Multiple Lower Cranial Nerve Palsies Due to Nasopharyngeal Actinomycosis Masquerading as a Tumour – A Diagnostic Conundrum

Sir,

Lower cranial nerve palsy though common, is often challenging with an extensive etiologies. Usually, they occur as multiple instead of isolated nerve lesions. Based on the cause of the lesion, they may be genetic, infectious, malignant, traumatic, vascular, immunologic, metabolic, nutritional or degenerative.[1] They may be acute, subacute or recurrent in the presentation. Infections causing lower cranial nerve palsies constitute 10% in a study done by Kaene, and his colleagues.[2] Extra-cranial lesions of the head and neck are also a common cause for lower cranial nerve palsies.[1] Tumors (benign or malignant) like nasopharyngeal carcinoma presenting as lower cranial palsies is a well-known entity. Other differentials include metastases, pituitary malignancies, lymphomas, plasmacytosis, inflammatory pseudotumour, rhabdomyoma, chordomas, and chondrosarcomas. In our case, a nasopharyngeal mass was mimicking as a carcinoma radiologically. Biopsy has confirmed to be a treatable infection caused by Actinomyces, which is a rarity, enlightened in this case report.

Here we discuss a 62-year-old elderly male presented with complaints of right ear pain and vertigo for ten days. Then he developed bulbar dysfunction (dysphagia to both solids and liquids with nasal regurgitation and nasal twang) for one week and global headache with multiple episodes of vomiting for three days. He was a reformed smoker and alcoholic, but he started on nicotine snuffing for past six months. He was neither diabetic nor any other known comorbid illness in the past. There was no history of any fever, loss of weight or appetite, trauma, recent travel to endemic areas or any exposure to tuberculosis contact in the past. Clinically, he was conscious alert, moderately built and nourished. He had no cervical or generalized lymphadenopathy. Oral cavity showed dark brown stained teeth with no caries, ulcers or pus points. The otoscopic examination was normal except for mild bulging of the right tympanic membrane with no discharge or perforation. Nasopharyngoscopy showed right fossa of rosenmuller's fullness with congestion and minimal mucopus discharge. Clinically, he had asymmetrical cranial nerve palsies. The left hypoglossal nerve and bilateral glossopharyngeal and vagal nerves were involved. He had a nasal twang. Right palatal arch movements were reduced with a deviation of the uvula to the left with absent gag reflex and deviation of the tongue to the left. No pharyngeal congestion or discharge is noticed. Rest of the neurological examination was normal, with no evidence of any meningeal signs. Blood investigations showed normal hemogram (Total white cells – 11.4 × 103/microliter, Red blood cells count – 4.5 million/microliter, haemoglobin – 13.3 g/dl). His erythrocyte sedimentation rate was elevated (48 mm in 1 hour). His glycosylated haemoglobin was 5.9%. His renal and liver parameters were normal. Chest Rontgen imaging was normal. Magnetic Resonance Imaging (MRI) of the brain showed a large ill-defined mass in the right side of the nasopharynx [Figure 1a]– involving torus tubarius, eustachian tube and obliterating the fossa of Rosenmuller on the anteromedial aspect. The mass was extending across the midline and involving the contralateral posterior pharyngeal wall and showed heterogeneous solid and ring enhancement with gadolinium contrast [Figure 1b]. The features in the images like – poorly differentiated mass crossing fat and tissue planes, a central area of hypo-intensity (probable central areas of necrosis) [Figure 1c] with intense enhancement on contrast, and clival bone erosion – favoured more of nasopharyngeal carcinoma with infective aetiology being as one of the differentials. Contrast-enhanced computed tomography of the neck showed peripherally enhancing hypodense area probably abscess with surrounding mildly enhancing inflammatory mass in the nasopharynx [Figure 2a, b] with clival bone erosion [Figure 2c] and no nodal enlargement. Imaging also showed soft-tissue

growth in bilateral otitis media [Figure 2d, e] with thickening of the sphenoid sinus. Mass caused the narrowing of the left internal carotid artery just below the base of the skull. He had under-went myringotomy with nasopharyngeal lesion biopsy on March 29th, 2019. Biopsy showed nasal mucosa lined by ciliated columnar epithelium exhibiting squamous metaplasia with mixed inflammation with eosinophilic predominance [Figure 3a]. Also, there were numerous actinomycotic colonies, surrounded by an mixed with eosinophils predominant inflammation with no evidence of malignancy [Figure 3b-d]. Gram staining and acid fast bacilli staining was not done as the picture was very classical for actinomycotic colonies. Though it can normally be seen as a commensal in the human oral cavity and rarely in the nasopharynx, presence of multiple colonies with mixed inflammation and classical histopathological pattern, favoured active infection. Immunohistochemistry showed no positive cytokeratin cells amidst the lymphoid follicles.

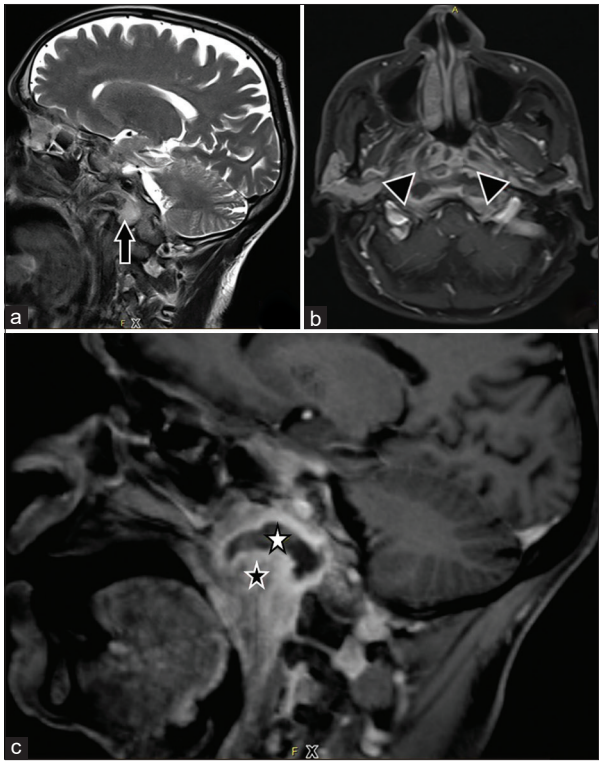

Figure 1: Magnetic resonance imaging – Sagittal (a, c) and axial sections (b) – showing hyperintense nasopharyngeal mass (arrow) with intensely enhancing (solid and ring – triangular heads) on gadolinium contrast and area of central hypo-intensity (white star) suggestive of necrosis

He was started on intravenous ceftriaxone two grams once daily for six weeks and then changed to an oral cephalosporin (Cefixime 200 milligrams twice daily) with doxycycline (100 milligrams twice daily) continued for a total of six months of treatment. He was under regular follow up – his vertigo and nasal twang recovered after one and two months respectively. He had persistent ear discharge initially and later subsided after six months of treatment. Dysphagia gradually resolved after two months course of treatment with minimal residual weakness. Repeat contrast-enhanced computed tomography of the neck showed the inflammatory mass with an abscess had been resolved entirely [Figure 2e, f,g].

Nasopharyngeal actinomycosis is usually a subacute to chronic infectious disease. Actinomyces, the causative organism, is a gram-positive, branching and filamentous bacterium.[3] It is often a commensal inhabiting oropharynx, gastrointestinal and urogenital tract.[4] Healthy people usually get infected after a mucosal trauma or inoculation into an anaerobic environment. Infection can also occur without any prior incident making the situation more difficult for diagnosis. Clinical variants

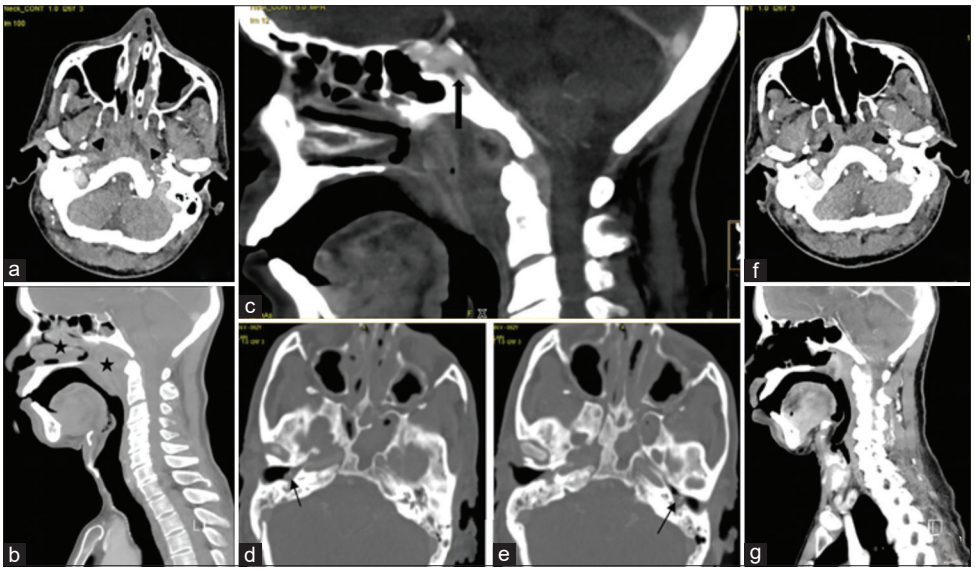

Figure 2: Computed tomography of neck – Axial (a, d, e, f) and sagittal (b, c, g) sections showed – a, b – arrow heads and star showing ill-defined Nasopharyngeal mass with mucosal thickening. c – showing soft tissue inflammatory mass with clival bone erosion. d, e – presence of soft tissue inflammation in bilateral otitis media. f, g – showing the mass and surrounding abscess have been resolved with clear sinuses

include cervicofacial, thoracic, abdominal and pelvic form.[5] The cervicofacial one is more common variant with characteristic fistula formation, while the opposite is noticed in nasopharyngeal form – occurring more commonly in young to middle-aged people with no fistula formation.

In our case, occurrence in an elderly male with an unusual site and its acute onset makes it a rarity. Here, nicotine sniffing was probably considered, as a provoking factor for causing mucosal trauma. There were reported cases, showing its occurrence without prior incident, leading to late diagnosis but had a great response to antibiotics without any death nor deformity. There was a decrease in the incidence of actinomycosis after the introduction of broad-range antibiotics and maintaining good oral hygiene.[3] In late presentation or delay in diagnosis, they can present as an invasive lesion with surrounding tissue destruction. These can mimic either as a malignant lesion or chronic infective lesions like tuberculosis or fungal infection. Actinomycosis typically spreads contiguously, ignoring tissue planes and aggressively invading adjacent tissues including bone. Unlike nasopharyngeal carcinomas, however, lymphatic dissemination of actinomycosis is highly unusual and hematogenous dissemination occurs at any stage of the infection. The evidence of lymphadenopathy and the nature of lymph nodes can be one of the distinguishing features from malignancy and chronic infections, where actinomycotic diseases usually lack lymphadenopathy which is seen in our case.[6] Similar cases have been documented elsewhere in 1997 and 2004, where a prior nasal trauma was reported. In these cases, the nasopharyngeal mass was mimicking as a nasopharyngeal carcinoma radiologically, but the biopsy was proven to be an infection.[7,8]

Infiltrative lower cranial palsies are often challenging in etiological evaluation for treating physicians. Benign and easily treatable conditions like Actinomyces shall always be considered as one among the differentials for large irregular and aggressive nasopharyngeal masses, that carry low mortality and better prognosis with treatment. Histopathological diagnosis of the lesion is foremost and paramount for definitive diagnosis.

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Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

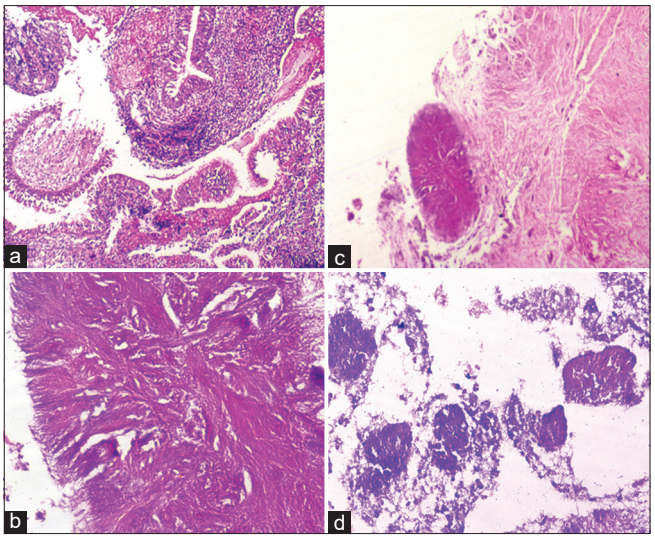

Figure 3: Histopathological picture of nasopharyngeal mass showed: a – ciliated columnar epithelium exhibiting squamous metaplasia with mixed inflammation and predominant eosinophilic inflammation. b (10X magnification) – Actinomycotic infection showing splendor hoepple phenomenon. c (30X magnification)– Actinomyotic colony. d (10X magnification) – Multiple actinomycotic colonies with inflammation

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