

“Klinefelters Syndrome” Presenting with Deep Neck Fascial Infection with Uncontrolled Diabetes - A Case Report

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Abstract

Klinefelter syndrome is chromosomal abnormality seen in males, linked with hypergonadotrophic hypogonadism with increased risk to metabolic syndromes; usually diabetes and cardiovascular diseases (CVD). Diabetes is a prevalent metabolic disorder which compromises the individual's immunity and makes them vulnerable to infection. Oro-fascial infections in the maxillofacial region are very frequently diagnosed in Diabetes Mellitus patients with uncontrolled glycaemic levels. Delay in treatment can cause spread into various Oro-fascial spaces which could be fatal. Prompt surgical treatment with medical management should be strictly undertaken. Here we discuss a patient with klinefelter syndrome reported with potential deep neck space infection with uncontrolled glycaemic index, both surgical and medical management been discussed for the same.

Keywords: Deep Neck Space Infection; Diabetes Mellitus; Klinefelter Syndrome (KFS); Medical Management; Surgical Drainage; Cardiovascular Disease; Endotracheal Intubation

Introduction

Odontogenic Oro-fascial infections are the most commonly seen infectious disease in head and neck region both in diabetic and non-diabetic patients though commonly prevalent in diabetic patients. Diabetes is characterized by undesirable elevation in plasma blood glucose level associated with changes in protein, lipid levels where absolute or relative lack of insulin hormone is the causative factor [1]. Klinefelter syndrome is a chromosomal abnormality that shows supernumerary X chromosome, aneuploidy with hypergonadotrophic hypogonadism. Phenotype of KFS is 47XXY, addition of extra X chromosome. In comparison with the general population, patients with KFS shows high incidence of cardiovascular diseases and diabetes. In the same way, low androgens, and low HDL cholesterol concentrations in KFS results in increased body fat [2]. Infections in diabetic patients is mainly due to increased glycaemic levels which impairs

host defence mechanisms, other degenerative complications such as microangiopathy and compromised leucocyte migration due to thickening the capillary basement membrane [3]. It has been documented that in odontogenic oro-fascial infections, streptococcus and Klebsiella pneumonia are the causative organisms. Those that affect the diabetic patients differ from non-diabetic patients [4].

Case Presentation

47-year-old male patients reported to the Department of Oral and maxillofacial surgery, College of dental sciences and hospital, Davangere. Presented with chief complaint of pain and swelling over his left side of the face from past 6 days, with acute localised, intermittent dull aching pain over lower left back tooth region since two weeks KFS and swelling since 6 days. Pain aggravates on taking food, reduced mouth opening, pus discharge was noted

from mouth since 4 days, no history of fever noted, no history of difficulty in swallowing or breathing noted, patient consulted Primary health care centre for the same from there he was referred to our department for further management. On medical history k/c/o type II diabetes mellitus, h/o right side mastectomy one year back for the suspicious carcinoma of breast, blood glucose level at the time of admission was 351mg/dl and ESR - 25mm/hour, TLC - 16480cells/cumm. Endocrinology reference was taken for the uncontrolled glycaemic levels, incidentally patient was found to have Klinefelter syndrome. On examination: Ill-defined diffused swelling extending from left supra orbital rim to lower border of mandible on left side, Skin overlying is stretched and color change is visible, puckering over the left zygomatic region (Dumbbell shape swelling featuring superficial temporal space), Little obliteration of the Nasolabial fold, mouth opening was 5-10mm, on palpation swelling is soft, non-fluctuant, compressible and tender, Pitting edema present, Localized rise in temperature, No edema or tenderness towards the neck or sternal region, Diagnosis made was Left buccal, submandibular, submental, infra orbital, peri orbital, temporal space infection secondary to 38.

Discussion

Potential deep neck space infections are to be managed by hospitalization with both surgical as well as medical management especially in medically compromised patients. They should be treated under general anesthesia with thorough workup.

Pre-operative workup: complete blood profile (CBC, RBS, RFT, LFT, SE, AEC, Coagulation profile, Serology, Blood grouping) including lipid profile as patients with KFS more prone for cardiovascular events and ESR, CRP which are the indicators of infection. Anesthetic evaluation to assess the airway and difficulty in intubation. Here fiber optic intubation was done as mouth opening was 5-10mm, endocrinology reference was taken for increased glycemic levels and for KFS which advised no management from their side at present situation and nil contraindication for surgical drainage. The concern was the patient with KFS are prone diabetic with uncontrolled glycaemic levels which causes aggression of infection. So long-acting insulin therapy was preferred.

Intra-op considerations: In patients with potential deep neck space infections, antibiotic therapy should be started on admission and choice of drug regimen are penicillin's and metrogyl through

intravenous route. Patient can be kept nil per oral or can be given IVF. Here 10 units of human actrapid was put in 500 ml of DNS and given at rate of 60-70 ml/hour as per Endocrinologist's advise. Antibiotics were given 30 minutes before the induction, was draped in conventional manner and the infected spaces were explored thoroughly, irrigated with betadine and normal saline, corrugated rubber drain placement following the removal infection foci tooth (38). In postop recovery, the patient was not extubated till POD-1, as there was no considerable swelling after drainage any tracheostomy was not performed. Culture and sensitivity test was with the pus sample collected and the organism isolated was Klebsiella. Antibiotic sensitivity was confirmed for Piperacillin, Tazobactam and Amikacin.

Case pictures

A 47 year old male patient reported with fascial space infection involving Left buccal, submandibular, submental, infraorbital, periorbital and temporal space infection secondary to 38. Typical features of Klinefelter syndrome as gynecomastia, sparse body hair, obesity are manifested.

Figure 1a: Pre- operative.

Figure 1b: Post-operative.

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Conclusion

Klinefelter syndrome and its association with odontogenic oro-facial infection are rare and diagnosis of the KFS is mainly by strong suspicion with symptoms such as increased body fat, gynecomastia, less facial/body hair, diminished genitals. Prompt referral to specialty of Endocrinology is needed for uncontrolled glycemic levels hand in hand with surgical management by exploration of the spaces to remove the infection foci, invariably the surgical management is also related to reducing the uncontrolled glycaemic levels. Management of KFS patients with odontogenic oro-facial infections is not challenging with availability of multidisciplinary team approach. Broad spectrum antibiotics have a synergistic effect both prior and later to surgery as the local bioavailability of antibiotics will be more to the affected site after intervention. Patients with KFS are prone for uncontrolled diabetes which can aggravate the infection. There are no contraindications for surgical approach or medical management in patient with Klinefelter syndrome. So to conclude with, odontogenic fascial space infections have a higher magnitude of severity in KFS patients because of the uncontrolled Diabetes and the treatment protocol remains same as of any other patients.