Numb chin syndrome: a case series and discussion

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VERIFIABLE CPD PAPER

IN BRIEF

- Raises awareness of numb chin syndrome.
- Outlines aetiological factors for numb chin syndrome.
- Explains the association of numb chin syndrome with malignancy.

Numb chin syndrome is a sensory neuropathy in the distribution of the mental or inferior alveolar nerve. It may occur in benign disease, both systemic and dental in origin. It is also an under appreciated sign of malignancy. We present six cases from our experience highlighting the varied presentation and briefly review the aetiology, pathogenesis and management of numb chin syndrome and stress its importance with regards to the association with malignant disease.

INTRODUCTION

Numb chin syndrome (NCS), also referred to as mental neuropathy, is a sensory neuropathy characterised by altered sensation and numbness in the distribution of the mental nerve, a terminal branch of the mandibular division of the trigeminal nerve. Though there are a number of benign pathologies associated with it, it is an under appreciated manifestation of metastatic malignancy. The long course of the trigeminal nerve and its branches can lead it to be compromised at various levels, either intra-cranially or extra-cranially. It can occur as a consequence of compression of the mandibular division of the trigeminal nerve at the base of the skull by a tumour mass or by leptomeningeal invasion, or involvement of the mandibular division of the trigeminal nerve along its course. This syndrome is most often a forerunner to malignancy progression and relapse.

We present six cases that highlight the varied aetiology for NCS covering both benign and malignant causes. We will then discuss the various causes of NCS and its pathophysiology particularly in relation to malignancy.

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CASE 1

A 53-year-old female was referred with progressive numbness of the right side of her face which started three weeks previously. The numbness was initially around the right side of the upper lip and premolar region and slowly increased in size to involve the right side of the nose, around the eye and onto the skin of the lower face and jaw, including the lower lip. It was associated with a tingling and crushing sensation over the affected regions.

On examination, the patient had reduced sense of smell and taste and also numbness of the right half of the palate, right tongue and right lower lip. The patient had a heavily restored dentition with a healthy gingiva. Initial radiographs, including occipito-mental (OM) and dental panoramic tomograms (OPT) views demonstrated no abnormality in the facial skeleton. She was referred to the neurology department for further review. Blood tests and MRI scan were normal. The patient was subsequently given a diagnosis of idiopathic trigeminal neuropathy. The patient has been followed up and has been considered for trigeminal ganglion intervention.

CASE 2

A 66-year-old female, with a background history of hypertension, diabetes mellitus and stroke, presented complaining of intermittent pain of the lower left canine region of one week duration. On examination there was numbness of the affected area and the canine tooth was tender to touch.



Fig. 1 Coronal reformat of abdominal CT showing the renal cell carcinoma (arrow)

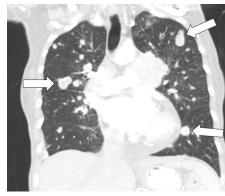


Fig. 2 Coronal reformat of chest CT showing multiple metastasis (arrows)

A periapical radiograph demonstrated no associated pathology of the canine tooth. On follow up a few days later, symptoms and clinical examination findings were similar and an OPT revealed no abnormality in the jaws.

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Fig. 3 Inflammatory lesion in the lower right canine/premolar gingival region



Fig. 4 Previously described inflammatory lesion has now healed

The patient was seen again two weeks later, now complaining that the pain was present over the left half of the face and that there was numbness in the region of the lower left half of the mandible. On this visit, she described the pain as stabbing in nature. Clinical examination demonstrated a reduction in light touch sensitivity over the distribution of the third division of the left trigeminal nerve, particularly in the region of the mental nerve. In the absence of abnormality in the face or jaws, the patient was soon referred to the neurology department, and subsequent investigation revealed that the patient had metastatic renal cell carcinoma (Figs 1-2). The patient died within six months of diagnosis.

CASE 3

A 55-year-old female presented with a four day history of an inflammatory lesion in the lower right canine/premolar gingival region, associated with intermittent pain and numbness (Fig. 3). On examination the numbness extended to the right infraorbital region, and right submandibular region. Intra-oral examination demonstrated a sloughy area of labial gingival in the region of the right lower right canine/premolar teeth with no apparent dental abnormality. Both periapical radiograph and OPT were normal. A biopsy of the affected gingival confirmed the lesion to be of inflammatory origin.

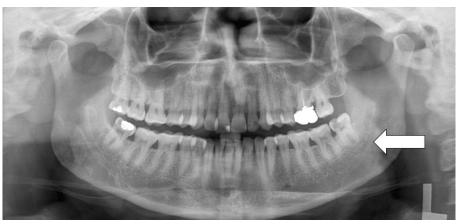


Fig. 5 Orthopantomogram demonstrating the periapical pathology in relation to lower left wisdom tooth (arrow)

Given the absence of abnormality detected in the radiographic examination of the face and jaws, the patient was referred to neurology. A subsequent MRI demonstrated an intracranial arteriovenous (AV) malformation which was presumed to be compressing the trigeminal nerve root. This vascular malformation was managed conservatively. On subsequent dental reviews, the gingival lesion had healed (Fig. 4), but the numbness with occasional tingling persists but with no further ulcerations.

CASE 4

A 40-year-old female was referred with a one week history of pain involving the left posterior mandible and a one day history of numbness in the left mental nerve. Extra-oral examination revealed frank numbness in the distribution of the left mental nerve and on intra-oral examination, there was dental caries in relation to the lower left wisdom tooth. An OPT demonstrated a periapical radiolucency in relation to the lower left wisdom tooth (Fig. 5). The patient was referred for surgical removal of the wisdom tooth. At subsequent review, the numbness had resolved. In this case the numbness was secondary to periapical infection of the lower left wisdom tooth.

CASE 5

A 47-year-old female was referred by her dentist with numbness affecting the left side of the lower lip which had started a few months previously. The patient had no relevant medical history.

Extra-oral examination determined there was a profound numbness of the left

side of the lower lip. Intra-oral examination showed fractured cusps of the lower left first and second molars, for which the patient was awaiting dental treatment. An OPT demonstrated an unerupted horizontally impacted lower left wisdom tooth associated with a 1 cm diameter cystic lesion (Fig. 6). This was in close association with the inferior alveolar nerve. The patient was referred for surgical removal of the wisdom tooth and associated cyst under general anaesthetic. At post-operative review the numbness left side of the lower lip had resolved. The histopathological examination confirmed the lesion to be a dentigerous cyst.

CASE 6

A 57-year-old non-smoking female patient presented with a progressive numbness of the right side of the chin and lower lip, which had started four months previously. On examination, a firm swelling was noted adjacent to the lower right second molar with pus discharging from this area. No tenderness was elicited on tooth percussion. The patient had a heavily restored dentition. In addition to hypertension, she had been treated for breast cancer three and a half years before, which involved radiotherapy and chemotherapy. Her current medication included anti-hypertensive drugs, thyroxin and intravenous bisphosphonates (three monthly infusions) which had been prescribed in relation to her cancer diagnosis.

An OPT demonstrated a patchy radiolucency centred over the apices of the lower right second premolar and second molar teeth (Fig. 7). Her lower right second premolar and second molar were removed,



Fig. 6 Orthopantomogram demonstrates an unerupted horizontally impacted lower left wisdom tooth associated with a 1 cm dentigerous cyst (arrow)

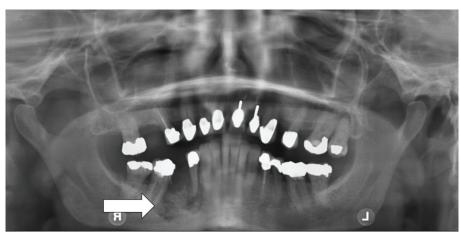


Fig. 7 Orthopantomogram demonstrates a patchy radiolucency centred over the apices of lower right second premolar and second molar teeth (arrow)

along with some associated bone, and the soft tissue around the area was biopsied and antibiotics given for the infection. Histological examination was reported as necrotic bone with resorption. At review, the swelling had resolved, but there remained a persistent discharging sinus. The area of continuing infection was debrided and on later review the sinus had healed but the numbness in relation to the lower right distribution of the inferior dental nerve persisted. The patient was diagnosed with osteochemonecrosis with secondary bacterial osteomyelitis in the lower right side of the mandible with the ongoing inflammation causing inferior alveolar nerve compression. Her numbness persists.

DISCUSSION

Charles Bell provided the first description of numb chin syndrome in 1830.¹ It was, however, not until 1963 when Calverley *et al.* used the phrase 'syndrome of the numb

chin' while describing five of their cases.² Since then there have been many reports of this syndrome. It is also referred to as mental neuropathy or numb lip syndrome.^{3,4}

Numb chin syndrome is a sensory neuropathy in the distribution of the mental or inferior alveolar nerve.³ It is characterised by numbness of the skin in the region of the lower lip, chin and mucous membrane inside the lip and extending to the midline.⁵ Patients may sustain injury leading to ulcers secondary to the altered sensation.⁵ It can be associated with pain and swelling when the primary pathology is destructive.⁶

The particular importance of this syndrome is its association with malignant disease. Breast, thyroid, renal, lung and prostate cancers are the most common malignancies that metastasise to mandible,³ but tumours of any cell origin can spread to the mandible.³ Other benign lesions can also cause NCS, as well as systemic conditions including amyloidosis, sickle cell

Table 1 Aetiology of NCS

A. Dental causes:

Odontogenic

- Dental abscesses
- Osteomyelitis
- Benign tumours

latrogenic causes:

- Facial trauma
- Wisdom teeth extraction (0.68-4.4%)
- Sensory disruption after ID block
- Orthognathic surgery of the mandible
- Implants
- Mandibular surgeries
- Orthodontic treatment (due to excess pressure)
- Faulty dentures (compressing mental foramen during mastication)
- Endodontic treatment (mechanical/chemical injury to ID nerve – less common)

B. Malignant causes:

- Breast cancer
- Haematological malignancies
- Lung cancer
- Renal tumours
- Melanoma
- Gastrointestinal cancer
- Multiple myeloma
- Head and neck cancer

C. Systemic causes:

- Diabetes mellitus
- Aneurysms
- Amyloidosis
- Sickle cell anaemia
- Vasculitis
- Sarcoidosis
- Syphillis
- Demyelinating disorders

anaemia, vasculitis, aneurysms, diabetes mellitus, drugs and age related mandibular atrophy (Table 1).^{4,6-9} Bisphosphonates are increasingly used in cancer therapy to protect the bone from destruction by metastatic tumour deposits, and from the effects of cancer therapeutics.¹⁰ There are reports in the literature where NCS has occurred secondary to demyelinating or inflammatory causes like multiple sclerosis and vasculitis.¹¹

In spite of its sinister connotation, dental causes are by far the commonest reasons for its occurrence.¹² Dental causes can include trauma to the mental nerve from ill fitting dentures, iatrogenic causes, dental abscesses, osteomyelitis, cysts and tumours of dental origin.^{4,6,12} Around 0.68-4.45% of lower third molar extractions result in numbness of the distribution of the mental nerve.¹³ A range of between 54 to 100% of patients who undergo orthognathic surgery experience paraesthesia of the inferior alveolar nerve and a numb lower lip and

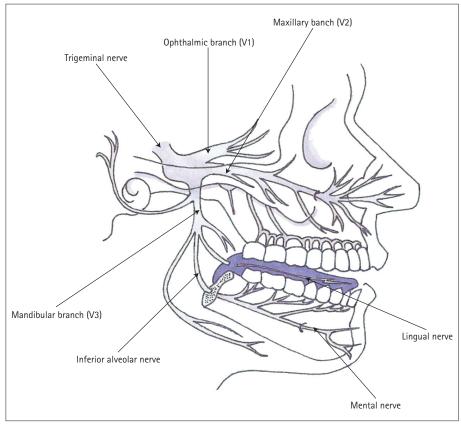


Fig. 8 Anatomy of the trigeminal nerve. Nerve can be involved intracranially, inferior alveolar nerve or mental nerve

chin. This is, however, typically temporary, though permanent damage can occur in some cases.¹⁴

Various explanations have been postulated regarding the pathogenesis of NCS. Direct infiltration or the compression of either the mental or inferior alveolar nerve occurs in tumours involving the plexiform course of the nerve in the mandible (Fig. 8).3,15 Mandibular nerve involvement can occur at the base of skull. Lesions here can cause osseous destruction and involvement of other cranial nerves.8,16 Leptomeningeal involvement can occur as a consequence of base of skull involvement or meningeal carcinomatosis.8,15 In some tumours including lymphoma, squamous cell carcinoma and malignant melanoma, perineural invasion has been described.8 In their paper Dalmau proposed that an immune reaction against still unknown antigens could precipitate mental neuropathy.17

Accurate localisation of the area of numbness can point to whether the nerve involvement is central or peripheral. Accurate history is equally important in determining the cause. A patient with NCS should be thoroughly investigated, including radiological investigations such as intra-oral periapical, OPT and radiographs of the facial skeleton, possibly supplemented with CT, MRI and bone scintigraphy where indicated. Other investigations may include blood tests and CSF analysis. In their paper Lossos et al. found adjunct investigation helped identify 89% of malignant cases. 16 It is important not only to diagnose the NCS but also to identify the aetiology. Maillefart et al. reported a six month median survival in those patients where the first sign of malignancy was NCS.13 Leptomeningeal involvement is associated with a better prognosis when compared to localised

mandibular involvement and have a 12 month median survival. 15,16

CONCLUSION

NCS may result from compression by benign lesions, by localised infection and inflammation, but is also an under-recognised sign of malignancy. It is important for clinicians to be aware of this seemingly trivial symptom. Appropriate investigations, referrals and follow-up should be undertaken to rule out a malignant aetiology. Its presence in a patient with known malignancy signifies poor prognosis.

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