Oro-Naso-Sino-Orbital-Cutaneous Fistula From Prolonged Cocaine Use

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Abstract

Presentation
We present the case of a 48-year-old man with nasal cellulitis and subsequent oro-naso-sino-orbital-cutaneous fistula from prolonged cocaine use.

Diagnosis
Initial laboratory investigations reported a raised white cell count (WBC) and C-Reactive Protein (CRP) and subsequently a positive atypical anti-neutrophil cytoplasm antibodies (ANCA) and positive anti-proteinase (PR3). Perihilar lung nodularity on chest imaging raised the possibility of a systemic autoimmune response. His urinalysis was positive for cocaine.

Treatment
He was commenced on Augmentin, Amphotericin B and Prednisolone. An obturator was created to manage the oro-nasal fistula. A subsequent naso-cutaneous defect was re-approximated. Daily nasal saline douche and abstinence of cocaine were recommended.

Discussion
Cocaine use in the community is rising and poses a challenge to multiple facets of our health care system.
Introduction

Cocaine is the most commonly used illicit stimulant drug in Europe. Ireland has one of the highest prevalences of cocaine use among young adults.\(^1\) Cocaine blocks the reuptake of norepinephrine and dopamine and provokes a sense of euphoria. Cocaine is commonly ‘cut’ or bulked up with levamisole. Levamisole is an immunomodulator and is used as an antihelminthic drug in veterinary medicine.\(^2\) It is the most common cocaine adulterant in Ireland, being present in 73% of samples analysed.\(^3\) It increases the euphoric effect of cocaine and is implicated in secondary ANCA vasculitis of the nose.\(^4\) Levamisole can cause cutaneous haemorrhagic bullae and necrosis of the skin.\(^5\)

Cocaine induced mid-line destructive lesions (CIMDL) are one facet of the spectrum of cocaine abuse. Prolonged cocaine use can not only induce septal perforation, but nasal destruction extending to the clivus, skin and palate.\(^6\) The case we present is one such extreme example of oro-naso-sino-orbital-cutaneous fistula.

Case Presentation

A 48-year-old male presented with six-day history of nasal cellulitis and swelling. He reported a four-month history of hyper-nasal speech, nasal congestion and nasal crusts on a twenty-year history of cocaine use. General inspection showed erythema and swelling over the skin of the left upper lateral cartilage and excoriation of the nares. Nasal examination showed copious crusts with complete loss of the nasal septum, turbinates and a 1.5cm hard palate communicating with the nasal cavity. Preliminary laboratory investigations showed a raised WBC 11.7 x10\(^9\)/L (4-11 x10\(^9\)/L) and C-Reactive Protein 62mg/L (0-5mg/L).

Computer tomography (CT) and Magnetic Resonance imaging (MRI) of sinuses showed near complete destruction of the bony and cartilaginous nasal septum and medial walls of both maxillary sinuses. There was erosion of the lamina papyracea bilaterally and floor of left orbit. (Figure 1)
Figure 1. T2 Weighted MRI Nose and Paranasal Sinuses: MRI demonstrating the oro-nasal fistula and loss of the septum and turbinates.

He proceeded twice to theatre for biopsy and re-approximation of the cutaneous defect respectively. Multiple nasal cavity biopsies were negative for malignancy and primary vasculitis was not demonstrated. Culture of the nasal biopsy material grew proteus mirabilis, and multiple other organisms. Invasive mucormycosis culture was negative in the submitted specimens.

The case was discussed at the Tallaght Vasculitis & Allergy Group multi-disciplinary meeting. He continued on oral prednisolone (40mg reducing to 7mg maintenance). An obturator was created to manage the oro-nasal fistula. The patient was discharged with a small pimple over his nose. He continued to use cocaine. He returned a week later with a significant larger defect. (Figure 2)

Figure 2. Clinical Photograph of Nose: Naso-cutaneous fistula.
Discussion

In this case report, we present a rapidly destructive process involving skin, nasal airway and oral cavity. The aetiology of muco-cartilaginous destruction secondary to cocaine is uncertain. It is postulated to be as a result of direct trauma, prolonged and recurrent vasoconstriction and bacterial colonisation. Lood and Hughes report that cocaine and levamisole may contribute to the development of ANCA by inducing release of inflammatory neutrophil extracellular traps with exposure of the neutrophil elastase autoantigen and B Cell-activating factor.

Nasal management of this atypical vasculitis is largely supportive. Consistently shown throughout the literature, cocaine abstinence is paramount to the management of this destructive process.

Authors have advocated surgical reconstruction be delayed for up to 24 months post cessation of cocaine ingestion. Palatal perforations may be treated conservatively with an obturator. We performed surgery on our patient due to the very obvious nasal cosmetic defect, and secondly to reduce the likelihood of crust formation.

This case serves to raise awareness among the ENT and wider medical community of the potential increasing complex local and systemic complications of cocaine use. An underlying or co-existing vasculitis should be considered in such patients. The management of these complex patients would benefit from a multi-disciplinary team approach.

Patient Consent:
Received

Declaration of Conflicts of Interest:
The authors have no conflicts of interest to disclose.

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