

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

S. Boyle<sup>1</sup>, M. Hussain<sup>1</sup>, C. Kirby,<sup>2</sup> S. Brennan,<sup>3</sup> L. Clarke,<sup>3</sup> R. Mullan,<sup>2</sup>  
D. Halpenny,<sup>4</sup> N. Conlon,<sup>5</sup> M.A. Little,<sup>6</sup> B.J. Conlon<sup>1</sup>, S. Abdulrahman<sup>1</sup>

1. ENT Department Tallaght University Hospital, Dublin 24, Ireland.
2. Rheumatology Department Tallaght University Hospital, Dublin 24, Ireland.
3. Pathology Department, Tallaght University Hospital, Dublin 24, Ireland.
4. Radiology Department, Tallaght University Hospital, Dublin 24, Ireland.
5. Autoimmune Department, St James Hospital, Dublin 8, Ireland.
6. Trinity Health Kidney Centre, Tallaght University Hospital, Dublin 24, Ireland.

### **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 nasocutaneous 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**

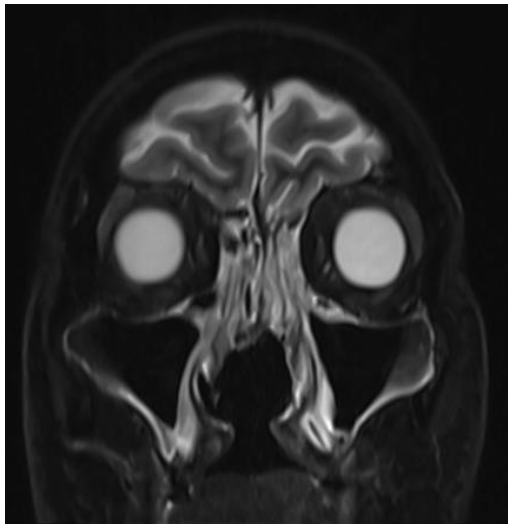
Cocaine is the most commonly used illicit stimulant drug in Europe. Ireland has one of the highest prevalence of cocaine use among young adults.<sup>1</sup> Cocaine blocks the reuptake of norepinephrine and dopamine and provokes a sense of euphoria. Cocaine is commonly 'cut' or bulked up with levamisole.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

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.<sup>6</sup> 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  $11.7 \times 10^9/L$  ( $4-11 \times 10^9/L$ ) 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 *serratia marcescens*, *streptococcus mitis*, *candida dubliniensis* and scanty *proteus mirabilis*. 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.<sup>7</sup> 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.<sup>8</sup>

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.<sup>9</sup>

Authors have advocated surgical reconstruction be delayed for up to 24 months post cessation of cocaine ingestion.<sup>9</sup> Palatal perforations may be treated conservatively with an obturator.<sup>10, 11</sup> 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.

### **Corresponding Author:**

Seamus Boyle, ENT SpR,

RCSI

E-Mail: [seamusboyle@rcsi.com](mailto:seamusboyle@rcsi.com)

## References:

1. Report ED. European Drug Report Trends and Developments. 2019.
2. (2022). NCfBI. Compound Summary for CID 26879, Levamisole. .PubChem
3. Geraldine O'Neill DCBL, Drugs Section. Purity of Illicit Drugs submitted to Forensic Science Ireland 2015. 2015.
4. Magliocca KR, Coker NA, Parker SR. The head, neck, and systemic manifestations of levamisole-adulterated cocaine use. *Journal of oral and maxillofacial surgery : official journal of the American Association of Oral and Maxillofacial Surgeons*. 2013;71(3):487-92.
5. Saikaly SK, Saikaly LE, Melamed MT, Norman RA. Levamisole-Induced Necrosis and Vasculitis. *Skinmed*. 2021;19(1):60-2.
6. Molteni M, Saibene AM, Luciano K, Maccari A. Snorting the clivus away: an extreme case of cocaine-induced midline destructive lesion. *BMJ case reports*. 2016;2016.
7. Trimarchi M, Bertazzoni G, Bussi M. Cocaine induced midline destructive lesions. *Rhinology*. 2014;52(2):104-11.
8. Lood C, Hughes GC. Neutrophil extracellular traps as a potential source of autoantigen in cocaine-associated autoimmunity. *Rheumatology (Oxford, England)*. 2017;56(4):638-43.
9. Lancaster J, Belloso A, Wilson CA, McCormick M. Rare case of naso-oral fistula with extensive osteocartilaginous necrosis secondary to cocaine abuse: review of otorhinolaryngological presentations in cocaine addicts. *The Journal of laryngology and otology*. 2000;114(8):630-3.
10. Colletti G, Allevi F, Valassina D, Bertossi D, Biglioli F. Repair of cocaine-related oronasal fistula with forearm radial free flap. *The Journal of craniofacial surgery*. 2013;24(5):1734-8.
11. Hofstede TM, Jacob RF. Diagnostic considerations and prosthetic rehabilitation of a cocaine-induced midline destructive lesion: A clinical report. *The Journal of prosthetic dentistry*. 2010;103(1):1-5.