An inferior alveolar intraneural cyst: a case example and an anatomical explanation to support the articular theory within cranial nerves

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The authors describe the case of an intraneural ganglion cyst involving a cranial nerve (V3), which was found to have a joint connection in support of an articular origin within the cranial nerves. An inferior alveolar intraneural cyst was incidentally discovered on a plain radiograph prior to edentulation. It was resected from within the mandibular canal with no joint connection perceived at surgery. Histologically, the cyst was confirmed to be an intraneural ganglion cyst. Reinterpretation of the preoperative CT scan showed the cyst arising from the temporomandibular joint. This case is consistent with the articular (synovial) theory of intraneural ganglion cysts. An anatomical explanation and potential joint connection are provided for this case as well as several other cases of intraneural cysts in the literature, and thus unifying cranial nerve involvement with accepted concepts of intraneural ganglion cyst formation and propagation.


**KEY WORDS** intraneural ganglion; intraneural cyst; cranial nerve; trigeminal; hypoglossal; facial; peripheral nerve

**Case Report**

**History and Examination**

A 46-year-old woman presented to an oral surgeon for edentulation. Prior to surgery her general dentist obtained a radiograph, which showed a radiolucent cystic dilation near the right angle of the right mandible. The patient did not describe any pain, numbness, or other symptoms attributable to the lesion. Subsequent CT scanning of the facial bones with sagittal and coronal reformatting was performed (Fig. 1) and showed an intrasosseous cyst within the mandibular canal near the mandibular angle. The cyst was not connected to any dental structures. The cortical layers of the adjacent bone were markedly thinned, but there was no destruction or breakthrough.

**Treatment**

Five months later the patient underwent excisional biopsy at a local hospital. The cyst was approached intraorally using a sagittal split osteotomy. An approximately 2-cm cyst was dissected free from within the inferior alveolar nerve. No joint connection was noted. The inferior alveolar nerve remained intact.
Posttreatment Findings

Histological preparations were sent to the University of Minnesota Division of Oral Pathology for consult and to confirm the diagnosis. Subsequently, the case was re-reviewed at the Mayo Clinic (Fig. 2), and the diagnosis of intraneural ganglion cyst was favored.

Aware of the articular theory, we hypothesized that the cyst arose from a neighboring joint and propagated through the articular branch from the auriculotemporal nerve to the parent inferior alveolar nerve. The preoperative CT scan was re-reviewed. The available image data were manipulated using standard workstation tools (Volume Viewer, Advantage Windows 4.2, GE Healthcare) to create oblique maximum intensity projection (MIP) images with both bone and soft tissue windowing. A previously unrecognized connection to the temporomandibular joint (TMJ) was apparent (Fig. 1). Attempts to obtain postoperative MRI to look for persistent or recurrent cyst were unsuccessful given the patient’s claustrophobia.

Discussion

This case is unique in that it describes an intraneural ganglion cyst affecting a cranial nerve that was found to have a joint connection, in this case to the TMJ.

Synovial, or ganglion, cysts derived from synovial joints are well known. (Note that the terms “ganglia” and “synovial cysts” are used interchangeably in the literature.) The TMJ is a synovial joint with more than 50 cases of (extraneural) cysts reported in the literature. Extraneural cysts in this location can be asymptomatic or symptomatic (causing neural symptoms due to extrinsic nerve compression\(^{3,23,33}\) or mastication difficulties because of the mass\(^{2,7,20,32}\)). They are derived from the TMJ via non-neural pedicles and can extend in different directions, even intraosseously\(^{2,8,16,26}\) or intracranially\(^{23,33}\) (Fig. 3B). As a synovial joint, it is logical that this joint can also form an intraneural ganglion. According to the articular (synovial) theory, intraneural cysts are formed when joint

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FIG. 1. Preoperative CT scans. A: Axial nonenhanced CT scan of the facial bones showing a benign intraosseous cyst within the right mandible (asterisk). B: Coronal reformatted image (from same data set as in panel A) showing the intraosseous cyst (asterisk) with benign anterior and lateral erosion of the cortex (arrows). C: Sagittal reformatted image showing the cyst (asterisk) extending into an expanded mandibular foramen (arrow). D: Oblique sagittal maximum intensity projection (MIP) from the CT data set demonstrating proximal extension of the cyst from the mandible posteriorly and medially through the expanded foramen (black arrow) to the capsule of the TMJ (white arrows). The soft tissue windowing displays the cyst as an intermediate-density structure.

FIG. 2. Histology. A: Cystic lesion surrounded by loose connective tissue. H & E, original magnification ×40. B: A higher magnification features another area of the lumen and surrounding wall. H & E, original magnification ×100. C: The cyst lining was composed of perineurial cells highlighted by epithelial membrane antigen (EMA). EMA–avidin biotin complex with hematoxylin counterstain, original magnification ×20.
inferior alveolar ganglion cyst with an anatomical explanation

Fluid propagates from the neighboring synovial joint along the articular branch into the main parent nerve. The TMJ is innervated by the mandibular portion of the trigeminal nerve (V3), namely an articular branch of the auriculotemporal nerve. Accessory innervation is provided by a branch from the masseteric nerve or a branch penetrating the lateral pterygoid muscle (the latter, therefore, most probably arising from the lateral pterygoid nerve: Fig. 3A). In our case, we believe that the auriculotemporal nerve acted as a conduit for propagation of the cyst to the inferior alveolar nerve (Fig. 3C).

Our review of the literature revealed several reports that did not propose a mechanism of cyst formation but could also be explained by the articular theory. The only other example of an intraneural cyst affecting the trigeminal nerve also involved the inferior alveolar nerve and had similar clinicopathological findings. Cases of intraneural cysts of the facial nerve have also been reported. We wonder if these cases are examples of intraneural ganglion cysts, which can be anatomically explained by articular branches from the TMJ via the communicating auriculotemporal nerve to the parent facial nerve (Fig. 3D).

A separate group are so-called hypoglossal cysts, ganglia producing hypoglossal nerve compression. There are 3 reported intraneural cases. Seven extraneural cases have been documented. Extraneural cases have been documented from the atlantooccipital joint and 2 from the atlantoaxial joint. There is some controversy about whether the 3 mentioned “intraneural” cases are in fact intra- or extraneural and whether joint connections existed in these cases. We believe that all of the reported examples are extraneural and have joint connections. We have also introduced a possible anatomical explanation that could explain the occurrence of an intraneural ganglion.

We report this case and our reexamination of the available imaging as a proof of concept; however, we acknowl-
edge the limitations of our report. The only available imaging in the patient was a CT scan of the facial bones, which was acquired with 2-mm axial slices without contrast. Images were not overlapping so that the coronal and sagittal reformatted images are somewhat limited by artifact as well as by the fact that images rather than raw data were used to create the reformatted images. We were able to manipulate the available image data through the use of a volumetric viewer to create MIP images at a variety of obliquities (including the image presented in Fig. 3D). Usable 3D reconstructions could not be performed because of the relatively low resolution and non-isotropic data set available to us. Ideally, dedicated MRI and CT scanning of the right TMJ would have been obtained with high spatial resolution and volumetric acquisitions, which would have allowed seamless 2D and 3D reconstructions and a high degree of detail. Contrast enhancement would have been useful to better delineate the intraneural cyst from adjacent muscle. Were it available in the patient, CT or MRI TMJ arthrography would have conclusively shown the cyst origin from that joint. Surgery was performed without exploration of the TMJ and without confirmation of a joint connection via the articular branch of the auriculotemporal nerve.

Conclusions

We described a mechanism by which a cranial nerve example of an intraneural ganglion cyst could be explained. We believe that this cyst arose from the TMJ, dissected within the articular branch (auriculotemporal nerve), and propagated within the inferior alveolar nerve, which would be supported by the articular theory for intraneural ganglia. We further suggest that our anatomical theory would similarly explain other rare examples of intraneural cysts within the trigeminal or facial nerve.

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References

7. Bonacci CE, Lambert BJ, Pulse CL, Israel HA: Inflammato-


Author Contributions
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