Case Report

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Acquired Cleft Palate Associated with Febrile Illness

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ABSTRACT

While congenital cleft palate is thought to be an interaction of hereditary and environmental factors, acquired cleft palate (ACP) is caused commonly by cocaine inhalation in western countries. Other causes include infective (Tb, tertiary syphilis); trauma; connective tissue disease (SLE, vasculitis Wagner's granulomatosis; sarcoidosis and idiopathic. An uneducated 45-year-old presented with febrile skin eruption which evolved into skin necrolysis and gangrene. This occurred following the ingestion of quinine sulphate 650 mg orally, tds, for seven days in a remote village in Sudan. This was treated conservatively with antibiotics and steroids resulting in healing by fibrosis. The patient was presented with fixed drug eruption 48 hours after the ingestion of quinine for presumed Plasmodium falciparum malaria in our endemic area. On general examination it was found that she is having an incidental cleft palate that she is not aware of its presence. On further inquiry she and her family member were unaware of any past history of hyper-nasality, speech problems, feeding problems, repeated upper respiratory infection and insisted that the main reason for her was to seek medical advice related to her skin problems. However, she admitted the presence of hyper nasality at the time of admission. Having knowing their illiterate and poor social background, we had no option but to confirm or exclude the presence of acquired cleft palate.

Keywords: Cleft Palate; Acquired; Idiopathic

Introduction

Syphilis and Cocaine abuse has been described as a cause of acquired palatal perforation in Europe and North America [1]. Maternal epilepsy and drugs administered in pregnancy such as diazepam, steroids, and phenytoin are known causes too [2]. Febrile illness associated with a palatal perforation in an African patient has not been reported.

Case Report

A 35-year-old female from al kamleen (central Sudan) presented to a local hospital with a 3 days history of low grade fever, generalized body ache and maculopapular skin rash over the front of her thigh. Being from a highly endemic area of malaria a presumptive diagnosis of malaria was made and as such she was admitted and received oral quinine, On the fourth day no improvement took place and the non-itching maculopapular rash spread to involve all of her lower limb. Two days later she developed nausea, vomiting and diarrhea that lasted for 48 hours. She consulted the physician at Soba University Hospital where she was admitted. She was prescribed her oral prednisolone 60 mg daily, hydrocortisone 100 mg six hourly iv for seven days. This was followed by extensive ulceration with superadded secondary infection in the areas of rash distribution and spread to involve the buttocks. She was put on IV ceftriaxone; heeling took place with scarring and fibrosis. Clinical history after the above-mentioned medication revealed that she was unaware of the presence of her cleft palate. On clinical examination She looked unwell but with no pallor, jaundice, or cyanosis. BP was normal at 130/80mmhg, her pulse was normal at 80 beats / min. cardiovascular, respiratory, neurologic, and abdominal examination revealed no abnormality. Head and neck examination revealed palatal perforation measuring 5x 4 cmm (soft palate). Investigations included: Complete blood counts (CBC), leukocytosis at 17,400. Erythrocyte sedimentation rate (ESR) at 119 mm/hr.

Urine analysis demonstrates pus cells count number of 10-15 / HPF and Red cells count number of 2-4 / HPF. Blood film showed neutrophilic leukocytosis. Normal red cells and platelets. Renal function had the following finding: NA 129 mmol/l, k 2.9 mmol. l, creatinine 0.6 mg / dl. Immunology screens for HIV, ANA, and anti-double stranded DNA test, were all negative. Skin swab grew staphylococcus aureus sensitive to ceftriaxone, gentamycin, cephalexin, and methicillin (Figures 1-3). Vascular biopsy came back with no evidence of vasculitis. Skin biopsy demonstrated excessive scarring and fibrosis. She settled on conservative treatment of steroids, antibiotics, and dressing. She underwent closure of her cleft palate (Table 1).

Table 1: Side effects of quinine sulphate.

SYTEM	SIDE EFFECT
Hematologic	Purpura, neutropenia, thrombotic thrombocytopenic purpura-hemolytic uremic syndrome, disseminated intravascular coagulation, petechiae, and ecchymosis.
Dermatologic	Flushing, pruritus, and skin rashes. Fixed drug eruption (nummular skin lesion) and fatal cutaneous vasculitis.
Gastrointestinal	abdominal pain, nausea, vomiting, diarrhea, and gastrointestinal upset.
Renal	Renal failure secondary to thrombotic thrombocytopenic purpura-hemolytic uremic s3. Syndrome.
Respiratory	Asthma symptoms, hemoptysis.
Ocular	Visual disturbances including blurred vision with scotomata, photophobia, diplopia, diminished visual fields, disturbed color vision, and blindness.
Cardiovascular	Cardiac dysrhythmias, including prolongation of the QT-interval.
Hepatic	Elevated alkaline phosphatase, lactate dehydrogenase, aspartate aminotransferase, alanine aminotransferase, and gamma-glutamyltranspeptidase.
Nervous system	Apprehension, restlessness, confusion, syncope, dizziness, vertigo, tinnitus, hearing loss, and nystagmus.
Metabolic	Hypoglycemia and electrolyte imbalance.
Other	Mucosal bleeding (gingival, gastrointestinal, epistaxis).

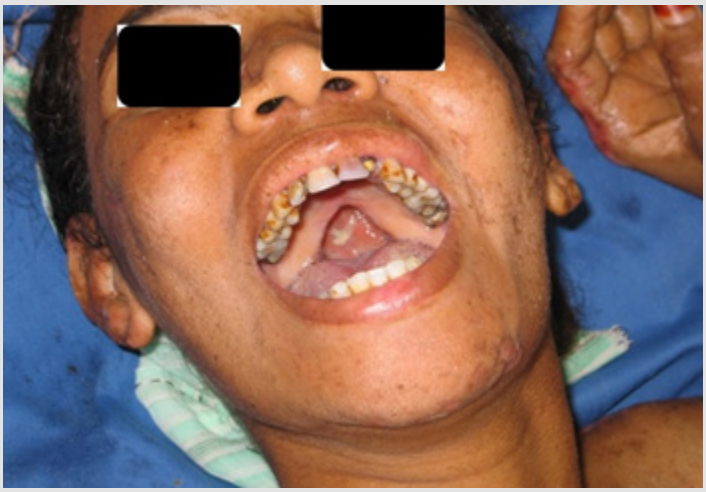

Figure 1: Congenital Palatal Perforation in a 35 years old.

Figure 2: Necrolysis of the Skin with Gangrene.

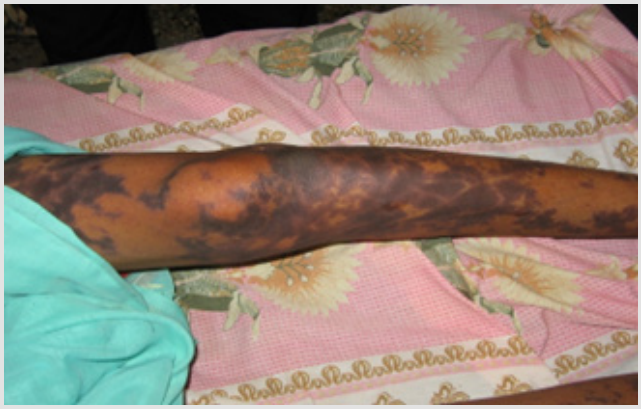

Figure 3: Healing after administration of oral prednisolone 60 mg daily, hydrocortisone 100 mg six hourly IV for seven days and intravenous ceftriaxone.

Discussion

Causes of Acquired cleft palate (ACP)include trauma, infections (TB, tertiary syphilis, Mucor mycosis), neoplasia (lymphoma, carcinoma), collagen vascular disease (SLE, Wegener's granulomatosis), sarcoidosis, idiopathic and cocaine abuse [2]. While cocaine abuse is not widely recognized in Sudan, syphilis and collagen vascular disease are common [2]. Palatal perforation due to syphilis and cocaine abuse has been reported (m. k.bains, m. hosseini) [1]. But perforation following a febrile illness in adults has not been reported in Sudan or anywhere else in Africa. It is uncommon for palatal fistula to be detected in individuals who have not undergone surgery, and only sporadic cases have been reported [3]. A complication after septoplasty procedure in a misdiagnosed sub mucous cleft palate case with the formation of post-operative Palatal fistula [4]. Cleft Palate Secondary to an Ingested Foreign Body (coin)in the nasopharynx has been reported by kanotra emphasizing the importance of evaluation of the nasopharynx in order not to miss such a foreign resulting in (ACP) [5]. Cocaine is a potent vasoconstrictor that can potentiate tissue necrosis when used chronically. Endonasal administration is the usual access route for cocaine, subjecting the contained anatomic structures to particular risk [6]. Ali has cited that "palatal perforation is a rare complication of septoplasty [7]. Long-standing cocaine use can jeopardize the integrity of the turbinates, nasal floor mucosa, and ultimately the maxilla and palatal musculature. Frank perforations into the oral cavity, from erosion through the nasal cavity above, can also occur especially after undocumented retained nasal pack [8].

Following septoplasty, complications such as poor quality of sleep, difficulty in breathing, decreased oxygen concentration and toxic shock syndrome can be encountered. Other possible complications include hemorrhage, vestibulitis, hematoma, adhesions and septal cartilage perforation [9]. Fixed drug eruption skin with lesions has been reported in a 23-year-old female following exposure to quinine in tonic water. An open oral challenge, approved by the patient, with 30 mg quinine sulfate triggered the appearance of pruritus, ery-

thema, and edema at the usual sites within 40 minutes of ingestion of the dose. Dermatologic side effects have included flushing, pruritus, and skin rashes. Fixed drug eruption (nummular skin lesion) and fatal cutaneous vasculitis have been reported [10]. Other side effects of quinine sulphate are tabulated in 1:1.

Conclusion

At the time of the patient admission, it was not clear to the patient or us whether this is a case of acquired cleft palate or a congenital one. The size of this cleft palate and the negative investigation confirmed this is probably a case of congenital cleft palate associated with fixed drug eruption after oral quinine for falciparum malaria rather than the initial suggestion of acquired cleft palate .The first message to take home is that in illiterate, low social class individual with a cleft palate in Africa the history can be an issue with confusion and uncertainty whether this is acquired or congenital cleft palate. In which case full investigation will clarify this issue. The second message is that many adults with poor education and low social class are unaware of the very presence of a cleft palate as it is concealed by its presence inside the oral cavity. The third message is that; cleft lip is a reason to create worry and anxiety even in illiterate families as the cleft lip is visible to the family members unlike isolated cleft palate. This case report is a stimulant to educate the public about early recognition of cleft palate and early seek for medical advice. As we come to see more and more patients with cleft palate at the age of 35 and above in remote rural areas in Africa.

Conflict of Interest

Authors declare that they have no conflict of interest.

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