

IN BRIEF

- Raises awareness of palatal perforations and the dental relevance of syphilis and cocaine abuse as rising problems for the profession.
- Explains the pathology and pharmacology behind the development of a syphilis or cocaine-induced perforation.
- Gives guidance for primary management including recognition, history, simple treatment and referral.
- Outlines secondary treatment options to seal or repair the defect with brief review of relevant case reports.

Palatal perforations: past and present. Two case reports and a literature review

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Syphilis and cocaine abuse are posing a growing public health problem on a global and national scale. Clinicians are increasingly likely to come across associated oral manifestations. We present two cases of palatal perforations caused by tertiary syphilis and cocaine abuse respectively. The literature review discusses issues specific to palatal perforations and those general to both conditions. The purpose of the report is to focus attention on syphilis and cocaine abuse as rising problems for the dental profession.

INTRODUCTION

The incidence of syphilis is rising worldwide.^{1,2} In the UK the number of new diagnoses increased by over 900% between 1996 and 2002.³ The resurgence is most marked in homosexual men and a series of large outbreaks have occurred particularly in London but also Manchester, Brighton, Newcastle-upon-Tyne, Dublin and Scotland.³ Recent interest has focused on the altered presentation in HIV positive men who are at greater risk of tertiary stage syphilis and have thus increased the likelihood of clinicians witnessing its signs and symptoms.⁴

Cocaine abuse is posing major public health problems worldwide.⁵ Rising levels of use seen in the USA are being mirrored in Europe with the UK showing its greatest increase in London and the South East, particularly among young people.^{5,6} It is speculated that this is associated with falling costs and increasing availability of the drug.⁶ The consequence to the dental profession is that practitioners are increasingly likely to encounter such patients. However they do not fit a classic

profile⁷ and so to identify them the health professional would need to recognise common signs and symptoms.

We report two cases of palatal perforation caused by tertiary syphilis and cocaine abuse respectively. We discuss their presentation, diagnosis and management and hope to raise awareness of the relevance of these conditions to the dental profession.

CASE REPORT 1

In October 1983 a 70-year-old gentleman was seen in the Department of Oral & Maxillofacial Surgery at Greenwich District Hospital. He had been referred by his GDP for the provision of dentures as 'he could not afford to pay for them'. On presentation he complained of inability to eat and recent onset of hoarseness. He also reported a recent faint in the street for which he had been examined at a local hospital, but there were no further details available.

Clinical examination confirmed edentulous arches, two perforations of the hard palate (Fig. 1) and nasal speech. There was abnormal scarring of the posterior part of the mouth and the throat (Fig. 2). The patient was unsure of how long the defects had been present. He confirmed that he had been married to his wife since the age of 29 years and they had one son. On further questioning he denied any extramarital sexual exposure. The chances of success with surgical repair were deemed poor and so dentures incorporating an obturator were constructed. Initial investigations suggested positive serology for syphilis

and the patient was referred to the Genito-Urinary Clinic for further investigation and treatment.

Serology results oddly confirmed an acute infectious syphilis as opposed to the suspected tertiary syphilis. This was explained as a fresh infection superimposed on the existing late syphilis. The patient's wife was invited to be tested. When she attended and discovered the investigations were for a venereal disease she promptly left the hospital never to be seen again despite repeated attempts to contact her. Cardiovascular complications were also diagnosed consistent with syphilitic aortitis and thought to be related to his recent faint.

Management comprised a 10-day course of procaine penicillin under steroid cover and a referral to the cardiology department. Regular blood and serological testing diagnosed re-infection in late 1984 which was again treated with antibiotics. His last documented clinic attendance was in 1985. Mortuary records for the hospital confirm that the patient died in 1988 from a ruptured aortic aneurysm.

CASE REPORT 2

In February 1999, a 36 year old man presented to Accident and Emergency at Bromley Hospital. His reason for attendance was the presence of a hole in his hard palate which had been slowly enlarging since its development two weeks previously. He was referred to the Oral & Maxillofacial Surgery Department at Orpington Hospital where he was seen for an

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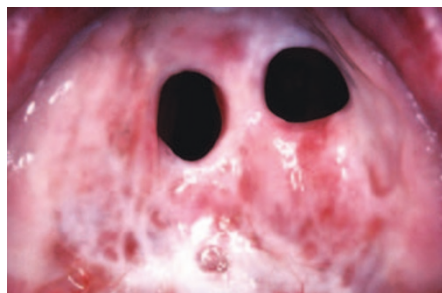


Fig. 1 Perforation of the hard palate caused by tertiary syphilis

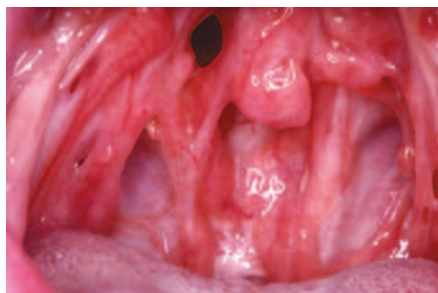


Fig. 2 Abnormal scarring of throat and posterior oral cavity



Fig. 3 Hard palate perforation caused by cocaine abuse

Table 1 Differential diagnosis of midline perforations ^{4,9,11,13-15,17,18}	
GROUP	EXAMPLES
Trauma	
Infection	TB, tertiary syphilis, typhoid, diphtheria, mucormycosis, actinomycosis
Neoplasia	Lymphoma, carcinoma, melanoma
Collagen vascular disease	Wegener's granulomatosis, SLE
Idiopathic	Midline lethal granuloma
Granulomatous disease	Sarcoidosis
Other	Cocaine abuse

initial consultation in March 1999. On presentation his chief complaint was of difficulty eating due to fluid and food escaping through the nose via the palate. More recently he had also noticed a change in his speech. He reported the hole had developed spontaneously and started as a pinhole which had gradually enlarged over the past four weeks. In addition he complained of three carious teeth which had been planned for extraction.

Further questioning confirmed a history of nasal cocaine abuse which the patient openly admitted to and cited as the cause of his palatal defect. He also admitted to smoking 30 cigarettes per day but did not drink alcohol. His relevant medical history included non-insulin dependent diabetes mellitus and depression for which he had been prescribed metformin, glibenclamide, lithium and venlafaxine.

Examination confirmed the presence of a small perforation, measuring 2-3 mm, in the centre of the anterior hard palate (Fig. 3). An immediate acrylic partial obturator was constructed and fitted which was successful in sealing the defect. The patient was advised that at this stage it was unwise to attempt any surgical repair, as this was most likely to fail, but that it could become a viable option once the cocaine abuse ceased. He was informed of the risk of further enlargement of the defect and regular review visits were recommended. However he failed to return

for any of these appointments.

One year later the patient was again referred to the same maxillofacial department having requested his GMP to do so. He claimed to have stopped using the cocaine and was eager for the defect to be surgically closed. Yet on examination it was seen to have increased in size and again surgery was deemed unwise. He was next seen in 2002 following a PDS referral for tooth extractions under GA. Again he insisted he had stopped using cocaine and had been clean for eight months. However he failed to return to collect a new obturator and to date the patient has not attended any follow-up visits.

DISCUSSION

Mechanism of damage

Syphilis is caused by the spirochaete *Treponema Pallidum* and may be congenital or acquired.⁴ Transmission can occur via direct contact with syphilitic lesions or infected blood. Entry is gained through epithelial breaks and spread occurs via the lymphatic and circulatory systems.² The incubation period lasts from 10 to 90 days after which the first two stages are highly contagious.⁸ Tertiary syphilis develops 3-10 years later in 30-40% of patients.⁴ It results from reactivation in an untreated or inadequately treated person or from reinfection in somebody previously treated.⁹ All features arise from endarteritis obliterans.¹⁰

Cocaine is an alkaloid extract derived from the *Erythroxylon coca* plant which

grows primarily in South America.¹¹ It is classed as a CNS stimulant and affects adrenergic nerve endings where it blocks the re-uptake of catecholamines and potentiates, in particular, dopamine resulting in transient euphoric effects.¹² Abuse of the drug dates as far back as the Inca era where coca chewing was part of the culture.¹² Nowadays the most common route of administration is snorting⁷ while smoking has increased in popularity with the advent of 'crack' cocaine. It is also abused intravenously.¹³ Its unique vasoconstrictive properties found use as an anaesthetic¹⁴ but if overused the intense vasoconstriction leads to ischaemia of affected tissues which can progress to necrosis and then perforation.¹⁵ The nasal septum is eroded and eventually disintegrates with collapse of the internal support structure.¹⁶ The area of damage extends to cause perforation of the hard and/or soft palate.¹⁷ Ischaemic necrosis of the soft palate musculature can lead to ulceration, infection and retraction with subsequent distortion and paralysis.¹⁶ The mechanism of necrosis is, however, probably multifactorial. It may be partly caused by the caustic effects of adulterants, local superinfection and inadvertent direct trauma as a result of the local anaesthetic effect. Such effects can be potentiated by smoking or injecting the drug.¹²

Clinical presentation

Three stages of syphilis are usually described. Each is associated with characteristic oral lesions. Primary syphilis uncommonly manifests as an oral chancre. Secondary syphilis can cause greyish ulceration in 'snail track' or 'mucous patch' formations.¹⁰ Tertiary syphilis is a non-infective multi-organ stage characterised by a painless localised granuloma (gumma)¹ which classically presents on the midline of the palate.⁹ Degradation of the mass leaves a deep pale ulcer with necrotic rolled margins.¹⁰ Chronic necrosis destroys the palatal bone to leave a clean perforation.⁸ Healing occurs with severe scarring which causes distortion or destruction of the soft palate and tongue.⁸ Systemic complications most seriously affect the cardiovascular and nervous systems. Aortic aneurysms, as seen in this case, are rarely seen.⁸

The local presentation of cocaine abuse is dependent upon its route of administration. Snorting most commonly results in an isolated septal perforation^{12,15} but can also lead to perforation of the hard palate.¹⁴ Patients complain of unintelligible nasal speech, nasal regurgitation and dysphagia possibly with resultant weight loss.¹⁶ Oral cocaine use is often associated with no more than a glossitis.⁵ Direct application of cocaine to the gingivae is used as a test for

substance purity and may cause gingival recession, ulceration, and necrosis.¹¹ Effects on hard tissue can present as erosion of enamel/dentine or corrosion of metal restorations.¹¹ Oral complications may also arise indirectly through related behavioural changes. Parafunction is one such example. Aggressive toothbrushing methods can result in cervical lesions or gingival trauma and neglect has been associated with higher rates of caries and periodontal disease.¹¹

Differential diagnosis

Diagnosis requires a detailed social or STD history. Unfortunately patients do not easily volunteer information concerning syphilis or cocaine abuse. While syphilis may be tested for, the diagnosis of cocaine abuse relies heavily on the patient admitting they have a habit. A urine drug screen could check for cocaine metabolites but this requires the informed consent of the patient. It is therefore necessary to eliminate other possible causes of midline destructive lesions. (Table 1)^{4,9,11,13-15,17,18}

Tests

The gumma is a granulomatous lesion characterised by endarteritis obliterans and the absence of scar tissue contracture. Histological changes can be non-specific and do not therefore form the basis of diagnosis.¹⁰ Non-specific serological testing (eg VDRL) indicates active disease and is positive in untreated tertiary syphilis. Specific tests (eg FTA) remain positive even in treated cases.⁸ Other tests may be appropriate at different stages. Testing normally extends to current and recent partners and also checks for other STDs.

Cocaine abuse is more difficult to diagnose as discussed above. Investigation therefore also involves elimination of other possible causes. Clinical examination may need to be followed by haematology/ biochemistry profiling, urine analysis, radiographs, nasal swabs, endoscopic evaluation, multiple biopsies and serological tests. However cocaine abusers may test positive for c-ANCA which is also positive in Wegener's granulomatosis and hence pose further difficulties in diagnosis.¹⁵

Management

Management is two-fold as it requires treatment of the underlying problem and local treatment of the perforation. Referral to a Genito-Urinary physician is necessary for treatment of syphilis. Penicillin forms the mainstay of treatment and is supported with preventive advice and regular sexual health check-ups.⁸ Treatment of late syphilis will not reverse the tissue damage but it may result in some improvement. Dental management can resume once the VDRL is negative.¹⁸

Management in cocaine cases is not an easy task due to the non-compliant lifestyle of the typical patient.¹² Many do not even seek treatment either because they are asymptomatic or because they have devised their own methods for managing their complications. Some may go so far as to attempt to seal their perforations with chewing gum or food. And, of course, there are those who remain in denial of their drug habit and will not come forward in order to avoid an admission to a health professional.¹² Those patients who do ask for help often compromise their management through failure to abstain from cocaine use and many are lost to follow-up. There is limited information available to explain the progression of disease and the influence of external factors.¹⁵ The published literature agrees that cessation of the drug habit plays a key role in reversing the pathological process and forms the basis for treatment.¹⁵ This is likely to require a referral for education and counselling as part of a drug rehabilitation programme.¹¹ Although there is not a firm consensus on further management most authors advise a period of conservative therapy. This incorporates analgesia, antibiotics, surgical debridement of necrotic tissue and nasal saline douches.^{12,15} Dental treatment carries risks with LA, GA and extractions.^{11,15} It is thus best avoided until medical clearance is given.

Specific management of the palatal defect may be to do nothing, to seal the defect or to repair the defect. Obturators are a successful method of managing the speech and masticatory problems. Surgery is another option but extensive scarring in syphilitic lesions makes any attempt at palatal repair hazardous. In cocaine abusers surgical intervention should be delayed for at least one year following confirmation of drug cessation and lesion stability.^{12,15} The patient should remain under review to check for progressive necrosis. Reconstruction is again controversial due to the aetiology of tissue damage. Ischaemia and necrosis render the tissues more likely to breakdown following surgical repair. The better option seems to be the non-surgical conservative approach.

When repair is indicated it is the site and size of the defect which dictates the most appropriate type of flap or graft.¹⁵ In most cases palatal defects and the floor of the nose are usually grafted using standard cleft palate surgery techniques.¹⁷ For the palate local flaps are the best option wherever possible.¹ A recent case, of congenital syphilis, reported successful repair of a large palatal defect using a tongue flap. The success was attributed to the excellent vascular supply and the proximity of the donor and recipient sites.¹ Speech therapy

was advised as part of the rehabilitation programme. Distal flaps may be an option particularly for large defects. Recent evidence suggests promising results are obtainable with the use of microsurgical forearm flaps.¹⁹

Acquired palatal perforations are produced by many conditions. The unusual causes are exemplified by the gumma of tertiary syphilis. Hopefully such cases will remain rare. However the incidence of defects caused by cocaine abuse is likely to increase and such patients will frequently present to their dental surgeon requesting treatment. Obturators remain a simple and effective way of managing their perforations.

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