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## Case Report

# Intraosseous Schwannoma Originating in Inferior Alveolar Nerve: A Case Report

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## Abstract

Schwannomas (neurilemmomas) are benign neoplasms derived from Schwann cells of the neurilemma and appear most frequently on the auditory nerve or peripheral nerves of the skin. They arise in the oral and maxillofacial region infrequently, and very rarely in the center of the jaw. We herein present a case of a rare mandibular intraosseous schwannoma derived from the main trunk of the inferior alveolar nerve in a 33-year-old man. Fusiform expansion in the mandibular canal was observed and a mass showing the target sign in the mandibular canal was confirmed on T2-weighted and Gd contrast-enhanced T1-weighted MRI. Based on these findings, an inferior alveolar nerve-derived schwannoma or other benign nervous system neoplasm was diagnosed. A buccal side cortical bone flap in the mandibular molar region was removed to expose the mass, which was then peeled away from the nerve fibers and completely removed. Some inferior alveolar nerve fibers that were connected to the mass were removed at the same time, but the remaining nerve fiber bundle was preserved. Histopathology confirmed the diagnosis of a schwannoma with Antoni type A and Antoni type B regions. Although the patient experienced extremely mild paresthesia in the skin over the mental region and mental foramen at immediately after surgery, this had almost entirely disappeared at 7 years and 4 months later, and there has been no tumor recurrence.

**Key words:** Schwannoma—Neurilemmoma—Inferior alveolar nerve—  
Intraosseous—Mandible

## Introduction

Schwannomas (neurilemmomas) are benign neoplasms derived from Schwann cells of the

neurilemma and appear most frequently on the auditory nerve or peripheral nerves of the skin<sup>2)</sup>. In the head and neck area, they primarily appear in soft tissue and often in

the tongue, and intraosseous schwannomas are particularly rare<sup>11)</sup>, making up less than 1% of all benign primary bone tumors<sup>13)</sup>.

While it has been suggested that intraosseous schwannomas appearing in the mandible are linked to the inferior alveolar nerve, they are often slow-growing and progress without symptoms, and are discovered only after they have grown quite large. By that time, they are removed from the inferior alveolar nerve and encapsulated, making it impossible to determine whether they originated in the main trunk or a peripheral branch of the inferior alveolar nerve<sup>13)</sup>. As a result, resection of the neoplasm often means removing both the nerves connected to the lesion, and all the other adjacent nerves. We herein present a case in which an inferior alveolar nerve-derived schwannoma presenting with fusiform expansion of the mandibular canal was excised while preserving the nerve, leaving the perceptual function of the inferior alveolar nerve largely intact.

### Case

The patient was a 33-year-old man who presented with throbbing pain in the area around the left mandibular first molar. A diagnosis of pulpitis was made and a pulpectomy performed. The pain persisted, and a panoramic radiograph revealed expansion of the mandibular canal near the left mandibular molar area. The patient was then referred to our department. An initial examination of the oral cavity only revealed mild pain on percussion of the left mandibular first and second molars, but no spontaneous pain, local swelling, or anesthesia in the mental nerve area. No abnormal findings were observed in the regional lymph nodes.

Panoramic radiography and computed tomography (CT) showed vertical and lateral expansion in the wall of the mandibular canal and, a radiolucent fusiform area apparently unconnected with the teeth or root tips (Fig. 1). Magnetic resonance imaging (MRI) revealed a mass with low signal intensity on

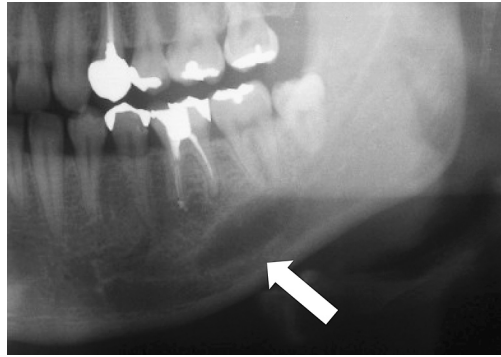


Fig. 1 Panoramic radiograph showed vertical and lateral expansion in wall of mandibular canal and radiolucent fusiform area with no connection with teeth or root tips (arrow)

T1-weighted imaging, but high signal intensity on T2-weighted and Gd contrast-enhanced T1-weighted imaging (Fig. 2). Horizontal MRI revealed a region with clear boundaries accompanying a target sign with partial non-uniform contrast inside. The coronal view revealed that the mass occupied a large portion of the mandibular canal. A benign intraosseous nervous system neoplasm derived from the inferior alveolar nerve was strongly suspected based on these findings.

A benign neoplasm was resected from the mandible *via* tracheal intubation under general anesthesia by intraoral technique. A sagittal split osteotomy was performed on the segment of the exterior cortical bone from the back of the mental foramen to the back of the mandibular second molar. The cortical bone flap was removed, revealing a lesion surrounded by a fibrous capsule in the expanded mandibular canal. Despite the anterior and posterior sides of the lesion being connected to the inferior alveolar neurovascular bundle, it was easily peeled away from the nerve fibers, making it possible to preserve the nerve fibers while stripping away only the mass. However, it was suspected that part of the posterior side of the lesion adhered to the nerve fibers in a section where there were unclear boundaries between them. In this section only, the nerve fibers were excised

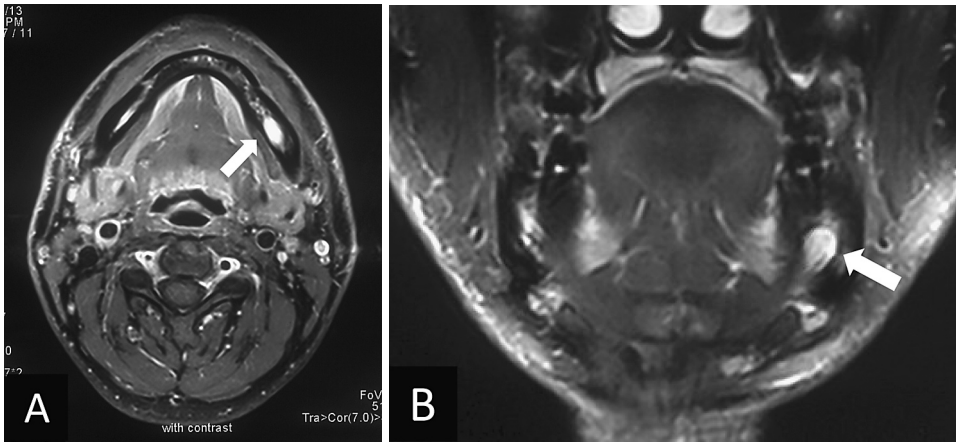


Fig. 2 A: MRI revealed mass with high signal intensity on Gd contrast-enhanced T1-weighted image  
 B: Characteristic target sign of schwannoma was observed on T2-weighted image (arrows)

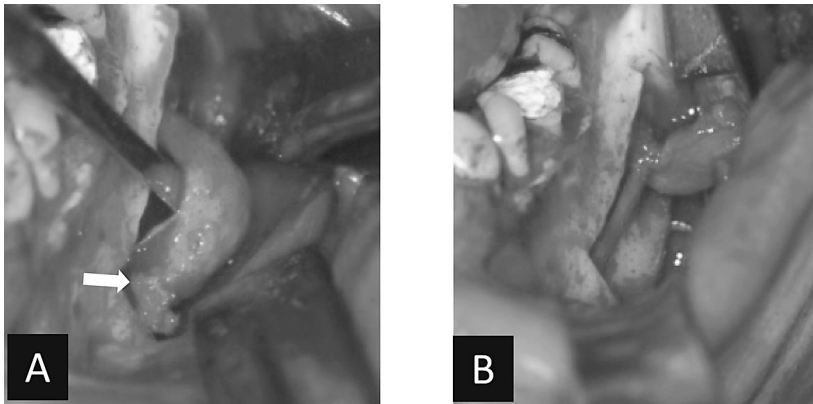


Fig. 3 A: Direct attachment of tumor to nerve was noted during removal (arrow)  
 B: Continuity of inferior alveolar nerve was preserved

together with the mass (Fig. 3). The provisionally removed cortical bone flap was put back and fixed in place with a bioabsorbable polylactate osteosynthesis miniplate and the wound was closed (Fig. 4).

An internally homogenous, soft, rubbery, yellow-whitish mass approximately 10×20 mm was extracted (Fig. 5). Histopathology by H-E staining revealed a proliferation of spindle-shaped and elliptical tumor cells in loose fibrous connective tissue with small-to-medium-sized vessels with ectasia and thrombosis. A portion of the tumor consisted of a



Fig. 4 Cortical bone flap was put back and fixed in place with bioabsorbable osteosynthesis miniplate

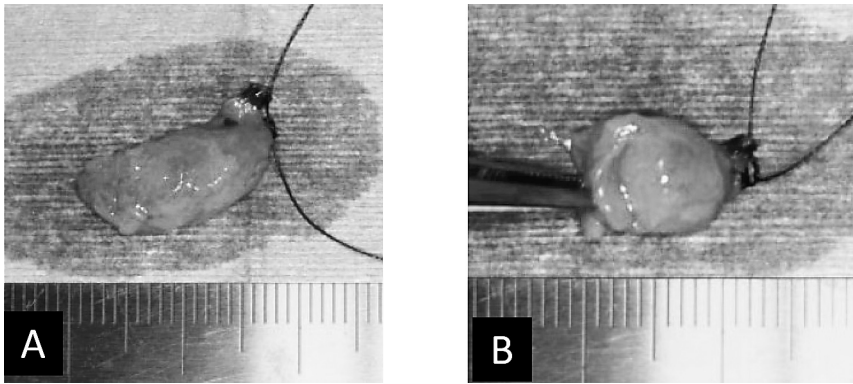


Fig. 5 A and B: Approximately 10×20 mm, soft, rubbery, yellow-whitish, solid mass that was internally homogenous was extracted

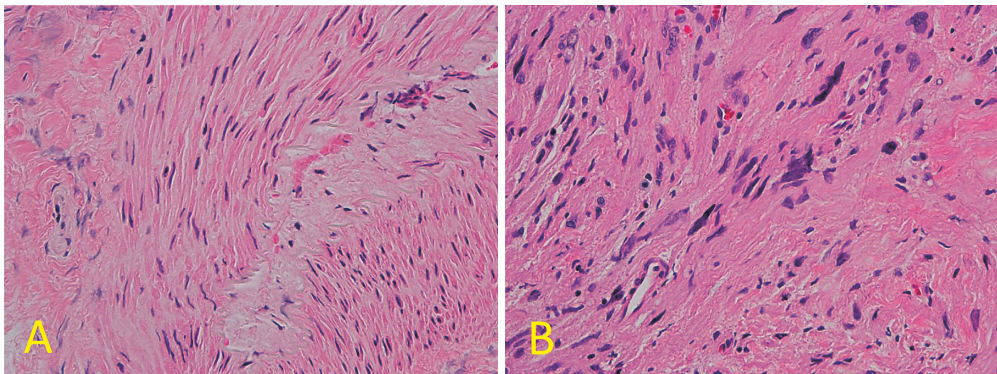


Fig. 6 A: Photomicrograph showing Antoni type A tissue consisting of dense proliferation of spindle-shaped cells with palisading arranged nuclei (hematoxylin-eosin, original magnification ×20)  
 B: Photomicrograph of lesion showing irregular, dense arrangement of cells and elongated spindle tumor cells. Antoni A was predominant microscopic pattern, alternating with Antoni B areas occasionally (hematoxylin-eosin, original magnification ×40)

dense proliferation of spindle-shaped cells with palisading arranged nuclei, a characteristic feature of Antoni type A schwannoma (Fig. 6-A). The greater portion of the tumor, however, consisted of spindle cells with large, hyperchromatic nuclei, which were haphazardly arranged within a delicate fibrillar microcystic matrix, and myxoid degeneration (so-called Antoni type B areas). Antoni A areas were the predominant microscopic pattern, alternating with Antoni B areas occasionally (Fig. 6-B). Immunohistochemical staining was positive for S-100 protein and weakly positive for NSE in the tumor cells, confirming the diagnosis of a nerve-derived tumor (Fig. 7).

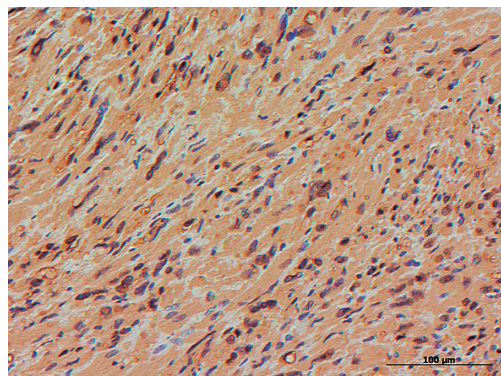


Fig. 7 Immunohistochemical findings  
 Staining was positive for S-100 protein (S-100, original magnification ×20)

Based on the above findings, a schwannoma with Antoni type A and B areas was diagnosed. Imaging findings at 7 years and 4 months after surgery showed new bone formation where the mass had been, the disappearance of mandibular canal expansion, and recovery of regular mandibular canal structure. Further MRI revealed that the signal value in the mandibular canal was the same as that of the healthy side. In tests to investigate impairment of the inferior alveolar nerve soon after surgery, the patient felt slight discomfort in the left mandibular incisor and extremely mild paresthesia in the skin over the mental region and mental foramen. Perceptual function tests with a Semmes-Weinstein pressure esthesiometer at 7 years after surgery showed recovery to nearly normal. There has been no recurrence of the tumor to date.

### Discussion

Schwannomas may appear anywhere in the body, but mostly originate in the auditory nerve or peripheral nerves of the skin. In the head and neck area, they primarily appear in soft tissue and often in the tongue, and intraosseous schwannomas are extremely rare<sup>11</sup>. Unni<sup>13</sup> reported their prevalence as making up less than 1% of all benign primary bone tumors. In a review of 152 cases of schwannoma occurring in the oral and maxillofacial region, Gallo<sup>6</sup> reported only 18 cases (11.8%) of intraosseous schwannoma in the mandible.

While it has been suggested that intraosseous schwannomas arising in the mandible are associated with the inferior alveolar nerve, Martins *et al.*<sup>10</sup> and de Lacerda *et al.*<sup>3</sup> have also reported them originating in the dental root and alveolar bone of the mandibular symphysis. Schwannomas are often slow-growing and develop without symptoms. As a result, they are often discovered only after they have grown quite large. In many cases, they are already disassociated with the inferior alveolar nerve and encapsulated upon discovery, and it cannot be determined whether they

originated in a peripheral nerve branch or in the main trunk of the inferior alveolar nerve<sup>13</sup>. According to Chi *et al.*<sup>1</sup>, there are three mechanisms by which schwannomas may involve bone: (1) a tumor may arise centrally within bone; (2) a tumor may arise within the nutrient canal and produce canal enlargement; or (3) a soft tissue or periosteal tumor may cause secondary erosion and penetration into bone. In the present case, fusiform expansion was observed in the mandibular canal, but no abnormalities in the surrounding mandibular bone or connection with the dental root. This corresponds to (2) as proposed by Chi, indicating that the tumor arose not in a peripheral branch, but in the main trunk of the inferior alveolar nerve.

Schwannomas exhibit two characteristic histopathological patterns: Antoni type A, in which the tissue consists of spindle-shaped cells with parallel rows of palisading nuclei organized in whorls and waves. These cells often surround an acellular eosinophilic zone (Verocay body), representing reduplicated basement membrane; and Antoni type B, consisting of spindle cells haphazardly scattered in a delicate fibrillar microcystic matrix. Most schwannomas contain a mixture of both Antoni type A and B tissue. The present schwannoma was also a mixture of these two types, with Antoni type A as the predominant microscopic pattern, alternating with Antoni type B areas occasionally.

Lesions that present with expansion of the mandibular canal may be hemangiomas, simple bone cysts, or malignant lymphomas, among other possibilities. It is necessary to carefully distinguish the schwannoma from these other diseases. Koga *et al.*<sup>8</sup> stated that the presence or absence of characteristic target signs on MRI may be very useful for imaging diagnosis of schwannomas. In tumors comprising a mixture of Antoni type A and B areas, the signal value will differ between sparse and closely packed areas, and the image will generally be non-uniform. Therefore, a schwannoma or other benign nervous system neoplasm must be suspected in cases where diagnostic imaging reveals fusiform

expansion of the mandibular canal, a solid lesion affected by contrast enhancement, clear boundaries, and no apparent destruction of the bone in the mandibular canal wall. Moreover, a schwannoma should particularly be suspected if target signs are confirmed by MRI. In the present case, imaging findings also indicated target signs in one section, leading us to suspect schwannoma. We did not perform a biopsy because it would have been highly invasive due to the small size of the lesion and its central location in the jaw bone, and schwannoma was most strongly suspected from the imaging findings. Malignant schwannoma very rarely appear in the oral region<sup>7,12)</sup>. Ducatman *et al.*<sup>4)</sup> reported a 4.6% incidence of malignant schwannoma concurrent with von Recklinghausen's disease and only a very small 0.001% incidence among patients without the disease. We therefore made a clinical diagnosis of a benign tumor and planned a tumor excision with an approach that made extended surgery possible should the lesion have turned out to be a malignant tumor.

The treatment of choice for schwannoma is surgery, as radiation therapy and chemotherapy are ineffective. There are various arguments as to whether nerves connected to the tumor should be preserved. While there are comparatively more reports of radical resection including excision of the tumor together with the nerves connected to it, paralysis of the inferior alveolar nerve when it is excised together with the tumor imposes a large burden on the patient. While there have been rare cases of recurrence of schwannoma in patients undergoing incomplete tumor extraction, malignant transformation of the tumor is exceedingly rare. Furthermore, there have been no reports of malignant transformation of intraosseous schwannomas to date<sup>5,9,14)</sup>. It is therefore preferable to preserve the inferior alveolar nerve if it is possible to peel the lesion away from the nerve fiber during surgery. In the present case, we excised a portion of the inferior alveolar nerve that was attached to the tumor, but were able to peel the rest of the tumor away from the

nerve fiber, thereby excising only the tumor while leaving most of the inferior alveolar nerve bundle intact. The patient experienced extremely mild nerve paralysis in the postoperative period, and perceptual function tests at 7 years after surgery showed recovery to nearly normal levels.

The results in this case indicate that target signs on MRI are a characteristic feature of and helpful in the diagnosis of schwannoma, and that it is preferable to preserve the inferior alveolar nerve if it is possible to peel the lesion away from the nerve fiber.

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