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Case Report

Isolated Perioral/Perinasal (Onion Peel Skin) Sparing Pattern of Facial Sensory Loss: A Clinical Clue to Pars Caudalis Involvement in Posteroinferior Medullary Infarction

Sandhya Manorenj[1*], Zeyba Haider[1]

[1]Department of Neurology, Kerala Institute of Medical Sciences (KIMSHEALTH) Trivandrum, Kerala, India.

***Corresponding Author:** Dr. Sandhya Manorenj, Department of Neurology, Kerala Institute of Medical Sciences (KIMSHEALTH) Trivandrum, Kerala, India.

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Abstract

Background: Sensory disturbance of the face can occur either due to involvement of the trigeminal nerve or the trigeminal nucleus located in the brainstem and its connections. The chief sensory nucleus, mesencephalic nucleus, and spinal tract of the trigeminal nucleus in the brainstem mediate multiple sensory modalities of the face. Isolated involvement of the spinal tract of the trigeminal nucleus is very rare; hence it's often mislabelled as peripheral trigeminal neuropathy.

Objective: Here we describe a unique case of left facial sensory loss with clinicoradiological correlation.

Case Presentation: A 59-year-old woman with a prior history of hypertension presented with a one-month history of burning sensation in the left half of the face that worsened over 2 weeks. She did not have any motor complaints. Examination showed perioral and perinasal sparing of sensory loss of pain and temperature modalities, with preserved proprioception. Brain imaging showed a left posterior inferior medullary infarct.

Conclusion: An onion-peel skin pattern clue to involvement of the spinal tract of the trigeminal nucleus. Small medullary infarction may present with an isolated concentric

Keywords: *Face, sensory loss, medulla, onion skin*

Key Message

- Isolated facial sensory loss may be the sole manifestation of a lateral medullary infarction.
- Look for the characteristic onion-peel pattern of facial sensory loss to identify involvement of the spinal tract of trigeminal nucleus.
- Careful charting of facial sensory deficits can provide valuable clues to the anatomical localization of the lesion.

Introduction

Facial somatic sensation is primary mediated by trigeminal nerve. The facial sensory loss can occur due to involvement of parietal cortex, thalamus (ventral posteromedial nucleus), mesencephalic nucleus (midbrain), chief sensory nucleus in mid-pons, spinal trigeminal nucleus extending from caudal pons to C3 cervical spinal cord, and trigeminal nerve. Sensory loss to all modalities will be affected in parietal cortex lesion and thalamic lesion. Trigeminal nerve (V) involvement has peripheral nerve distribution pattern either ophthalmic, maxillary or mandibular nerve distribution pattern. [1, 2] Intrinsic brainstem lesion usually has segmental distribution pattern [1, 2] because of somatotopic representation in each nucleus and most of the time are associated with other long tract signs. Isolated sensory loss of face is very rarely reported due to intrinsic trigeminal nucleus involvement. They represent less than 1-2% of brainstem structural or vascular event. [3]

The authors present a prospective case study of a rare clinical entity, emphasizing the clinical and radiological correlation and illustrating the neuroanatomical substrates underlying the observed clinical pattern. Authors have obtained informed consent from patient/relatives for publication of the case.

Case Presentation

A 59-year-old lady with history of hypertension presented with one month history of burning sensation over left side of face especially forehead and upper ear region. On examination BP was 136/84 mmHg, pulse rate was 76/minute regular, pain, temperature and crude touch were impaired over left side of face with sparing of perioral and perinasal region, highest sensory loss was there on forehead and cheek region. Medial and lateral pterygoid movements, masseter, temporalis motor power were normal. Corneal reflex was sluggish bilaterally. Sensory examination showed normal sensation right half and reduced sensation by 50% for pain, temperature, crude touch and vibration sensation in left half [Figure 1]. There was no facial weakness, limb motor deficit, cerebellar signs or gait abnormality. Tandem walking was normal.

MRI brain showed acute to early subacute lacunar infarct in the posteroinferior aspect of left medulla, with normal angiogram brain [Figure 2]. Stroke work up showed denova detected diabetic state, glycosylated hemoglobin was 7.6 %; dyslipidaemia [LDL 224mg/dl, TC 293mg/dl]; 2D echo and Holter was normal. Since she came after a month, she was managed with single antiplatelets, high dose statins, diabetic medication, gabapentin, and antihypertensive and lifestyle modification. A diagnosis of lacunar stroke of medulla with onion peel skin pattern of facial sensory loss due to involvement of pars caudalis of spinal tract of trigeminal nucleus along with left upper cervical cord dorsal horn was considered secondary to involvement of small vessel branches of posterior inferior cerebellar artery or vertebral artery was considered.

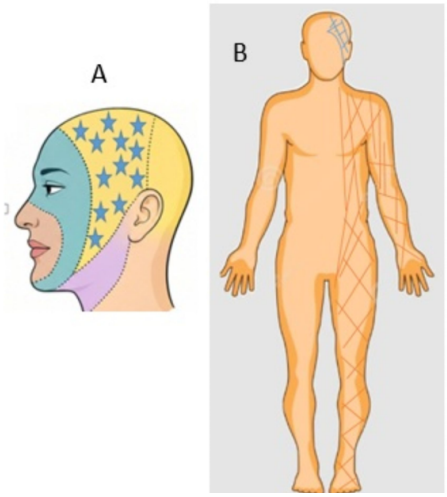


Figure 1. Schematic representation of ipsilateral sensory loss involving forehead, part of cheek (A) sparing perioral and perinasal region and involvement of ipsilateral half of body (B).

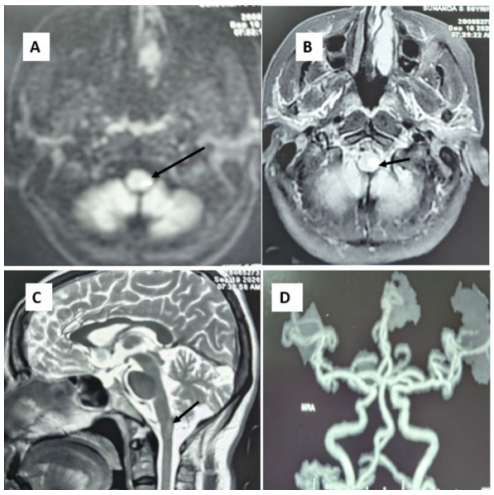


Figure 2. Magnetic resonance imaging brain diffusion weighted images (A) showed faint diffusion restriction in left posteroinferior medulla (marked by arrow), that is appearing hyper intense in T2 flair images axial section(B), T2 sagittal section shows extension of lesion (C) in lowermost medulla to upper cervical cord (hyperintensity marked by arrow) and angiography (D) is normal.

Discussion

The trigeminal nuclear complex consists of three major sensory nuclei. Principal sensory nucleus located in pons, mesencephalic nucleus in midbrain and spinal tract of trigeminal nucleus that extends caudally from pons. Proprioception of face is mediated by mesencephalic nucleus and principal sensory nucleus, while fine touch of face is mediated by principal sensory nucleus. [1, 2]

Spinal tract of trigeminal nucleus extends from the caudal pons through the medulla into the upper cervical cord (usually C2-C3). It carries pain, temperature sensation through the Aδ and C fibres and crude touch via larger low threshold mechanoreceptor afferents. [1,2,4] It has three subdivisions; pars oralis (rostral) that carries tactile sensation mainly from the oral/perioral region; pars interpolaris (located caudal pons and upper medulla) carries tactile and dental afferents; pars caudalis (in lower medulla) represent forehead, scalp, and outer cheek (peripheral facial region). Rostro caudal somatotopic representation of spinal tract of trigeminal nucleus cause an onion-skin pattern of sensory loss in brainstem lesions [Figure 3].

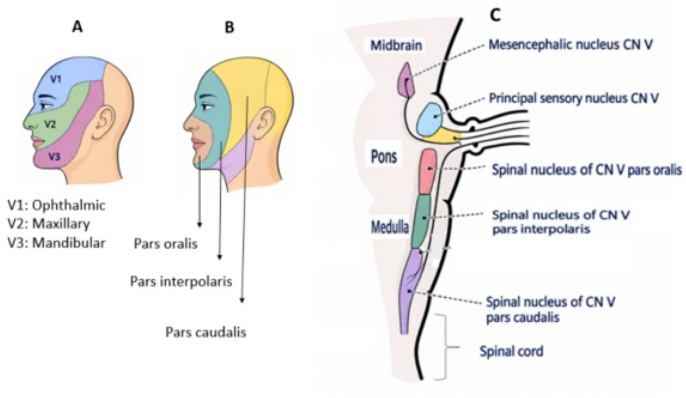


Figure 3. Shows patterns of trigeminal sensory loss, peripheral pattern or divisional pattern(A) involving branches of trigeminal nerve (V1, V2, V3) and segmental pattern (B) showing concentric rims of sensory pattern in intrinsic brainstem lesion and somatotopic representation (C) of spinal tract of trigeminal nucleus in brainstem.

The present case there is involvement of ipsilateral facial sensory loss along the outermost concentric ring of segmental distribution of trigeminal nerve and ipsilateral sensory symptoms in limbs. That shows perioral/ perinasal sparing suggesting pars caudalis involvement. Lateral medullary infarction associated with ipsilateral face and limb sensory symptoms is a rare entity but distinct variant caused by caudal lesion extending dorsomedially that probably involves ipsilateral pars caudalis trigeminal nucleus, ipsilateral dorsal column or decussating lemniscal fibres. [4] Present case lesion is extending from posteroinferior medulla to upper cervical cord. Pars caudalis extends into upper cervical cord ((C2-C3) that continues with the substantia gelotinosa of the spinal cord dorsal horn. Involvement of upper cervical cord dorsal horn in this case would explain partial loss of proprioception, pain, temperature in ipsilateral half of the body.

There are various patterns of sensory dysfunction described in lateral medullary infarction. [5] The classical ipsilateral trigeminal and contralateral limb pattern was observed in only 26% of patients. [5] Bilateral trigeminal pattern occurs when lesion are large ventrally extending at the middle rostral medulla. Isolated trigeminal and body involvement occurs with very small lesion as seen in our case.

Conclusion

Isolated facial sensory loss can be the only presentation of small infarcts in lateral medulla**.** Presence of onion peel skin pattern clues to intrinsic brainstem lesion

Conflict of Interest

The author declares no competing interests.

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