

Management of bilateral odontogenic infections in a systemically compromised sickle cell trait child-a case report.

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Abstract

Patient with sickle cell trait (SCT) inherits one sickle cell gene (HbS) from one parent and one normal gene (HbA) from the other parent making them heterozygous. People with SCT usually do not have any of the symptoms of sickle cell disease (SCD), but they can pass the trait on to their children and has increased susceptibility to infections.

Chronic inflammation of pulpal origin is one of the reasons for an extra oral sinus of odontogenic origin. Children exhibit many pathological lesions involving the jaw bones. Among them, odontogenic cysts constitutes an important aspect as they are derived from epithelium associated with the development of the dental apparatus. The most common among these lesions of inflammatory origin are radicular cysts. Radicular cysts are known to be the most common cystic lesions of the jaws. Because any

infection in a patient with SCT disease can become life-threatening. We present a special case of managing two infections in a sickle cell trait child patient to call attention to this serious problem and share our clinical management and experience, especially with surgeons who practice in areas with limited resources. The early diagnosis and the management of Odontogenic infections and cysts for patients with blood disorder requires medical consultation and cooperation between dentist and physician to avert adverse effect in sickle trait child.

Keywords: Sickle cell trait (SCT), Radicular cyst, draining, sinus, benign.

Introduction

Patient with sickle cell trait (SCT) inherits one sickle cell gene (HbS) from one parent and one normal gene (HbA) from the other parent making them heterozygous. People with SCT usually do not have any of the symptoms of sickle cell disease (SCD), but they can pass the trait on to their children. The prevalence of sickle cell carriers among different tribal groups varies from 1 to 40 per cent and sickle cell trait constitutes 2.1%.^[1]

The term sinus tract refers to a tract leading from an enclosed area of inflammation to an epithelial surface. It also states that instead of dental fistula, the more proper term is sinus tract.^[2] A chronic inflammation of pulpal origin is one of the reasons for an extra oral sinus of odontogenic origin.^[3] Microorganisms and their by-products, which are present in the periradicular area, might perforate the cortical plate with the infection draining onto the mucosal or cutaneous surface following the path of least resistance, after exiting from the necrotic pulp canal system.^[4]

Children exhibit many pathological lesions involving the jaw bones. Among them, odontogenic cysts constitutes an important aspect as they are derived from epithelium associated with the development of the dental apparatus.

The most common among these lesions of inflammatory origin are radicular cysts.^[5] Radicular cysts are known to be the most common cystic lesions of the jaws. They comprise about 52.3% of jaw cysts and 62% of cysts of odontogenic origin. They develop from cystic degeneration of cell rests of Malassez, secondary to inflammatory stimulation arising from the necrotic pulp. Surprisingly, those arising from primary teeth are considered rare, with the prevalence of only 0.5%.^[6]

We present a special case of managing two infections in a sickle cell trait child patient.

A 11-year-old female reported with a draining sinus on the lower right side of the face. Sinus was present since past 3 days before visiting the clinic. Patient was having a past dental history of previously done access opening with both lower permanent mandibular molars. Patient reported with medical history of sickle cell anaemia. Oral hygiene status was poor for patient, and a maternal history of sickle cell anaemia. On extraoral examination, on inspection, a diffuse, nontender, and bony hard swelling in lower one third region extending superio-inferiorly 2cm away from ala-tragus line upto lower border of mandible and antero-posteriorly 2cm away from corner of mouth extending posteriorly till the angle of mandible was noticed on the lower right side of the face (Fig1). Also, draining sinus was evidently noted with mild restricted mouth opening. On extraoral examination left side seems to appear normal. On palpation right side was febrile, mild tenderness and soft to firm in consistency. Intraoral examination revealed grossly destructed mandibular deciduous molar and permanent molars (fig2). Buccal Vestibular obliteration was noticed on both the sides. On investigation the panoramic radiograph showed periapical radiolucency with lower right permanent molar involving pulp. A well defined unilocular radiolucent lesion with smooth, well-defined borders measuring about 3.5 cm × 4 cm in

relation to lower left first permanent molar extending from lower left deciduous second molar upto ramus of mandible (anterioposteriorly); well defined radiolucency extends from 1cm above coronal portion with lower left mandibular second molar upto lower border of mandible which was roughly elliptical in shape (fig 3). The differential diagnosis for left side should include traumatic lesions, fungal and bacterial infections, neoplasms, chronic tuberculosis lesion, osteomyelitis, actinomycosis, and gumma of tertiary syphilis. A provisional diagnosis of chronic periapical abscess leading to submandibular space infection and cutaneous draining sinus and benign lesion with respect to molar-ramus region of left side was given. For treatment, emergency access opening with lower right permanent first molar followed by extraoral debridement of the area (saline, betadine, hydrogen peroxide) was done and patient was kept on antibiotic coverage. On the very next day, extraction with deciduous canine and molars was planned to remove source of infection and recalled after 1 week.

Further, management of cystic lesion was planned. On blood investigations, the haemoglobin level was found to be 9.5 gm/dl for which patient was advised to consult a physician and biopsy was planned. Aspiration was done followed by incisional biopsy. Before biopsy consent was signed from parents. Under all aseptic condition patient preparation was done. Local anesthesia was administered in left mandibular vestibule, crevicular incision was given from lower left second deciduous molar till posterior region. Full thickness mucoperiosteal flap was raised 2.5 ml approx and fluid was aspirated. Then thinned out buccal bone of involved tooth was removed using surgical bone cutting bur. Cystic lining was separated from the surrounding bone and was excavated followed by extraction with 36 73 74 75. Thorough curettage and smoothing of bony margins followed by irrigation was

done. After achieving the haemostasis the distal releasing incision was sutured while the remaining cavity was left open to heal by secondary intension (fig4). The cavity was packed with soframycin gauge piece. Tissue was collected in sample bottle, aspirated fluid in syringe and sent in lab for testing (fig 5). Patient was under regular follow up. Histopathological report revealed non keratized stratified squamous epithelium of variable thickness overlying the connective tissue capsules. Epithelium shows arcading pattern. Connective tissue stroma consists of haphazardly arranged collagen fiber, few fibroblasts, numerous chronic inflammatory infiltrate chiefly plasma cells, lymphocytes and numerous budding capillaries, which was suggestive of radicular cyst. The cutaneous draining sinus was healed after 2 weeks (fig 6). The follow up opg was taken after 2 months in which satisfactory healing was observed (fig 7).

Discussion

Sickle cell trait is benign because patients do not get vaso-occlusive crisis; they have a better quality of life and mortality is the same as the rest of the general population. Sickle cell trait patient with an average HbS level of 34% or higher are more likely to get papillary necrosis compared to those who have HbS of 20%.^[8] In our case the HbS level was 38.8% with no such complications but had fewer systemic imbalance. The total bilirubin level was 0.10 mg/dl higher than the normal range, direct bilirubin level was 0.20 mg/dl higher than the normal range which was suggestive of mild jaundice for which she was kept on medication. SGOT (serum glutamic-oxaloacetic transaminase) value was 6.44mg/dl higher than the normal range. Alkaline phosphatase level showed decreased value of 156.77 with the normal range of 250.0-770.0.

Winstock described cutaneous lesions with dental infections. Kaban elaborated the path of spread of chronic

dental infections. Approximately 80% of the reported cases are associated with mandibular teeth and 20% with maxillary teeth.^[9,10] Submandibular region and chin are the most common location where this sinus tract are often formed.^[11] For this case, both the sides pathology was seen in mandible and the submandibular area was infected. Patients with SCA should receive preventive dental care including oral hygiene instructions, information on a proper diet to help control tooth decay, proper tooth brushing and flossing technique, as well as fluoride gel applications.^[12] Here, we have done oral prophylaxis, sealants for permanent molars and treated all the carious tooth to prevent all the source of infection.

Mandibular extra-oral sinus is often misdiagnosed with suppurative osteomyelitis. It is most often associated with a dental pathology. By proper diagnosis and treatment we can minimize patient discomfort and complication like sepsis and osteomyelitis.^[13]

Acute periapical abscess is drain through a path of least resistance intra orally or extra orally by making a sinus tract. It can also spread to the deeper oral tissue and caused space infection. Chronic periapical abscess may remain asymptomatic for longer duration of time.^[14] The evaluation of a cutaneous sinus tract must begin with a thorough patient history and awareness that any cutaneous lesion of the face and neck could be of dental origin.^[15]

Root-canal therapy is the treatment of choice if the tooth is restorable. Extraction is indicated for nonrestorable teeth.^[16] For this case , we have taken detailed case history, initial debridement and cleaning of the sinus extraorally through irrigation with betadine and saline and extraction of offending primary teeth and root canal of permanent right mandibular molar followed by dressing .

The following features can help in confirming the diagnosis of radicular cyst.

- There should be evidence of carious/traumatized/endodontically treated tooth
- Loss of lamina dura around the roots of the suspected tooth
- Follicular space around the permanent successor is intact and clearly visible.^[17]

The present case clinical radiographic features were in accordance with these criterias.

As odontogenic or orofacial infections are caused by a mixed flora of aerobes and anaerobes, bacterial synergism enhances growth of different types of organisms. Consequently, amoxicillin in combination with clavulanate acid is the better empirical choice of antibiotics, Thus, the dose and duration of treatment depends on the general condition of the patient and also by the resolution of the infection, with on average approximately seven to twelve days.^[20]Protein (g/ dl) range for radicular cyst is 6.1-11.7.^[18]. In this case, the report of the aspirated fluid from the infected cavity of left side showed that the protein content of the fluid was 6.29 mg/dl which was suggestive of radicular cyst. In case of radicular cyst, complete enucleation of the cyst and preservation of permanent successor is a suitable treatment option.^[19]

In the present case cystic lining was enucleated and open dressing was given with soframycin and was changed every 2 days to heal by secondary intension.

Conclusion

Odontogenic infections and cysts are of concern for surgeons, physicians and dentists because they are commonly seen in clinical practice, challenging to manage, and can lead to fatal complications if not treated in a timely manner. Early diagnosis is important to avert adverse effect. The diagnosis and the management of Odontogenic infections and cysts for patients with blood disorder requires medical consultation and cooperation

between dentist and physician. All dental appointments should be short, stress free. Long and complicated procedures should be avoided. With regular follow-up with the dentist and physician, we can assure to manage systemically compromised patients.



Fig 1: Pre operative extraoral picture showing swelling and draining sinus on lower right side of mandible.



Fig 2: Grossly destroyed 74 75 84 85 36 46



Fig 3: preoperative OPG



Fig 4 : Immediate post operative image.



Fig 5a : cystic tissue removed.



Fig 5b: Aspirated fluid collected in syringe



Fig 6: Healed cutaneous draining sinus after 2 weeks.



Fig 7: follow up OPG after 2 months

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