

SOLITARY INTRAOSSEOUS NEUROFIBROMA OF THE MANDIBLE: REPORT OF A CASE

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ABSTRACT

A case of solitary intraosseous neurofibroma of the mandible is reported. The tumor was located in the body of the right mandible. Segmental mandibulectomy including the surrounding tissue was performed and the mandible was reconstructed with the free scapular osteocutaneous flap. From histological examination of the resected mandible, typical findings of a tumor invading the adjacent bone along the mandibular nerve were observed. Immunohistochemical examination of the tumor showed positive staining of the tumor cells for anti-S100 protein antibodies. Numerous nerve-like fibers were also noted by Bodian stain.

Key Words: Neurofibroma, Mandible, Inferior alveolar nerve

INTRODUCTION

Neurofibroma of the oral lesion is common in association with neurofibromatosis (von Recklinghausen's disease), while solitary intraosseous neurofibroma is rare. Das Gupts *et al.*¹⁾ found that among 303 cases of benign nerve sheath tumors, about 45% involved the head and neck region and approximately 9% occurred in the oral cavity. Recently, Polak *et al.*²⁾ reported a case of solitary neurofibroma of the mandible. Their analysis of 66 cases of neurofibroma revealed the following distribution concerning the site of occurrence of neurofibroma in the head and neck area: tongue, 12; palate, 12; mandibular ridge/vestibule, 15; maxillary ridge/vestibule, 9; buccal mucosa, 10; lip, 4; mandibular intrabony, 2; gingiva, 1; and floor of mouth, 1. Moreover, in the past literature, they found only 29 cases of solitary neurofibroma of the mandible. The total number of cases of intraosseous neurofibroma was only 30 including the present case. It can be said that mandibular intrabony neurofibroma is extremely rare.

We report a case of a neurofibroma of the mandible that showed the typical histological findings of the tumor invading the surrounding tissue and discuss its origin.

REPORT OF A CASE

A 37-year-old man visited the dentist to receive usual dental treatment in May 1988. A panoramic radiograph was taken, and showed an oval radiolucent area in the body of the right mandible, underlying the premolar and first molar region (Fig. 1). The area was well circumscribed and well demarcated with a homogeneous background. The connection of the radiolucent area to the mandibular canal was observed on the radiograms. The teeth overlying the lesion showed

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Accepted for Publication in November 24, 1992

vital pulp, and there was no evidence of root resorption.

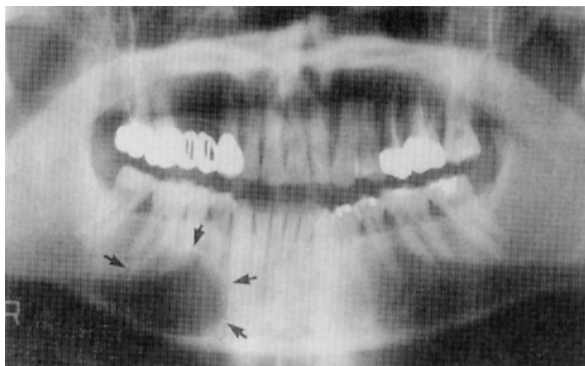


Fig. 1. Panoramic radiogram of the patient, showing a radiolucent area.

The patient did not complain of pain from the lesion. There was a remarkable swelling in the mandibular body. A CT scan of the mandible showed a round radiolucent image in the right mandibular body, and thin cortical bone around the tumor (Fig. 2). There was no lymphadenopathy. There were no café-au-lait spots or subcutaneous nodules on the patient's trunk, and there was no maxillary freckling.



Fig. 2. CT scan radiogram of the patient, showing an oval radiolucent lesion.

Incision biopsy of the oral lesion was performed and specimens were submitted for histopathologic examination. The microscopic appearance of the specimen showed ovoid or spindle-shaped cells intermingled with a delicate fibrillar stroma. Numerous mast cells were scattered throughout the stroma (Fig. 3). Immunohistochemical studies showed positive staining of the tumor cells for anti-S100 protein antibodies (Fig. 4). Also numerous nerve-like fibers were noted by Bodian stain (Fig. 5). Therefore, the final diagnosis was neurofibroma of the mandible.

Under general anesthesia, segmental resection of the right mandibular body was performed because the lesion was too big to preserve the mandibular continuity. The adjacent periosteum of the right mandible was also removed because of the potentiality of tumor invasion into the

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peripheral soft tissue. From the histopathological findings of the resected mandible, there was no tumor capsule, and deep invasion of the tumor cells into the surrounding bone along the mandibular nerve was observed. Also, the mandibular nerve was observed in the central part of the tumor (Fig. 6). A 7×5×3 cm defect that included the overlying mucosa remained after surgery. The free scapular osteocutaneous flap was transplanted by microvascular anastomosis to the facial artery and vein. The blood circulation was excellent after the operation and the flap survived completely. Three years postsurgery there was no evidence of recurrence or functional problems.

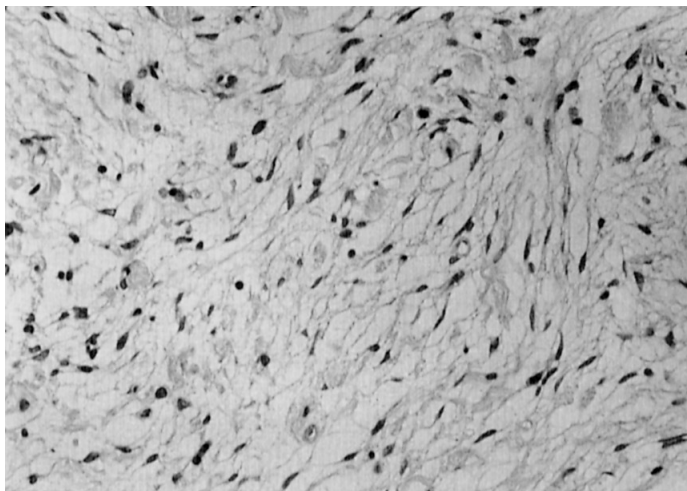


Fig. 3. Photomicrograph of a section from the tumor showing bundles of spindle-shaped cells within a delicate stroma. (Hematoxylin-eosin stain. Original magnification, ×400)

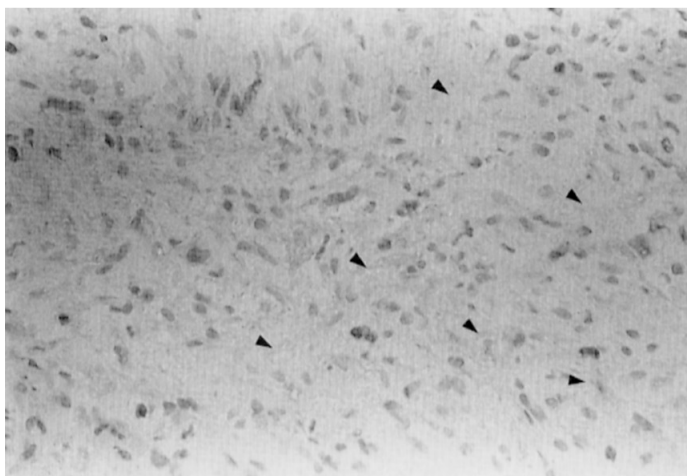


Fig. 4. Immunohistochemical staining of the tumor cells, showing positive staining for anti-S100 protein antibodies. (Allows showing S100 protein positive cells, ×400)

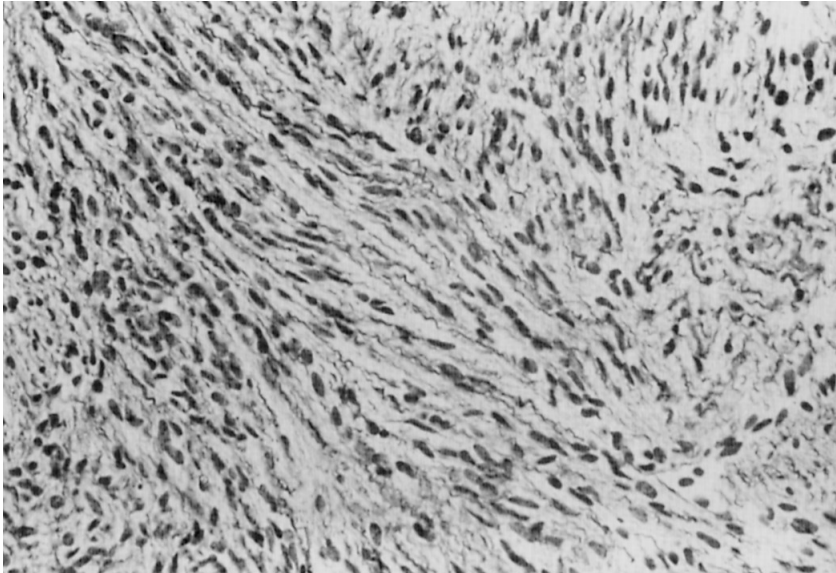


Fig. 5. Bodian staining of the specimen, showing positive staining of numerous nerve-like fibers. (Bodian stain, $\times 200$)

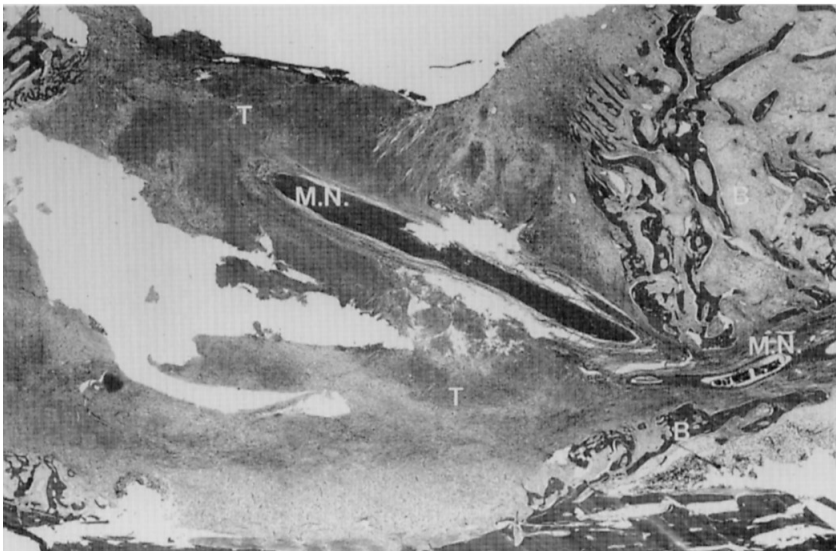


Fig. 6. Histological findings of the resected mandible. (Hematoxylin-eosin stain, original magnification, $\times 80$) The mandibular nerve was observed in the central part of the tumor. The tumor cells invaded the surrounding bone along the nerve.

(M. N.) Mandibular nerve (T) Tumor (B) Invaded bone

DISCUSSION

Polak²⁾ and his associates reported on neurofibromas of the mandible. According to their report, neurofibroma of the mandible appears to have a predilection for females under 24 years of age, and for the posterior mandible.

Most neurofibromas occurring centrally in the mandible have been associated with neurofibromatosis. Our patient did not have café-au-lait spots on the trunk nor any other common clinical features of Von Recklinghausen's disease. Solitary neurofibroma is a benign, slow-growing neoplasm, relatively circumscribed but not encapsulated, originating within a nerve and composed of Schwann cells, perineural cells, and mature collagen. In the present case the mandibular nerve was observed in the central part of the tumor on histological examination. It may be a typical finding that the tumor arises from the mandibular nerve.

The absence of encapsulation makes complete surgical removal of a solitary neurofibroma difficult, which probably accounts for some cases of recurrence.

In the present case, deep invasion of the tumor cells into the surrounding bone along the mandibular nerve was observed on histological examination of the resected mandible. Therefore, the mandibular body including the surrounding bone and overlying mucosa were removed because of the potentiality of tumor invasion into the surrounding tissue. In addition, the mandibular nerve with positive extension along the canal was excised³⁻⁵⁾.

In the Japanese literature, three cases of intraosseous neurofibroma in the mandible have been reported. One was a case report of a 24-year-old Chinese man who lived in Japan. The other two cases were a 32-year-old woman with involvement in the left premolar region and a 21-year-old woman with a tumor in the anterior part of the mandible. These Japanese patients had a solitary oval radiolucent image on a panoramic radiogram. The tumors were found in a routine radiographic examination for dental treatment, and the patients were without special symptoms such as mandibular swelling, severe pain, or mental nerve palsy. All tumors were removed surgically with segmental resection of the mandible. Their prognoses are not identified because of the absence of follow-up reports of the patients.

ACKNOWLEDGEMENTS

We are grateful to Dr. M. Hananouchi, Division of Pathology, Chubu Rosai Hospital, for his technical assistance.

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