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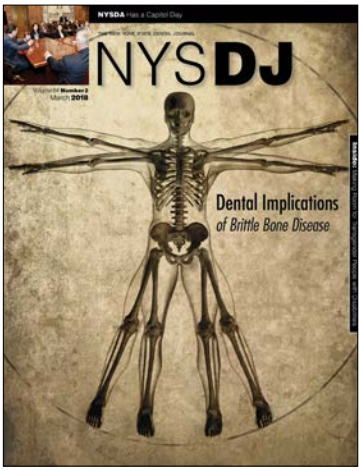

Cover: Victims of osteogenesis imperfecta, or brittle bone disease, may be afflicted with hearing loss, blue sclera, thinness of skin, weak joints, easy bruising, deficient growth and dental abnormalities that require intervention to improve and control oral diseases and restore proper occlusal function.

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Know Where You are Going to Get There

A written plan is essential to a successful career and personal life.

What will my personal life and career as a dentist look like in five years? We all wonder what the future holds for us. Depending upon our circumstances at the time of the inquiry, the possibilities may comfort or disturb us. Since we know we cannot control the future, we often abandon further analysis. However, when we avoid all thought about what we want and how we will get it, we risk not knowing where we are going. In most cases, not knowing our destination makes it less likely we will ever get there.

Dentists visualize desired clinical results before initiating treatment every day as a prerequisite to success. Yet, we often fail to apply the same principles to personal and practice decisions. Sure, it involves less work to put on the blinders, live life and accept that others will determine where and when we will arrive. After all, it is intimidating to confront our fears that we are not good or deserving enough to attain our goals. We succumb to the enormity of the task of how we technically could achieve our aspirations, and it forces us down the slippery slope of inaction. We lose our focus and it impairs our ability to lead others. However, when we strategically plan for what we want, the process forces us to ask the more central question of why we want it in the first place. Ultimately, as we discover our true motivations, we will uncover our inherent value as an individual and our purpose as a dentist.

Leaders know where they are going and why they want to get there. Only commitment to clearly defined goals can inspire others to join their cause. We can achieve these self-realizations through careful drafting and pursuing our own personal and professional vision and mission statements. These writings force us to focus on what is important and to organize our plan. We can more easily select which challenges we will undertake, and those we will decline, when we know our objectives and why we chose them.

We set specific goals and use these objectives to motivate us to become the best version of ourselves. Putting our thoughts and dreams into writing further crystallizes abstract ideas. Our manuscript memorializes the terms that strengthen our own commitment to their achievement. We can then more easily share our personal goals with our significant others and our professional goals with our staff. Only then can others truly buy-in and contribute to our success.

We should start with a vision that projects where we want to be in five years, both in our personal and professional lives. The vision includes a general description and specific objectives. As life presents new challenges, we must periodically revise the statements to reflect the changes. Our vision will create the foundation for personal and professional mission statements that focus upon today's conduct necessary to make tomorrow's vision a reality. Somewhere in the process, the magic occurs. As we dream, plan, succeed and fail, we discover, through trial and error, why we chose dentistry as

a career and what we were meant to do as individuals. We must then use this vital knowledge to redirect our path toward our true destination.

Ideally, our personal goals should set the parameters for our professional goals as a dentist. Our practices should enable us to better realize our personal objectives. When our practice wins, so should our personal lives. We should not let our practices set us up for personal failure. Hence, our personal visions, missions and objectives, to a certain extent, should stand independent from our professional lives. We will enjoy stronger commitment to both the personal and professional areas when we recognize our practices form only part of the puzzle, but never conflict with what we know to be important to ourselves.

The benefits of thoughtful, written statements regarding our goals and plans to achieve them apply to dentists at all stages in their careers and to our professional organizations. New dentists use their mission statements to assist them in the selection of employment opportunities and ownership options. Owner dentists strategically plan their practice profile and, based upon their goals, provide leadership for their family and practice. Senior dentists can transition out of practice on their own terms. Importantly, organized dentistry can only effectively lead our profession, its member dentists and our patients if it knows where it is going and why it wants to get there.

NYSDA's documented success in providing services and education, advocacy on the behalf of dentists and their patients, and representation to government agencies, all flow from its mission statement to "promote the public's health" and its "commitment to provide quality dental care accessible to everyone." Only a mission statement that identifies the value of organized dentistry to the profession, its members and the public and its purpose in the advancement of oral health can keep the organization relevant. Only mission statements that reflect our inherent value as individuals and our purpose as dentists will allow us to know where we are going and how to get there.

D.D.S., J.D.

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Osteogenesis Imperfecta

A Case Report

Alice Golden, D.D.S.; Taka Suzuki, D.D.S., Ph.D.; Veena Nanda, D.M.D., M.S., Ph.D.

ABSTRACT

Osteogenesis imperfecta (OI) is a rare and autosomal dominant connective tissue disorder that causes bone fragility and recurrent multiple bone fractures. The generalized decrease in bone mass results from mutations in Type 1 collagen. OI is further classified based on clinical presentations, mode inheritance, and if there is reduction in quantity or quality of collagen production. This disorder is commonly associated with blue sclera, hypermobility of joints, progressive ear deafness, dental abnormalities and opalescent teeth. The dental manifestations are consistent with opalescent appearance of the dentition and Class III malocclusions. Frequently, patients exhibit fractures of tooth enamel and heavy dentinal attrition.

The prototypical radiographic findings include marked cervical constriction with pulp chambers and root canals completely obliterated. This leads to a deficiency in the dentinoenamel junction, resulting in enamel chipping, attrition and loss of vertical dimension. Therefore, dentists continually face a challenge when treating patients with OI and must work closely with the primary care provider. It becomes a multidisciplinary approach involving orthogenetic surgery, periodontal, restorative and prosthodontic procedures. Early and appropriate dental care in these patients can improve and control oral diseases and restore proper occlusal function.

A case of OI is presented with the goal of presenting the dental and skeletal characteristics and dental management of a patient with OI.

Osteogenesis imperfecta (OI), also known as "brittle bone disease," is an autosomal dominant disorder of connective tissue. It is one of the most commonly recognized inheritable disorders of connective tissue (HDTC).[10] This heterogeneous disorder is characterized by bone fragility and recurrent, multiple bone fractures.[3] The incidence has been reported to be from 1:10,000 to 1:20,000 births; it affects males and females equally.[12]

The condition results from mutations in the genes COL1A1 and COL1A2 that code for Type 1 collagen.[10] A number of mutations have been identified that affect COL1A1 and COL1A2 genes.[3] This disturbance in collagen formation results in a variety of genetic mutations that code for proteins responsible for the assembly and maintenance of bone and connective tissues. This leads to poor bone quality and quantity, and causes bone fractures. The fractures result from deficiency of osteoblasts, osteoid and periosteal bone formation.[4] Therefore, severity and phenotypic presentation varies significantly.

Sillence et al. classified OI into four basic types in 1979. The classification is based on clinical presentation and mode of inheritance. A reduction in the quantity of collagen results in OI of Type I, while the qualitative and quantitative alterations in collagen synthesis result in types II, III and IV.[7] Type I is an autosomal dominant mild form of OI, with a typical presentation of blue sclera and hearing deficits in 50% of the cases.[10] Type II is the most severe form, where most infants are stillborn or die shortly after birth. Type III is almost as severe as Type II. Infants are born with fractures and deformity and are unable to survive the perinatal period as a consequence of severe skeletal changes. Type IV is an autosomal dominant form with moderate severity. The typical presentation is normal sclera and hearing, bowing bones and vertebral fractures.[10] These patients also present with shorter stature due to a lessened postnatal growth.

There has been a new classification of OI that expanded the old classification from four types into a classification consisting of 12 different types. OI Type V, an autosomal dominant mode of inherence, is similar to OI Type IV in the frequency of fractures and degree of skeletal deformity. Among the most defining features are large, hypertrophic calluses in the largest bones at fracture or surgical procedure sites. In addition, hypertrophic calluses can occur spontaneously. Type VI is an autosomal recessive inheritance that is extremely rare and is moderate in severity. This type is distinguished histologically by a characteristic mineralization defect in the bone.

Two recessive types of OI, Types VII and VIII, do not involve mutations in the Type I collagen. Instead, these types result from mutations in two genes that affect collagen by post-translational modification: the cartilage-associated protein gene (CRTAP) and the prolyl 3- hydroxylase 1 gene (LEPRE1). Type VII results from a mutation in the CRTAP gene and resembles OI Type II, except that

infants have white sclera, smaller heads and round faces. Patients also exhibit short stature, short humeri and femora. Mutation in CRTAP can lead to moderate bone dysplasia.

Type VIII is characterized by severe growth deficiency and severe under-mineralization of the skeleton. It is caused by mutations in the LEPRE1 gene, resulting in a deficiency of prolyl3-hydroxylase activity. Types IX, X, XI, XII have a mode of autosomal recessive inheritance as well. The complexity of the genotypic and phenotypic variability of OI has expanded its classification into 12 distinct classifications.

Most cases are classified as Type 1 or Type IV, with or without teeth involvement. OI can be further subdivided based on the presence or absence of opalescent teeth.[7] There is a wide variety of presentation of OI, with and without association of opalescent teeth. There are patients with OI who present with considerable dentin involvement, and there are those who manifest no clinical or radiographic aberrations in the dentition. Therefore, clinical, radiographic and histologic findings are ways to differentiate the types of OI and the presence or absence of opalescent teeth.

The clinical and phenotypic presentation varies widely based on the type and severity of Type I collagen deficiency. Type I collagen is a major protein that is a constituent in tendons, ligaments, skin, sclera, teeth, and middle and inner ear.[10] Therefore, all tissues composed mainly of Type I collagen are affected. Infants and children with OI often present with a triangular-shaped face.

Most common abnormalities include hearing loss, blue sclera, thinness of skin, weak joints, easy bruising and deficient growth. The sclera is abnormally thin, and the pigmented choroid shows through giving the clinical characteristic of pale blue sclera. However, it is only present in about 30% to 65% of patients.[6] Patients present with short stature, opalescent teeth, asthma and spinal curvature.[10] Patients with OI are more prone to sustain fractures from minimal trauma due to bone fragility and abnormal bone formation. Therefore, the most frequent complication is bone pain.

Each of the four types of OI is further subdivided on the basis of the absence or presence of opalescent teeth. OI with involvement of opalescent teeth affects both primary and permanent dentition. The main changes of opalescent teeth associated with OI are the color of the teeth and enamel fractures. Patients may also exhibit skeletal Class III malocclusion and an open bite. Class III malocclusions occur in 70% to 80% of types III and IV OI cases, with a high incidence of anterior and posterior crossbites and open bites.[7]

Primary teeth are more severely affected; and the teeth have an opalescent, gray, brown or yellow hue. Enamel begins to chip away from the incisal portion of anterior teeth, from the occlusal surface of posterior teeth, and from the vertical buccal or lingual surfaces of all teeth.[10] Abrasions occur on the exposed dentin; and the dentin surface is smooth with the gingival tissues. The permanent dentition may appear healthy, but have crowns that are short and abraded.

The radiographic findings demonstrate a marked cervical constriction; crowns tend to be bulbous; and roots are short and slender.[10] Pulp chambers and canals are obliterated over time. Tooth enamel tends to crack off due to deficient dentinoenamel junction, resulting in attrition and loss of vertical dimension. The enamel is more irregularly mineralized, and there is a lower degree of mineralization. This results in enamel chipping, which leads to dentinal exposure, and can result in periapical radiolucencies.[10]

Histologically, opalescent teeth associated with OI will have abnormal dentin, in which dentinal tubules are reduced in number or completely absent. The tubules are narrow, tortuous, short and do not penetrate the whole thickness of dentin. This results in excess intertubular dentin.[10]

Opalescent teeth associated with OI vary within as well as between dentitions, but the loss of tooth structure is a consistent finding, which may result in tooth fracture. Therefore, dentists face a challenge when treating patients with OI because of the great number of failures in adhesive procedures. This is because bonding resin to defective tooth structure is problematic; however, it has been successful in some patients. Adhesive dentistry is

Figure 1. Extraoral view. Frontal and profile.

not contraindicated in patients with OI, but careful consideration must be taken on an individual basis, depending on the variability of dentin involvement. Veneers and full-coverage restorations have been useful to mask the discoloration.

The dental treatment should also be an integral part of the overall care of the patient and his or her sense of well-being. There are few articles addressing the specific type of dental management and the overall caries and periodontal risk. Full-coverage restorations and complete dentures have been implanted to preserve function, vertical dimension, normal growth and esthetics. Because of significant malocclusion and skeletal Class III and open bite, orthodontic and orthognathic therapy have been shown to be beneficial.

The unfortunate consequence of early tooth loss in patients with OI poses a treatment planning dilemma for the dentist. Since the quality and quantity of bone is of paramount significance for implant osseointegration, dentists are hesitant to treat these patients with implants because of the osteoporotic condition of bone. Therefore, there has been limited research relating the use of dental implants in patients with OI. Few reported cases address the success of osseointegration of the implant or the risk of failure in implant-supported prostheses in such patients. The long-term success of implant placement, healing and loading has not been answered in patients with OI.

A case of OI is presented here with the goal of presenting the dental and skeletal characteristics and the dental management of a patient with OI.

Case Report

A 32-year-old African-American male presented to New York University College of Dentistry for comprehensive dental care. His major concern was his "cracked teeth," which impaired masticatory function. The patient's medical history was significant for Type IV osteogenesis imperfecta (OI). The patient presented with a mobility aid for assistance and accessibility.

Past medical history revealed that the patient was diagnosed with OI at age 4 following bilateral compound fractures in his femurs. The diagnosis was reached after multiple lab tests and radiographs. Following the fractures, half-body plaster casts were used for initial treatment during the acute stage of the fracture. Telescopic expanding rods, Dubow-Bailey rods, were placed in both femurs to lengthen the long bones during growth. Throughout his adolescence, the patient reported having seven to eight fractures, all involving the long bones, and occasional cracked ribs. He was further treated with plaster casts and splits. Besides his condition of OI, he had no other significant medical history. Socially, he was single, with an older brother diagnosed with sickle cell anemia. To the patient's knowledge, there was no family history of bone disorders.

Past dental experiences revealed that the patient did not receive dental care regularly. Most of his dental visits consisted of extractions and occasional dental prophylaxis. The patient's oral hygiene practices were reported and were minimal. He brushed only once a day, used a mouthwash sparingly and never flossed.

Physical examination revealed he was short in stature, at a height of four feet and five inches (Figure 1). Orofacial clinical examination also revealed a triangular-shaped face, with a convex facial profile, competent lips and eyes with normal sclera. The tongue, lingual, labial frenum, soft and hard palate were normal.

Intraoral exam revealed an opalescent dentition with a Class III malocclusion (Figure 2). The patient exhibited an anterior open bite with a diastema and bilateral posterior crossbite. Multiple teeth were rotated and had a bell-shaped appearance. Maxillary and mandibular molars had abrasions on the exposed dentin; the dentinal surface was smooth and continuous with the gingival tissue. The crown fractures involved either the lingual or buccal wall of the tooth and were not related to caries severity. Clinically, the lingual walls of teeth #2, #3, #14, #19, #30 and the buccal wall of #13 were all fractured to the gum line (Figure 3). Clinical exam did not reveal detectable caries in any of the fractured crowns.

The patient exhibited gingivitis, with probing depths measurements between 2 mm and 5 mm pockets. There was bleeding on probing on almost every surface. There was mild recession

throughout the dentition, with moderate clinical attachment loss. There was extensive bone loss in the maxillary left posterior region (Figure 4). The mandibular incisors exhibited +1 to +2 mobility, with widened PDL space.

Radiographically, the crowns of the teeth were bulbous. There were also marked cervical constrictions. The pulp chambers were obliterated (Figure 5). The roots were short, slender and dilacerated. The enamel appeared slightly thicker and more radiopaque than dentin.

Discussion

The patient's chief concern was his cracked teeth, which caused problems and pain during oral functions, such as chewing. The patient was not interested in correcting either his Class III malocclusion or his discolored teeth. His primary concern was to restore the fractured crowns for proper function.

The first phase of the treatment plan consisted of four quadrants of scaling and root planing to debride the infected tissue and create a biologically compatible root surface. Extraction of the maxillary right and left third molars was also included, to prevent food impaction and further periodontal bone loss. The patient was taught how to properly brush and floss to reduce gingival inflammation. A re-evaluation at four to six weeks following initial therapy was arranged to determine patient compliance with oral hygiene prior to starting the restorative phase of treatment and to allow optimal gingival healing time.

The second phase of treatment, the restorative phase, addressed the patient's chief concern. The maxillary right second molar, tooth #2, would receive a porcelain-fused-to-metal crown because of extensive crown fracture. A conservative approach with tooth #3 was decided. A distal-lingual composite would be performed initially. If the composite filling failed, a modification would consist of fabricating an onlay. The maxillary left second premolar, tooth #13, would be extracted because of extensive facial crown fracture. A bridge would then be fabricated from tooth #12 to tooth #14. The mandibular left and right first molars, teeth #19 and #30, would receive lingual composites, as a conservative approach prior to fabrication of onlays or crowns. The edentulous space for tooth #31 would receive an implant, and tooth #32 would be extracted after integration and loading of the implant.

The first phase of treatment was critical to implement because of the patient's history of OI and his moderate periodontal risk. Although studies have suggested that periodontal disease is rare in patients with OI, our patient demonstrated gingival inflammation with moderate probing depths and beginning periodontal bone loss. Increased mobility of the lower anteriors was associated with increased width of PDL and reduced height of the alveolar bone. All of these factors can stem from the patient's poor oral hygiene. His lack of appropriate daily home care can

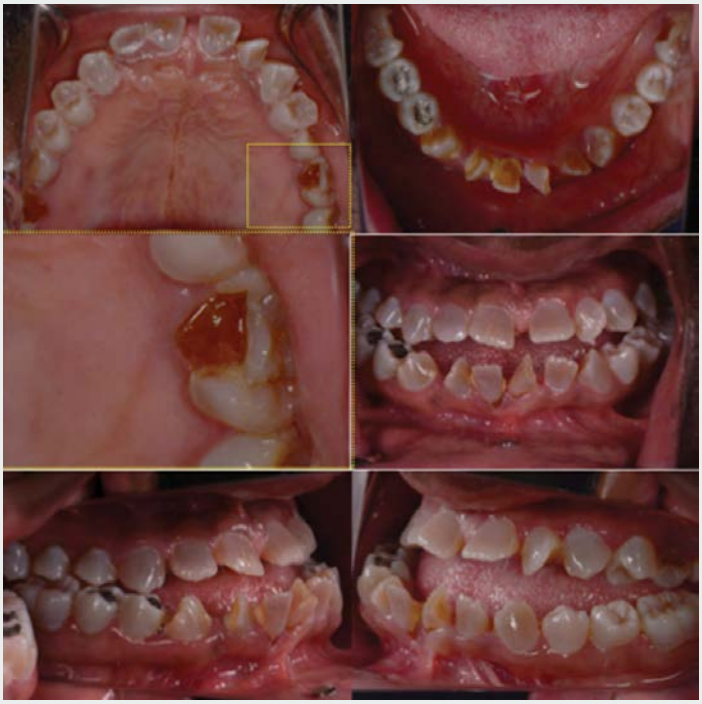

Figure 2. Intraoral: Maxillary and mandibular occlusal views (top); Palatal view of tooth #3, anterior view (middle); Right lateral, left lateral (lower).

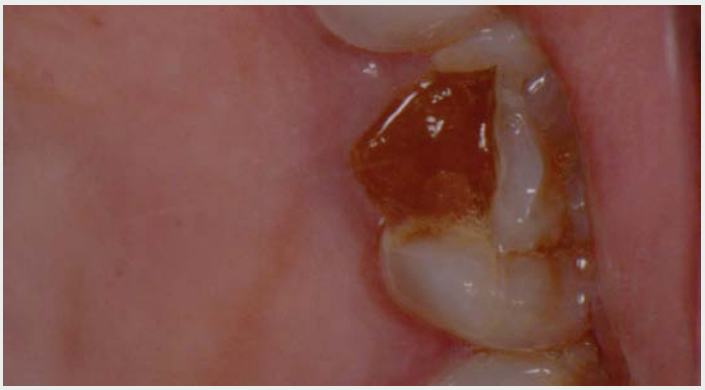

Figure 3. Fracture of lingual wall on tooth #3.

be attributed to the fact that he was not instructed or educated on the proper technique and regimen for oral hygiene. Therefore, scaling and root planing were recommended as the initial phase of treatment.

The coronal fractures detected clinically can be attributed to the weakness in the dentin, as a result of the molecular defect in collagen. Studies have demonstrated that deviations within the DEJ and decreased mineral content can cause coronal tooth fractures.[10] Therefore, the dentinal tubules are reduced in number and are considered to be shorter, narrower, tortuous, and do not penetrate the whole thickness of the dentin.[10] This can explain the multiple coronal fractures observed in our patient in each quadrant. These fractures posed an intense problem during oral function.

Treating these coronal fractures may be difficult due to adhesive failures. Although it can become problematic to bond resin

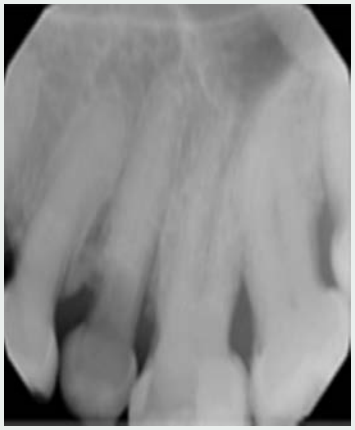

Figure 4. Bone loss in UL quadrant.

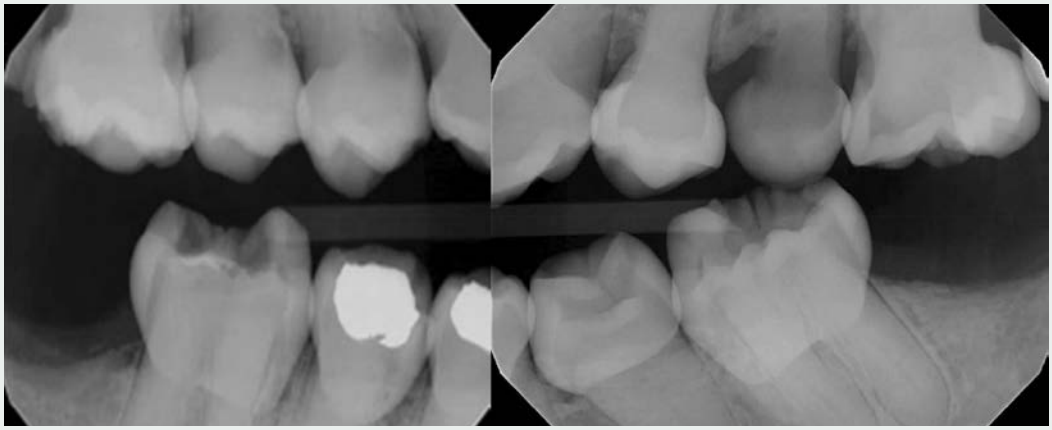

Figure 5. Obliteration of pulp chambers on BW

to defective tooth structure, it has been shown clinically to be successful.[7] Adhesive dentistry is not contraindicated in patients with OI and should be considered. Therefore, we decided on a conservative approach to restore the coronal fractures with resin-bonded restorations.

Implant-supported fixed prosthesis for the edentulous space on the lower right is a feasible treatment decision rather than fabrication of a bridge. As supported by literature, implant surgery and prosthetic rehabilitation have been successful for edentulous patients with osteogenesis imperfect.[8,9] Extraction of the third molars would not facilitate oral hygiene compliance, but would serve to benefit the patient's occlusion and functional dentition.

Conclusion

Dental treatment for patients with osteogenesis imperfecta has been shown to be challenging, but not impossible. The essential objective is to maintain dental health by facilitating proper preventive measures. Treatment involves a multidisciplinary approach with proper periodontal, restorative and prosthodontic procedures. Early and appropriate dental care in these patients can improve and control oral diseases and restore proper occlusal function.

Queries about this article can be sent to Dr. Golden at ajg427@nyu.edu.

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Case of Bilateral Multilocular Expansile Traumatic Bone Cysts

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ABSTRACT

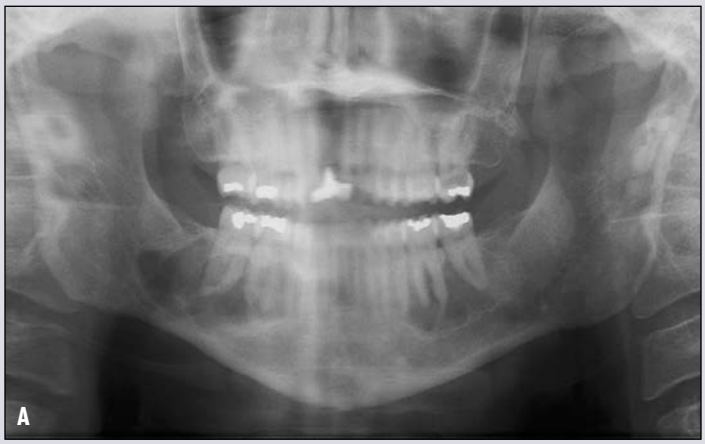
Traumatic bone cyst (TBC), unlike other bone cysts, is a benign lesion with no epithelial lining. Diagnosis is achieved by clinical, radiographic and histologic findings, and confirmed during surgical exploration of the "empty" bone cavity. Most TBCs appear unilateral. Radiographically, they generally present with distinct margins, and are located above the alveolar canal. The lesions are often seen in close proximity to vital teeth. TBCs do not commonly present with pain or neurological changes; they are most frequently an incidental finding on radiographic analysis. TBCs rarely present in an aggressive manner. This is a case report of a patient who presented with multilocular expansile traumatic bone cysts of the bilateral posterior mandible. The clinical presentation, diagnostic criteria, treatment and outcome for such an aggressive presentation will be discussed.

Traumatic bone cyst (TBC) is a diagnosis of exclusion. The etiology is unknown. Patients with TBC present with asymptomatic, radiolucent lesion(s) with or without a history of trauma to the affected area. Typical clinical and radiographic presentation is a unilateral radiolucency with scalloping effect above the alveolar canal between the roots of vital teeth. However, 20% of traumatic bone cysts present with multilocular radiolucencies, with higher chances of cortical expansion and enlargement of the lesion along the body and ascending ramus of the mandible.

Presentation of bilateral TBCs with multilocular radiolucencies is rare, with only one other reported case.[1] The discussion presented here will involve a rare case of a 50-year-old female who presented with bilateral multilocular expansile bony lesions of the posterior mandible. The presentation, diagnosis, treatment and long-term management of a more aggressive occurrence of TBC will be detailed in this report.

Case Report

A 50-year-old African-American female with asymptomatic, slowly enlarging bilateral mandibular lesions presented to the hospital upon referral from her primary care physician, who had discovered the lesions on a computed tomography (CT) scan.



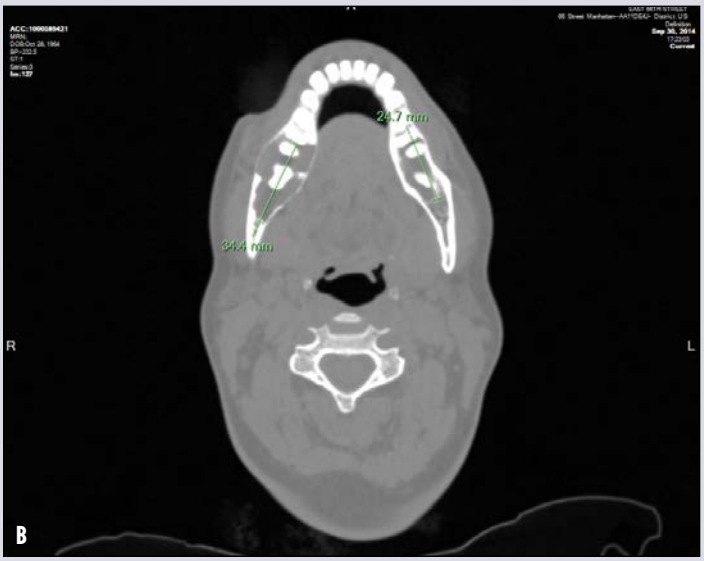


Figure 1. Preoperative images. A. Panoramic image: bilateral radiolucent expansible masses of posterior mandible #18, #19, #20, #29, #30, #31 extending along ascending ramus. B. Axial view: expansible mandible bilaterally, with bone loss in alveolar bone with buccal and lingual expansion.

The patient reported that in 2007, her dentist incidentally discovered a mandibular lesion on radiographic review. She underwent an incisional biopsy by an outside oral and maxillofacial surgeon, who reported, "the lesions were empty." The patient received no further treatment at that time and had an uneventful course following the biopsies. In September 2014, the patient developed bilateral cervical lymphadenopathy after a scaling and root planing procedure. She reported to her primary care physician, who ordered a head and neck CT scan, which detailed the bilateral expansile multilocular mandibular lesions. Upon initial evaluation in August 2015, the patient denied symptoms of pain, mobility of the teeth or paresthesia; she reported no history of trauma to the region.

The patient's past medical history was significant for sickle cell trait, kidney stones and uterine fibroids that were treated with a hysterectomy. The physical examination was unremarkable except for the bilateral enlargement of the bony alveolus in the posterior mandible. All teeth were vital to cold testing; neighboring teeth showed no signs of mobility; and normal periodontal probing depths were found. Panoramic images and CT indicated bilateral, well-defined multilocular radiolucencies intimately associated with teeth #18–#20 and #29–#31, extending posterior to the second molar above the level of inferior alveolar canal, with buccal and lingual alveolar expansion (Figure 1).

The treatment rendered included decortication of the lesions, with histologic examination of the buccal cortex, as no gross intralesional soft tissue was detected during exploration of the lesions. A gentle curettage of the bony walls was performed under intravenous sedation. Prior to decortication, a clear yellow, straw-color aspirate was obtained. The surgical findings were notable for bilateral empty bone cavities; no cystic lining was seen. The decortication was limited to avoid disturbing the tooth roots and inferior alveolar canal.

Histopathologic examination indicated viable cancellous bone containing loose fibrovascular marrow with infiltration of

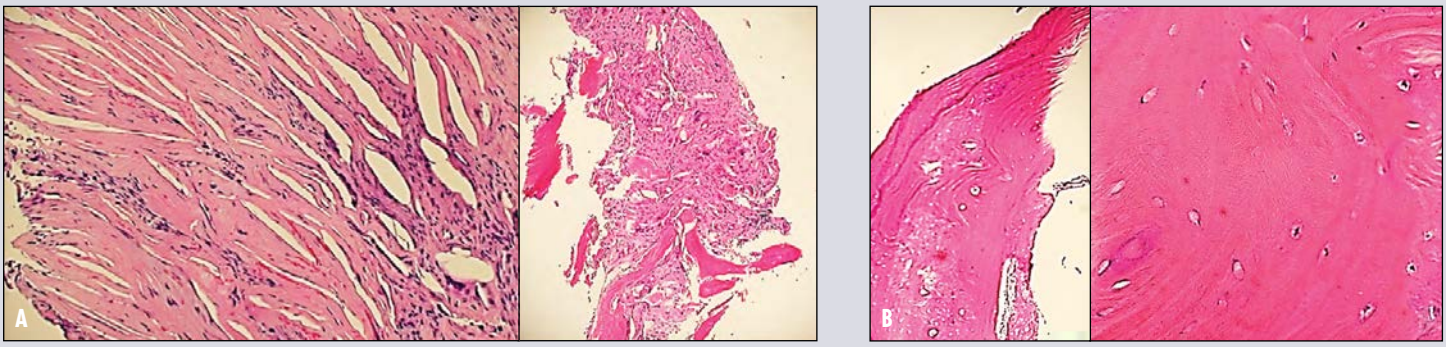


Figure 2: Histology. Right (A) and left (B) bone biopsy indicated viable cancellous bone containing loose fibrovascular marrow, infiltrate of lymphocytes, plasma cells.

lymphocytes and plasma cells bilaterally and no cystic lining. This indicated the diagnosis of traumatic bone cysts, a diagnosis of exclusion (Figure 2). No packing or graft material was used at the time of surgery. The patient had an uneventful postoperative course and denied any neurosensory disturbances. The patient was followed over the next several months, with periodic imaging to monitor for bony fill of the lesions. Postoperative panoramic image indicates filling of bone (Figure 3).

Discussion

Traumatic bone cyst (TBC) is an intra-osseous, pseudocystic, non-epithelial-lined lesion of the jaws.[2-4] TBC is mostly diagnosed in young patients during the first two decades of life[3,5] and accidently detected by panoramic radiograph during a routine dental exam.[2,5] TBC has an even sex distribution but, according to some authors, males are more affected.[2] Results of a study involving 255 cases indicate that TBC occurs in the posterior region of the mandible 89% of the time, while showing 11% of the time in the anterior region of the maxilla.[6]

Etiology of the lesion is unknown; however, there are various hypotheses that suggest trauma and/or multifactorial causes, such as bone tumor degeneration, low-grade infection, ischemia, intramedullary bleeding, local alteration in the bone growth, increased osteolysis, venous obstruction, altered calcium metabolism or a combination of such factors.[3,6] The most widely suggested theory is that of trauma.[2,5] Unlike normal healing, where an organized blood clot forms leading to an area of healing, the clot liquefies, and the surrounding bone is destroyed by enzymatic activity.[3,6]

In most cases, patients with a traumatic bone cyst present with no symptoms. Only 10% to 30% of patients experience pain from the lesion; other rare symptoms include paresthesia, tooth sensitivity, fistulas, delayed eruption of permanent teeth and pathologic fractures.[3,7] On clinical exam, a TBC does not cause change in soft tissue overlying the lesion.[6,8] Neighboring teeth are vital and present with no mobility, displacement or resorption of their roots.[3,6] A TBC usually appears radiographically as a well-defined radiolucent unilocular lesion with irregular or scalloped borders with or without sclerosis at the periphery.[5,8] However, 20.6% of traumatic bone cysts have multilocular radiolucencies with cortical expansion, and 44.1 % demonstrate slow, progressive enlargement.[2]

Histological findings typically reveal a cancellous bone cavity, fibrous connective tissue and normal bone without an epithelial lining.[3,8] The lesions may exhibit areas of vascularity, erythrocytes, fibrin and occasional giant cells.[3] Definitive diagnosis of TBC is made by clinical, radiographic and histological findings but, invariably, diagnosis is achieved at surgery when an empty bone cavity without epithelial lining is observed.[3]

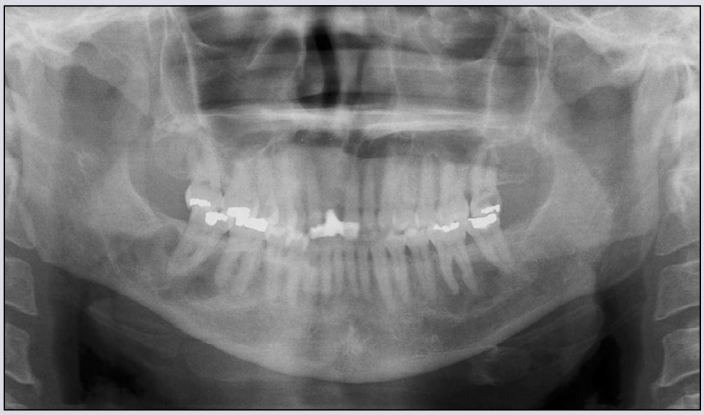


Figure 3. Postoperative panoramic image after two months follow-up. Bone beginning to fill into empty cavities bilaterally.

Surgical exploration followed by curettage of the bony walls is the treatment of choice, as a bleeding cavity forms a clot that will be replaced by normal bone despite the aggressive clinical presentation.[3] However, controversies regarding placement of a bone graft in the cavity remain. Application of gel foam, allogenic bone grafting with platelet-rich plasma (PRP), and injection of blood with hydroxyapatite and bone chips have also been proposed to have good results.[8]

In a review of orthopedic literature involving TBC of the femur, reports of curettage followed by grafting with tissue-engineered bone indicated good results.[9]

Rapid and complete healing may play a role in reduction of the incidence of pathological fractures.[9] Others indicated spontaneous resolution of the lesions without a bone graft.[10] Further investigation is needed to prove whether grafting in a site of a TBC is advantageous.

Queries about this article can be sent to Dr. Cho at juc9079c@nyp.org.

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Mucocle of Blandin-Nuhn

Case Report

Nathanel Shammay, M.S.; Louis Mandel, D.D.S.

ABSTRACT

Mucoceles of the Blandin-Nuhn salivary glands, or anterior lingual glands, are uncommon oral lesions infrequently found in adults. The following case report documents its presence in a 22-year-old female.

Other than in the gingiva, minor salivary glands can be found scattered throughout the soft tissues of the oral cavity.[1] The gland of Blandin-Nuhn, or anterior lingual gland, is anatomically situated in the ventral surface of the tongue, and is part of this minor salivary gland oral complex.

All minor salivary glands are located superficially in the submucosal tissues. Their excretory ducts open directly on to the overlying mucosal surface. The minor glands, along with the major salivary glands, are involved in the production of saliva and contribute to saliva's total protective, digestive, moistening and taste activities.

Cystic-like swellings, associated with minor salivary glands, are referred to as mucoceles, or mucous cysts. A minority of these lesions are true epithelially lined cysts that are formed as a consequence of secretory retention caused by duct obstruction. The majority of mucoceles are not lined by epithelium because they are not true cysts. Instead, because the swellings represent an extravasation phenomenon, a condensed granulation tissue lining will be found at the periphery of the escaped fluid.[2] These extravasation mucoceles usually originate from trauma to a minor gland's excretory duct, but patients may not recall the traumatic event that precipitated the lesion.[3] As a result of duct laceration, glandular secretions escape into the surrounding connective tissue and cause the formation of a reactive granulation tissue wall that encapsulates the leak.[4] This variety of mucocele is most commonly found in the pediatric population, and in areas most subject to trauma, such as the lower lip, ventral tongue surface and the cheeks.

Mucoceles are the 15th most common oral lesion, with a prevalence of 2.4 cases per 1,000 people.[5] Mucoceles of the gland of Blandin-Nuhn are considered uncommon and have a reported prevalence that varies. Epidemiological studies have reported incidences of mucoceles involving the tongue that range from 2.4% to 18.3% of all mucoceles.[2,5-8] After the lower lip, the ventral tongue is the second most likely location for a mucocele.

Classically, the mucocele appears as a fairly well-circumscribed, oval, fluctuant, painless mass. Its usual superficial submucosal location gives it a translucent, bluish appearance, caused by the contained colloid–like secretion that acts to scatter light, the Tyndall effect. Increased depth leaves the overlying mucosa with no obvious color change. Because the mucocele is thin-walled and close to the surface, it tends to rupture. Recurrence is frequent, but, occasionally, the mucocele can disappear spontaneously.[1] Mucoceles of the gland of Blandin-Nuhn vary in size.[5]

Superficially placed, they lend themselves to complete excision, which is the standard accepted treatment.[4,9-11] Excision of the cyst-like mass should include the immediate underlying, contributing glandular tissue and its associated duct.

From Salivary Gland Center, Division of Oral and Maxillofacial Surgery, Columbia University College of Dental Medicine, New York, NY.

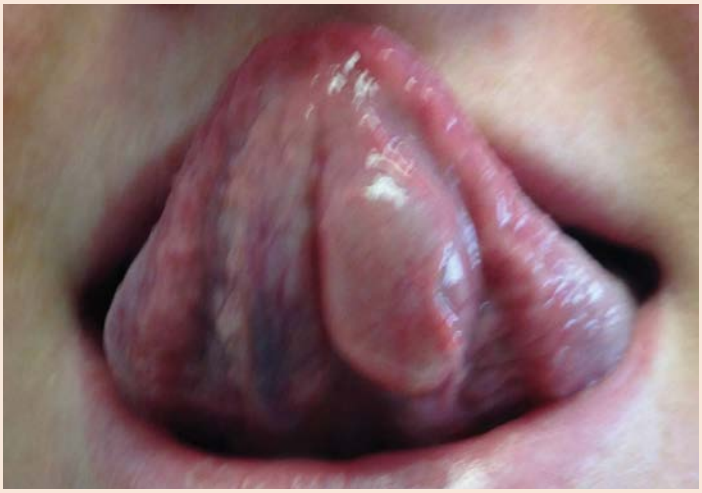

Figure 1. Clinical appearance of mucocele of Blandin-Nuhn gland.

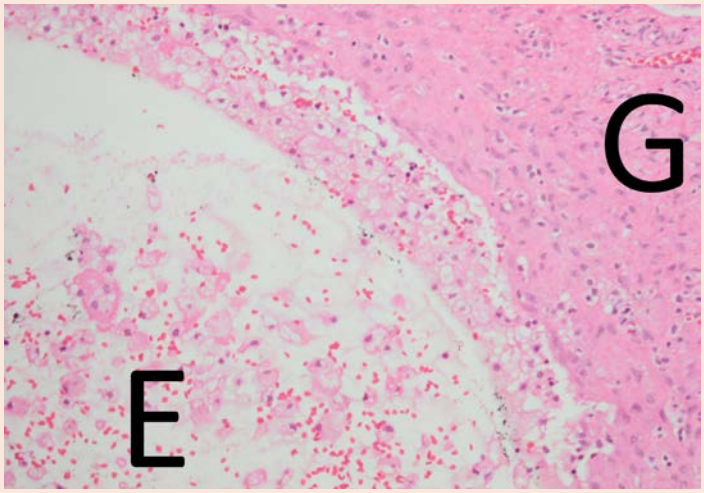

Figure 2. Microscopic photo of Blandin-Nuhn mucocele. Extravacated secretion (E) surrounded by wall (G) of granulation tissue. (H and E stain; magnification x 200)

Case Report

A 22-year-old female, in excellent health, presented to the Salivary Gland Center at the Columbia University College of Dental Medicine because of a swelling in the anterior aspect of the ventral tongue (Figure 1). The size and physical configuration of the lesion caused some discomfort, but only during normal mastication and speech. The swelling had developed two months previously. There was no history of any past swelling episodes.

Intraorally, a well-circumscribed swelling involving the left ventral tongue surface and measuring 3.5 cm x 1.6 cm was evident. The overlying mucosa had a bluish tinge. Palpation indicated that the swelling was not painful and that it contained fluid. A clinical diagnosis of a mucocele of the gland of Blandin-Nuhn was made. Surgical removal of the pseudocyst with its associated glandular tissue was accomplished using local anesthesia. The lesion was excised to the level of the genioglossus muscle, which served as a surgical cleavage plane. After undermining the margins of the wound, primary closure was attained.

Healing was uneventful, and the sutures were removed five days postoperatively. Microscopic examination confirmed the clinical diagnosis of a mucocele resulting from mucous extravasation (Figure 2).

Six months postoperatively, healing was complete and no recurrence had occurred.

Discussion

The tongue houses groupings of three minor salivary glands: von Ebner glands on the dorsal surface of the tongue in close relation to the circumvallate papillae; Weber glands in the root of the tongue and along the tongue's posterior lateral border; Blandin-Nuhn glands in the anterior ventral tongue.[12] The gland of Blandin-Nuhn, with five to seven ducts, is a mixed mucus/serous gland covered by a thin mucous membrane. Each duct exits from a lobe of the gland[5] and opens into the oral cavity just medial to the plica fimbriate, a fringed fold of mucous membrane on the undersurface of the tongue lateral to the lingual frenum.

Mucoceles of the gland of Blandin-Nuhn have variously been reported to have an equal gender predilection[2] or a 3:1 female

predominance.[5] They can occur at any time during an individual's lifetime,[11,13] but the large majority (76%) of mucoceles occur in the first two decades of life.[2] Our case report of a 22-year-old female is not rare, but it is uncommon among patients with Blandin-Nuhn mucoceles.

If a Blandin-Nuhn mucocele is left unattended, an increase and decrease in size may be observed as the lesion cyclically ruptures and accumulates mucin.[4] In order to stop the cycle, and also eliminate the oral discomfort or speech impediment caused by the swelling, proper identification of the lesion and referral for an oral surgical procedure are required.

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Sequential Intracranial and Pulmonary Abscesses with *S. intermedius* as an Extension of Severe Maxillofacial Infection

A Case Report and Review of the Literature

Michael Y. Nagai, D.D.S., M.D.; Robert Miskimen, D.D.S.; Matthew Rossen, D.M.D., M.D.

ABSTRACT

Intracranial abscesses are rare infections of the brain parenchyma that can arise from a variety of infectious sources, including, on rare occasion, the dentition and its supporting structures. Morbidity and mortality related to intracranial abscesses, although significantly improved from a historical perspective, remain quite high. The unfortunate case of Deamonte Driver, a 12-year-old boy from Maryland who succumbed to complications of an intracranial abscess related to uncontrolled dental disease, spurred a national conversation about the need for improved access to care.[1,2] Considering the number of people in the United States without reliable preventative care and limited access to oral health providers, discussions concerning intracranial extension of maxillofacial abscesses remain relevant. Here we present a case of a patient with an intracranial abscess that was preceded by a pulmonary abscess with the same causal organism one month prior. The etiology was determined to be odontogenic in nature.

A 65-year-old Caucasian male was taken to a local hospital via ambulance following a deterioration in mental status and a presumed seizure that lead to an unwitnessed fall. The patient was immediately transferred to our facility for a higher level of care and definitive treatment. After further history was obtained through the family, it was believed that the patient had undergone an acute change in his mentation that had progressively declined over the days leading up to his admission, along with an abrupt transformation in his appearance, with worsening lethargy and decreased responsiveness.

Initial workup revealed a left hip fracture and an intracranial lesion, suspicious for metastatic disease or abscess. MRI imaging demonstrated multiple large-ring-enhancing lesions in the right frontal lobe (Figures 1, 2). The patient was evaluated by the neurosurgery service and brought to the operating room for an emergent, open right frontotemporal craniotomy and drainage of what appeared to be an abscess. At the time of the initial surgery, multiple specimens were sent for culture and sensitivity.

Following the craniotomy, a search for the source of the abscess was undertaken, and a maxillofacial CT was obtained. This demonstrated significant opacification of the left maxillary sinus, along with multiple carious teeth and corresponding periapical radiolucencies (Figure 3). As a result, the oral and maxillofacial surgery service was consulted for management of the sinus disease and failing dentition with a presumption that the source of the infection was related to his chronic sinus and dental pathol-

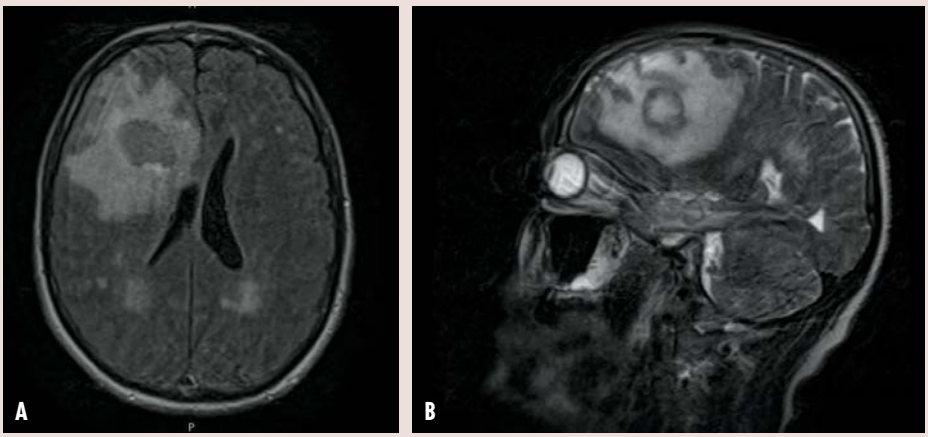

Figure 1. MRI brain FLAIR revealing large enhancing 3.5 cm x 2.2 cm right frontal lobe mass with significant vasogenic edema. A. Axial slice. B. Sagittal slice.

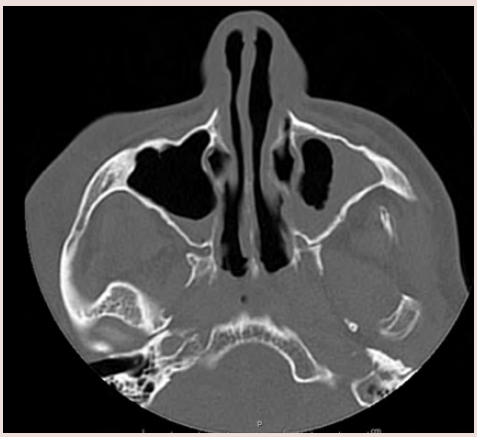

Figure 2. CT face reveals thickened mucosa of maxillary sinus with air fluid levels.

ogy. Cultures obtained during the neurosurgical procedure were positive for *Streptococcus intermedius*, likely odontogenic in origin.

The patient was returned to the operating room, where his remaining dentition was extracted, along with a left antrostomy via Caudwell Luc approach, in an effort to obtain source control of the infection. Culture data from the sinus was positive for coagulase negative *Staphylococcus* species, yeast and *Diphtheroids*. The patient was maintained on aggressive antibiotic and antifungal therapies and slowly improved over the ensuing days. He was discharged on hospital day seven to a local rehabilitation facility with marginally improved cognitive and functional deficits.

A unique aspect to this case was that one month prior to his presentation, the patient underwent a CT-guided biopsy of a right upper lobe lung mass. This ultimately returned as a focal abscess cavity with culture positive for *Streptococcus intermedius*. The patient was aware of his dental status and the need for extensive oral rehabilitation but continued to delay definitive therapy. According to his family, the patient had an extreme fear of the dentist and had "poor experiences as a child," which lead to his avoidance of necessary treatment.

Discussion

Over the past 60 years, the incidence of intracranial abscess has remained stable, at approximately 1 in 100,000,[3,4] while during the same time period, the rate of full recovery has more than doubled to 70% and the case fatality rate has decreased four-fold to 10%.[4] The significant improvement in outcomes may likely be attributable both to the development of advanced imaging techniques (CT, MRI, etc.), which allow for earlier diagnosis and better characterization of lesions, and to a shift in the prevailing treatment dogma of conservative management towards an excision of the infectious nidus, along with surgical drainage as an important adjunct to medical therapy. Broad spectrum intravenous antibiotic coverage has also led to a significant reduction in perioperative morbidity.[5]

The most commonly implicated organisms, making up a full third of those identified, are *Streptococcus species*, of which *viridans streptococci* are most common, lending particular suspicion to odontogenic sources in any case of intracranial abscess. *Staphylococcus* and gram-negative enteric species are also quite common, and a polymicrobial culture is identified in 23% of cases;[4] however, of the three routes of infection recognized throughout the literature,[6] cases with a polymicrobial culture are more likely due to either direct contamination (trauma or surgery) or contiguous spread of a localized infection (otitis media, sinusitis, mastoiditis, fascial space infection, etc.). The third route, hematogenous spread from a distant site (septic embolism from endocarditis or PE, transient dental sepsis, etc.) is considered comparatively unlikely in such cases due to the ability of the blood brain barrier to exclude most pathogens.[7]

Three viridans streptococci species, namely *S. anginosus*, *S. constellatus* and *S. intermedius* (the causative organism identified in the present case), make up the *Streptococcus anginosus* group (SAG, also known as *S. milleri* group). Though all three species have a propensity for causing suppurative infections within soft tissues, *S. intermedius* possesses the unique ability to produce both sialidase and hyaluronidase virulence factors, which digest host tissues into small nutrients utilized in bacterial growth.[8] The organism is frequently implicated in intracranial abscesses owing to the antigen I/II surface proteins present within its polysaccharide

capsule, which assist in binding to fibronectin and laminin of brain tissue.[9] Although it can also be found at various mucosal sites in the genitourinary and gastrointestinal systems, *S. intermedius* has been recognized as an inhabitant of the oral cavity since 1975.[10] In that same year, it was first implicated in a case of intracranial abscess.[11]

A concomitant *S. intermedius* infection of the lung (e.g., abscess, emphysema, bronchiectasis) has previously been identified as a predisposing factor for intracranial abscess.[12] Similarly, multiple reports have identified a prior liver abscess containing the bacterium as a risk factor;[13,14] however, such reports of odontogenic liver abscess are much sparser in the literature, possibly because of the differing routes of infection. The lung is most commonly infected by SAG bacteria through aspiration of oral secretions,[15] whereas hematogenous seeding after transient dental sepsis is the likely route of infecting the liver.[16] Other risk factors for developing an intracranial abscess containing SAG organisms include mucosal infection (paranasal sinusitis, periodontal diseases), pneumonia, alcohol abuse and diabetes.[17] In addition, unoperated cyanotic congenital heart disease (CCHD) accounts for 25% to 46% of intracranial abscess cases associated with *S. intermedius*, through a variety of proposed mechanisms.[9]

A particular concern for the case presented, and similar cases throughout this country, are the barriers to care that remain a significant problem, with potential delay in diagnosis and/or treatment. In 2000, the U.S. Surgeon General described the condition of oral health disparities in certain populations as a "silent epidemic."[18] Several factors play a role in limiting access to care with regard to oral health. These include financial considerations, non-coverage, poor oral health literacy, psychological factors and patients with special needs.[19,20] Fewer people have dental insurance than medical insurance, which seems to coincide with some views that oral health remains a luxury. One study looking at the decline of dental care utilization in adults from 2001 to 2010 found it due in large part to non-coverage and changes in insurance status.

Interestingly, there has been an increase in dental care utilization among publicly insured children from 2000 to 2011.[21] In 2013, Wall et al. reported that the main factor for declining adult utilization was a shift in pattern of dental benefits. The portion of adults with private insurance benefits decreased, while those without dental benefits increased. Severe morbidity and mortality can result from deferring dental problems, as this case plainly demonstrates. The number of emergency room visits for non-

traumatic dental complaints is increasing, and the majority of hospitals in the United States are not equipped with dental centers for definitive treatment. This results in temporizing measures, such as prescription narcotics and antibiotics.

Dental visits in the ED as a percentage of total ED visits increased from 1.06% in 2000 to 1.65% in 2010.[22] Additionally, healthcare costs associated with seeking dental care in the emergency setting is significantly higher than a routine preventative dental visit. From 2011-2013, Saloman et al. analyzed the effects of restricting preventative dental services to emergency-only treatment before and after passage of the SMART (Save Medicaid Access and Resources Together) Act in Illinois. After restriction of services, patients were more likely to have complex surgical interventions, increased admission lengths, and the total cost of care increased by $1.6 million.[23] In the case presented, dental treatment was delayed for many years, resulting in a life-threatening presentation, and the total burden to healthcare cost increased significantly.

Conclusion

We have presented a case of an intracranial abscess associated with *Streptococcus intermedius*, odontogenic in origin, preceded by a lung abscess of the same causal organism that was the unfortunate harbinger of future spread to the brain. The route by which the brain was seeded is not clear, whether by septic embolism from the lung or by hematogenous spread through transient dental sepsis. *S intermedius* was not confirmed to be within the oral tissues, neither were the two abscess cultures matched by DNA homology; however, both the presence of SAG organisms within the oral cavity and the mechanisms by which the brain and lung may be seeded are well documented in the literature.

While the incidence of CNS infection, including all cases of meningitis, subdural empyema, cerebritis, encephalitis, septic thrombophlebitis, and intracranial abscess from an odontogenic source, is frequently cited to be 1% to 2%,[24] more recent evidence suggests this etiology may have been underreported and should not be overlooked in cases of intracranial abscess.[4,6] The challenge will remain that susceptible individuals may not realize the pending danger of their dental problems and seek appropriate management. Patient education and routine preventive care will continue to be the cornerstone in avoiding this infrequent, yet devastating, condition.

Queries about this article can be sent to Dr. Rossen at rossenmsr@gmail.com.

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