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Case Report

A Rare Case of Hansen's Disease with Involvement of Buccal Branch of Facial Nerve with Lower Motor Neuron Palsy

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Leprosy is a usually disease of peripherally located nerves. In cranial nerves facial nerve followed by the trigeminal nerve being the most frequently affected. The involvement of the zygomatic branch of facial nerve has been reported previously but involvement of its buccal branch is rarely reported. Here, we report a 26 year old male, presented with insidious onset, gradually progressive, swelling and heaviness of the right cheek followed by sudden onset weakness of the right side of his face. Examination revealed an ill defined, hypoaesthetic macule over the right side of face associated with lower motor neuron type facial nerve palsy. The buccal branch of right facial nerve was thickened which was confirmed by ultrasonography and MRI of facial nerve. Histopathology, further confirmed the diagnosis of Hansen's disease (Borderline Tuberculoid). He was managed by Multi Drug Therapy (MDT) (Dapsone, Clofazimine and Rifampicin) and tapering dose of oral Prednisolone along with eye care and physiotherapy with satisfactory outcome.

Key words : Hansen's Disease, Facial Nerve, Buccal Nerve

Introduction

Leprosy is a disease of nerves which usually involves peripherally located nerves. In cranial nerves facial nerve is most frequently affected followed by the trigeminal nerve. The incidence of facial nerve involvement has been reported to be around 10% (Kumar et al 2006, Gopinath et al 2004). The involvement of the zygomatic branch of facial nerve has been reported previously but involvement of its buccal branch is rarely reported (Turkoff et al 2003). Here, we report a case of a Hansen's disease with involvement of buccal branch of facial nerve with lower motor neuron palsy for its rarity and uniqueness.

Case report

A 26 year old male, resident of West Bengal in India, presented with complaints of an insidious onset, gradually progressive, swelling and heaviness of the right cheek of 01 month duration. He reported to a dentist and was treated as a case of an impacted tooth. However, the swelling progressively increased over the next few days

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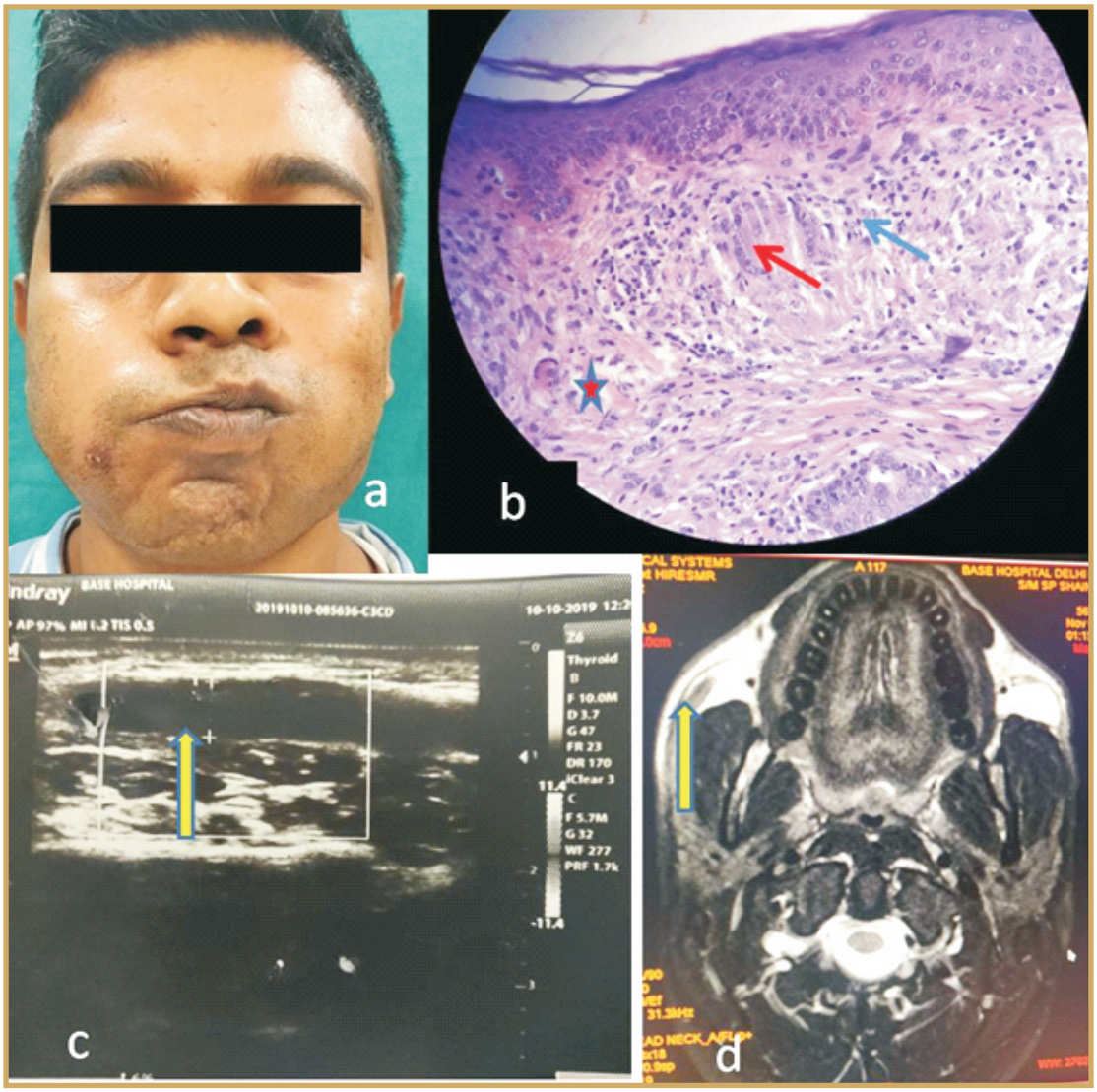

Fig. 1 : Results of radiological and histopathological investigations

1a. **Image of patient showing deviation of mouth towards left when asked to blowup.**

1b. **Histopathological examination (H&E stain 40X) showing well defined epithelioid cell granulomas (marked by red arrow) with central breakdown; focally and perivascular and peri-adnexal cell mononuclear cell infiltration (marked by blue arrow). Chronic inflammatory cells are infiltrating arrector pili muscle and there are few Langhans giant cells (marked by star) consistent with findings of Hansen's disease (borderline tuberculoid).**

1c. **Thickened buccal nerve in ultrasonography (yellow arrow indicating thickened buccal nerve between two plus marks).**

1d. **Thickened buccal nerve branch of facial nerve in right buccal space in a MRI image (indicated by yellow arrow).**

and there was no improvement in the heaviness. Over next 15 days, the patient developed sudden onset weakness of the right side of his face, which he noticed in the form of a change in appearance and difficulty in chewing food. He gave no history of trauma to face, fever, intercurrent infections, red, raised, painful lesions over body, shooting pain down the limbs or weakness of any other part of the body.

On examination, there was a solitary, ill defined, vaguely hypopigmented, mildly hypoaesthetic, hypotrichic, macule measuring approximately 1x1cm in size over the right side of angle of the mouth. There was no nerve to patch. Peripheral nerve examination revealed a non-tender and thickened right greater auricular nerve (WHO Grade 2) on same side as palsy. The greater auricular nerve on the other side was not visible. Rest of the peripheral nerves were also not thickened or palpable. The cranial nerve examination revealed a non-tender, thickened, buccal branch of right facial Nerve (WHO Grade 2) and facial nerve palsy with Lower motor neuron type (LMN) in the form of loss of forehead furrowing and naso-labial fold on the right side, Bell's phenomenon was present, there was deviation of the mouth to the left and he had difficulty in puffing out his cheeks (Fig 1a). Investigations revealed an essentially normal haemogram, renal function tests and liver function tests. A slit skin smear for lepra bacilli was negative however histopathology examination showed well defined epithelioid cell granulomas with dense perivascular and peri-adnexal infiltrate, suggestive of Hansen's disease (Borderline Tuberculoid) (Fig 1b). Nerve conduction study showed right facial nerve involvement. Ultrasonography revealed a thickened right facial nerve (Fig 1c) while MRI confirmed enhancing thickened nerve in right buccal space as buccal branch of facial nerve (Fig 1d). Based upon the history, examinations and investigation findings, our patient was diagnosed as a case of Hansen's disease (Borderline Tuberculoid) with LMN type of facial nerve palsy. He was started on Multi Drug Therapy (MDT) (Dapsone, Clofazimine and Rifampicin) and tapering dose of oral Prednisolone at 1 mg/kg along with eye care and physiotherapy. He has completed six months of MDT and steroid has been tapered over 5 months. The facial weakness has improved markedly though mild deviation of mouth remains. Hypoaesthesia over the patch has also improved significantly. He continues to be on three drugs MDT.

Discussion

Hansen's disease can even involve cranial nerves common being the facial nerve followed by trigeminal nerves. The zygomatic branch of the facial nerve is most affected because of its superficial location (Brand & Fytche 1985, Dastur et al 1966). However, it has been found that leprosy affects facial nerve in a scattered distribution from the main trunk to all peripheral branches and is not confined to peripheral zygomatic branch, as was thought earlier and involvement of the buccal branch has been rarely reported (Turkoff et al 2003). The involvement is more common in long standing cases of lepromatous leprosy, but can occur in other forms of leprosy with shorter disease duration also as was seen in this case.

Role of steroid in facial nerve palsy is still controversial. Study by Sullivan et al (2007) suggests early use of steroid has beneficial effects. However, recent Cochrane reviews suggest there is no benefit of steroids alone or in combination in idiopathic facial nerve palsy, Bell's palsy (Salinas et al 2004). However, administration of steroid is recommended modality in the patients of type 1 lepra reaction (Rao et al 2006, Smith et al 2004, WHO resource

accessed 2020). Our patient falls into category of facial nerve palsy due to infiltration of lepra bacilli/ its components as AFB were not demonstrable in histopathological sections in our case. He had shown significant improvement in lower motor neuron palsy subsequent to steroid administration which can also be partly explained by inflammation due to immune response in BT type of disease. Contribution of three drug combination having Clofazimine in it should also be taken into consideration. It is important to note that high index of suspicion of Hansen's disease should always be kept in differential diagnosis of facial nerve palsy particularly in endemic country like India (Kumar 2005). This will influence the choice of therapy including steroids as done in this case even when history was of 15 days and ultimately its outcomes.

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