extent of the lesion.

Figure 6. View of the lesion during resection showing an expansile lesion with thinning of the cortical plate.

Figure 7. Reconstruction plate in place to stabilize the ends of the resected segments and to maintain space for grafting.
of resection should go through uninvolved surrounding tissue to avoid manipulation of the lesion and to lessen the prospect of operative hemorrhage.

Shira and Guernsey expressed the belief that hemangiomas may occasionally regress spontaneously through a process of multiple infarctions and subsequent bony reformations.1

**Case report**

An 8-year-old male child of Indian origin reported to the Department of Pedodontics and Preventive Dentistry, Government Dental College, Rohtak, Haryana, India, with the chief complaint of a swelling on the left side of the face over the previous 10 months. The patient’s history revealed uneventful extraction of the carious primary predecessor before the onset of swelling. The swelling started small, reached its present size and form in 5 months, and had been the same ever since. An occasional toothache was the only accompanying symptom, and no history of parasthesia, spontaneous bleeding, or epistaxis was given.

Upon extraoral examination, facial asymmetry was evident on the left side. The swelling was approximately 4x5 cm in size and was present over the left mandibular body with diffuse margins (Figure 1). Palpation revealed a bony, hard, nonpainful swelling in relation to the lateral aspect of the mandible. The swelling was firm, nonhyperthermic, and sensitive only to deep palpation.

Intraoral examination exhibited obliteration of the buccal vestibule extending from the distal aspect of the mandibular left lateral incisor to the mesial aspect of the permanent mandibular left first molar. There was expansion of the mandible’s lateral aspect in relation to this area with blanching but without any pulsations. The dentition exhibited age specificity with the deflection of the crowns of the left lateral incisor and the first premolar associated with loss of occlusion on the same side. Teeth present in the involved area were vital and exhibited limited mobility, except for the first premolar—which was markedly (grade 3) mobile. All the other findings were noncontributory.

A complete radiographic analysis was carried out. Intraoral periapical radiographs showed nondescript findings. Mandibular occlusal radiographs exhibited an expansile lesion of the left mandibular body with erosion of the lateral cortex (Figure 2). A panoramic radiograph disclosed coarse trabeculations and increased radiodensity extending from the mandibular lateral incisor to the mandibular permanent first molar region (Figure 3). Small circular radiolucencies and multiple radio-opaque striae giving a honeycomb appearance were also observed. The tooth buds of the canine and second premolar were deflected. Laboratory investigations—which included complete blood report, urinalysis, and investigations for parathormone efficacy—were within normal limits. Since the lesion’s clinical and radiographic presentations were suggestive of hemangioma or a giant cell lesion, it was decided to confirm the diagnosis by CT scan. The scan’s findings revealed an expansile lesion of the left mandibular body with obvious vascular involvement (Figures 4 and 5). This strengthened the authors’ earlier suspicion of hemangioma. To confirm this finding, fine needle aspiration cytology (FNAC) was conducted. This revealed the presence of thin endothelial-lined vascular channels and fibrocollagenous soft tissue. After reviewing all the previous findings, a final diagnosis of central hemangioma was made.

In consultation with the Department of Oral and Maxillofacial Surgery, Government Dental College, Rohtak, Haryana, India, a joint admission and surgical intervention was planned. The patient was admitted 2 days prior to surgery and presurgical preparation was initiated, which included: (1) blood analysis; (2) urinalysis; (3) laboratory investigations; (4) chest X ray; (5) physical examination;...
(6) blood group determination; and (7) cross-matching. Since loss of blood during surgery was anticipated, a provision was made for rapid replacement of blood in case of massive hemorrhage.

After surgical preparation of the patient for general anesthesia, a submandibular incision was made extending from the angle of mandible to the symphyseal area. En bloc resection of the mandibular lesion—including 1 cm of healthy bone—was performed (Figure 6). A nickel-titanium reconstruction plate (spacer) was placed to cover the defect caused by the lesion and to maintain the space for bone grafting at a later stage (Figures 7 and 8). The mandibular fragment was sent to surgical pathology for histopathological diagnosis (Figure 9). The results confirmed the presurgical diagnosis of central hemangioma. The patient has been on follow-up since 8 weeks postsurgery, the wound is healing uneventfully, and the reconstruction of the mandibular defect via grafting is being planned (Figure 10a and b).

**Discussion**

The present case illustrates many features that are characteristic of central hemangioma, including the patient's: (1) clinical history; (2) findings of examination; (3) radiographic and scanning examination.

Radiographically, a differential diagnosis of ameloblastoma, cavernous hemangioma, giant cell lesion, cyst and myxoma could be made due to the characteristic sunburst appearance. Scanning reports showing vascular involvement limited the authors' horizon to hemangioma or a giant cell lesion, and the former was confirmed via FNAC. Due to the lesion's vascular involvement and the risk of hemorrhage, it was decided to conduct FNAC instead of a biopsy.

Radiotherapy was not the treatment of choice, considering the age of the patient and the retarding effects of radiation on oral and perioral tissues. Intraluminal injections of sclerosing agents would not have been effective because of the lesion's bony nature. Hence, surgical resection and osseous reconstruction was the treatment of choice due to multiple factors such as the: (1) lesion size; (2) child's growth potential; and (3) lesion's approachability. The mandible was resected beyond the lesion's radiographic boundaries to avoid any manipulation of the vascular lesion and to prevent any complications, such as extensive hemorrhage. Because of the serious consequences, hemangiomas must always be considered in the differential diagnosis and proper precautions must be taken in establishing the final diagnosis before any surgical treatment is undertaken.

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**References**


Abstract of the Scientific Literature

Obstructive Sleep Apnea in Children

The objective of this study was to perform respiratory-gated magnetic resonance imaging to evaluate airway dynamics during tidal breathing in 10 children with obstructive sleep apnea syndrome (OSAS; age=4.3±2.3 years) and 10 matched control subjects (age=5±2 years). The authors hypothesized that respiratory cycle fluctuations in the upper airway cross-sectional area would be larger in children with OSAS.

Studies were performed under sedation. Respiratory gating was performed automatically at 10%, 30%, 50%, 70%, and 90% of inspiratory and expiratory volume. Airway cross-sectional area was measured at 4 ascending oropharyngeal levels at each increment of the respiratory cycle.

The authors noted the following in subjects with OSAS compared with control subjects: (1) a smaller upper airway cross-sectional area, particularly during inspiration; (2) airway narrowing occurred during inspiration without evidence of complete airway collapse; (3) airway dilatation occurred during expiration, particularly early in the phase; and (4) magnitude of cross-sectional areas fluctuations during tidal breathing noted in OSAS at levels 1 through 4 were 317%, 422%, 785%, and 922%, compared with 19%, 15%, 17%, and 24% in control subjects (P<.001, P<.005, P<.001, and P<.001, respectively).

Fluctuations in airway area during tidal breathing are significantly greater in OSAS subjects compared with control subjects. Resistive pressure loading is a probable explanation, although increased airway compliance may be a contributing factor.

Comments: Obstructive sleep apnea often occurs in children with adentonsillar hypertrophy. Sedation is contraindicated in children with OSA, although in this study sedation was necessary to complete the measurements required to prove the authors’ hypothesis. Providing dental treatment to children with OSA can only be accomplished safely with local or general anesthesia. This study provides excellent evidence, albeit from a small sample size, that both airway resistance and compliance are highly variable and significantly elevated in OSA children and provides an important explanation why we should avoid sedation in the presence of OSA.

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