

Numb Chin Syndrome with Four Fulminant Attacks in 16 Years

Keywords: Numb chin syndrome; Mental neuropathy; Lip numbness; Long-term follow-up; Benign

Abstract

We present a case report of an otherwise healthy 68-year-old man with a 16-year follow-up of recurrent sudden and severe attacks of painful but benign numb chin syndrome (NCS).

The patient presented with only one subjective finding: recurrent mental nerve neuropathy, a periodic, localized, atypical facial pain continuing up to 2 weeks at a time. Undergoing tests on an outpatient basis, a complete lab work-up including dental examination and laboratory tests, electroencephalogram, chest x-ray, abdominal ultrasonography, head and neck magnetic resonance imaging and computed tomography were done.

All examinations and test results were within normal limits and with no remarkable findings in 16 years of follow-up. After family, social, surgical, medical, dental, and psychological histories, with appropriate and careful clinical work-ups, no definitive diagnoses, other than NCS, could be made. An extensive literature search in the PubMed interface using the key words, "numb chin syndrome," "mental neuropathy," and "mental nerve numbness," revealed 152 articles reporting 576 patients. As reported, NCS can be benign or a precursor of certain cancers, leukemia, other morbidity, and mortality. Though inconveniencing and embarrassing, painful and extremely worrying during the periods of infliction, the present case of NCS has been benign for 16 years.

Abbreviations

ALL: Acute Lymphoblastic Leukemia; CT: Computed Tomography; EEG: Electroencephalogram; MRI: Magnetic Resonance Imaging; NCS: Numb Chin Syndrome

Introduction

Numb chin syndrome (NCS) is also called, "mental nerve neuropathy," and is defined as a sensory neuropathy, characterized, in some cases, by pain and, in all cases, by numbness (hypoesthesia, paresthesia, dysesthesia, and anesthesia) of the unilateral or bilateral chin and lower lip within the mental or inferior alveolar nerve distribution. In 1932, Dandy first saw blood vessels on the trigeminal nerve in a patient presenting with hemifacial paralysis of the chin [1]. NCS could be a precursor of a systemic malignancy, neoplastic disease, or an as yet undiagnosed cancer [1-30]. The presenting symptom of numb chin, with or without comorbid lower lip anesthesia, is more often associated with cancer, either as the first symptom or manifesting during the outcome, than with benign diseases [31,32].

We present the first report documenting the benign, long-term, 16-year outcome of NCS. The aim of this study was to elucidate the ramifications of the possible prognoses of patients diagnosed with pernicious NCS in order to clarify some of the new science couching this neuropathy.

The patient's informed consent was obtained for all the procedures and the reporting thereof in this study.



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Case Report

The first NCS attack of this patient occurred at the age of 52 years old with only the presenting symptoms of dysesthesia and a thickening sensation of the right lower lip and chin, local fever, and slight pain. The patient also complained of insomnia, anxiety, and depression during the episode. Examination revealed local swelling, redness and a thickening sensation of the lower lip and chin, and lower anterior gingiva. The symptoms occurred synchronously in bouts of from 3 to 5 days for 2 weeks, after which, all the symptoms resolved spontaneously. However, to rule out a pernicious outcome, complete lab work-ups, including blood and dental laboratory tests, head and neck MRI and CT, electroencephalogram (EEG), chest x-ray, abdominal ultrasonography, were done. All the exam results came back within normal limits, without any remarkable findings. Possible dental problems, such as tooth extraction, trauma, abscess, or osteomyelitis, were all ruled out by thorough evaluation by a 20-year veteran, expert dentist.

He has been physically active and plays tennis at least once a week, never smoked cigarettes but enjoys a glass of wine with dinner. His annual physical health check-ups for the past 30 years have revealed normal results other than stage-2 hypertension which is controlled with medication.

After taking exhaustive family, social, medical, dental, and psychological histories, appropriate and careful clinical work-ups, no definitive diagnosis other than NCS could be made. The symptoms inexplicably resolved spontaneously.

Similar fulminant attacks with the same annoying, awkward, and worrying symptoms occurred at 4-, 3-, and 6-year intervals at the ages of 56, 59, and 65 years old, respectively. Subjective symptoms and findings on attacks were recurrent mental nerve neuropathy, a non-necrotizing symptom of a localized, dysesthesia, atypical facial pain in the right lower lip and chin, and general dullness in the jawbone. Again, at each episode, these symptoms caused insomnia, anxiety, and depression. Although complete lab work-ups, head and

neck MRI and CT, etc. were repeatedly done, all results were within normal limits and without any remarkable findings. As with the three previous episodes, all the symptoms resolved spontaneously without medical intervention.

The symptoms did not recur in 3 years of follow-up. At the final follow-up examination, at 66 years of age, which consisted of EEG, electromyography, an additional CT and MRI, which was 14 years after the first attack, all were again within normal limits. There was no disclosure of an underlying malignancy, neoplasm, or systemic disease. Although painful and an inconvenience during the periods of infliction, the present case has been benign for 16 years to date, at 68 years of age.

Discussion

In the last 55 years, 576 patients, reported in more than 100 journals worldwide, comprised our examination of the literature with the PubMed interface using the key words, “numb chin syndrome,” “mental neuropathy,” and “mental nerve numbness”. Our search revealed that of these 576 patients from 19 countries, 137 cases were benign, while alarmingly as many as 439 were malignancies including 11 deaths. Key data were extrapolated and compiled, from this sizable patient population, revealing broad generalizations.

Although relatively rare signs and symptoms, when they do present, they are known collectively as numb chin, and are most often a prodromal sign of a serious malady, as in the 439 cases (76%) in the present review. Numb chin turned out to be the prodromal sign and was the initial manifestation of acute lymphoblastic leukemia (ALL) of the B-cell type, Burkitt’s-type ALL, other types of leukemia, malignant lymphoma, breast cancer, prostate cancer, other types of cancers, metastatic, and systemic diseases, and even HIV/AIDS [3,7,12,14,17-23,25,28,30-47]. Henny C et al. cautioned, although benign in appearance, NCS is most often associated with visceral neoplasia or malignant hemopathy and a factor of poor prognosis [48]. Massey EW et al. pointed out a nontraumatic mental neuropathy should initiate a search for cancer [12]. Laurencet FM et al. reported survival after diagnosis is generally less than 1 year. The appearance of a mental nerve neuropathy should never be considered as a ‘banal’ symptom, and investigations to detect a possible cancer should be mandatory [30].

On the other hand, among the 576 cases reviewed plus the case presented in this study, there were 138 benign cases (24%). Baron JC observed that at variance with classical notions, the outlook of NCS is occasionally benign [49]. While those benign case reports described short-term states, among them there were no reports of any cases involving recurrent attacks or any long-term follow-up studies, as is the case with the patient in the present report. Fortunately, however, for this patient, the painful recurrences of numb chin will remain, for now at least, inexplicably diagnosed as benign. Though inconveniencing and embarrassing, painful and extremely worrying during the periods of infliction, the present case of NCS has been benign for 16 years.

Author Contributions

Mr. Robert E. Brandt was responsible for the study concept and design, analysis and interpretation of data, and critical revision of

the manuscript for important intellectual content and final approval. Dr. Akihiro Takeuchi, analysis and interpretation of data, and of the manuscript for important intellectual content. Dr. Toshikazu Hirayama, acquisition of data, final approval of the manuscript for important intellectual content.

Ethical Standards

The present work was validated by the Kitasato University Medical Ethics Organization, Kanagawa, Japan, registered as No. B17-284.

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