

# Solitary intraosseous neurofibroma in the mandible mimicking a cystic lesion: A case report and review of literature

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## Abstract

### BACKGROUND

Neurofibromas are benign tumors of a neurogenic origin. If these tumors occur without any other signs of neurofibromatosis, they are classified as isolated neurofibromas. Neurofibromas in the oral cavity mostly occur within soft tissues, indicating that solitary intraosseous neurofibromas in the mandible are rare. Due to the absence of specific clinical manifestations, early diagnosis and treatment of these tumors are difficult to achieve.

### CASE SUMMARY

A 37-year-old female patient visited our hospital due to numbness and swelling of the gums in the right lower molar area that had persisted for half a month. The patient's overall condition and intraoral examination revealed no significant abnormalities. She was initially diagnosed with a cystic lesion in the right mandible. However, after a more thorough examination, the final pathological diagnosis was confirmed to be neurofibroma. Complete tumor resection and partial removal of the right inferior alveolar nerve were performed. As of writing this report, there have been no signs of tumor recurrence for nine months following the surgery.

### CONCLUSION

This case report discusses the key features that are useful for differentiating solitary intraosseous neurofibromas from other cystic lesions.

**Key Words:** Neurofibromas; Mandible; Cystic lesion; Odontogenic cyst; Schwannomas;

## Case report

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**Core Tip:** We present the case of a 37-year-old female with a solitary intraosseous neurofibroma in the right mandible, accompanied by tooth root resorption and local sensory abnormalities. Imaging revealed low-density unicysts with clear borders. The area affected by the inferior alveolar nerve canal was locally dilated, without displacement. A histological evaluation of the mass revealed spindle-shaped fibroblasts and fibroblast-like cell proliferation with a slight increase in nuclear size. Immunohistochemistry results showed positive staining for S100 and CD34 markers. Complete tumor resection and partial removal of the right inferior alveolar nerve were performed.

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## INTRODUCTION

Neurofibromas are typically benign tumors that originate from the sheath of the peripheral nervous system. They can present as isolated lesions or as a part of neurofibromatosis type 1 (NF1), a systemic syndrome also known as von Recklinghausen disease[1]. NF1 is a common neurocutaneous disorder with an autosomal dominant inheritance pattern; however, solitary neurofibromas do not exhibit this pattern[2]. Neurofibromas occur predominantly in soft tissues and less frequently in bones. It is important to note that approximately 20%-60% of oral neurofibromas are associated with neurofibromatosis. They are commonly found in the tongue, buccal mucosa, and vestibular areas[3]. Solitary intraosseous neurofibromas of the head and neck are rare, with those in the posterior part of the mandible being the most prevalent site. The early diagnosis and treatment of neurofibromas can be challenging due to their lack of specific clinical manifestations. However, with the aid of imaging techniques, such as cone-beam computed tomography (CT) (CBCT), cystic lesions can be easily diagnosed, which can affect treatment plans. Timely local organizational biopsies are crucial for an accurate diagnosis, and confirmation can be obtained through pathological examination. This report describes a rare case of a solitary intraosseous neurofibroma in the right mandible accompanied by tooth root resorption and local sensory abnormalities.

## CASE PRESENTATION

### Chief complaints

A 37-year-old female patient visited our hospital with numbness and swelling of the gums in the right lower molar area that had persisted for half a month.

### History of present illness

Fifteen days prior, the patient had visited a local clinic due to numbness and swelling of the gums in the right lower molar area. Oral panoramic radiography revealed low-density lesions at the root tip in the right mandibular molar area, specifically affecting the lower right second molar. Despite discovering the anomaly, the patient did not report any significant pain, bleeding in the lower right gum or teeth, or unusual odor in the mouth. Additionally, the patient denied experiencing any abnormal sensation in the lower right lip.

### History of past illness

There was no significant medical history related to this illness.

### Personal and family history

The patient had no family history of hereditary diseases or malignant tumors.

### Physical examination

The patient's general health was satisfactory, and no anomalies were detected on the skin of the face or the rest of the body. The patient's facial features were symmetrical, with no apparent swelling. No significant buccolingual bulge was observed in the right mandible, nor was there any palpable ping-pong-like sensation. The right mandibular molars exhibited second-degree loosening and discomfort upon percussion. Slight redness and swelling were noticed on the buccal side of the right lower posterior tooth's gingiva, with no sinus, pus discharge, or bleeding. The patient reported

symmetrical sensory acuity in both upper and lower lips.

### Laboratory examinations

The results of routine blood tests were as follows: Mean platelet volume: 8.7 fL (reference value: 9.0-13.0 fL); platelet distribution width: 8.8 fL (reference value: 9.0-17.0 fL).

### Imaging examinations

CBCT revealed a hypodense image measuring approximately 4.7 cm × 2.3 cm × 1.6 cm in the body of the right mandible. A white bone line surrounding the lesion was observed, along with thinning of the cortical bone. The lesion affected the apex of the lower right second molar with evident root resorption. The apical lesion of the lower right first molar seemed isolated, while the lower right third molar was located outside the mass and buried within the bone. The affected area of the inferior alveolar nerve canal was locally dilated without displacement (Figure 1).

## FINAL DIAGNOSIS

After comprehensive assessment of the clinical presentation, blood test results, imaging findings, and pathological examination, the patient was diagnosed with a solitary intraosseous neurofibroma of the right mandible.

## TREATMENT

On September 7, 2022, we performed complete tumor resection, along with partial removal of the right inferior alveolar nerve and extraction of the lower right third molar. During surgery, the tumor appeared as a grayish-white solid and the main trunk of the inferior alveolar nerve was not visible. Microscopic examination revealed spindle-shaped fibroblasts and fibroblast-like cell proliferation, with a slight increase in nuclear size. The nuclei were short and spindle-shaped, the cytoplasm showed rich red staining, and axons were locally visible (Figure 2). Immunohistochemistry results showed positive staining for S100, CD34, and H3K27me3 (Figure 2) and negative staining for smooth muscle actin, neurofilaments protein, and epithelial membrane antigen (Supplementary Figure 1).

## OUTCOME AND FOLLOW-UP

Nine months after the surgery, there were no signs of tumor recurrence. However, the patient developed numbness in the lower right lip, which had not improved at the time of writing this report.

## DISCUSSION

Solitary neurofibromas are slow-growing benign tumors that are not encapsulated. In cases where patients do not exhibit other manifestations of neurofibromatosis, such as axillary freckling, iris hamartomas, or skeletal dysplasia, they are diagnosed with isolated neurofibromas. Neurofibromas of the oral cavity primarily originate from the mucosa. Bone involvement in neurofibromatosis is typically attributed to subperiosteal neurofibromas, which can cause bone erosion [4]. Solitary intraosseous neurofibromas are rare because the bone marrow space lacks nerve sheaths or myelinated nerves [5]. We have compiled the results from 38 studies on the subject, published since the discovery of solitary intraosseous mandibular neurofibromas (Table 1) [5-35]. The posterior part of the mandible is the most common location, with a higher incidence in females. The average age of presentation is 27.5 years [6]. The length and thickness of the inferior alveolar nerve bundles may contribute to the comparatively high frequency of this illness in the mandible [7].

In the early stages, solitary intraosseous neurofibromas may not cause any symptoms; however, as the lesions grow, they can lead to expansion of the mandibular cortex. This expansion may occur with or without destruction and can result in pain, anesthesia, or paresthesia [8]. Further invasion of surrounding tissues may lead to abnormalities that alter mouth opening. However, the clinical symptoms lack specificity. This is the third reported case of a solitary intraosseous neurofibroma in the mandible with tooth root resorption. Therefore, the early diagnosis of solitary intraosseous neurofibromas without obvious symptoms is difficult. In contrast to mandibular cystic lesions, which often manifest as local swelling, loose teeth, and discomfort during biting, our patient presented with local numbness of the gums in the posterior mandibular region. Our initial neglect led to a diagnostic bias. Performing pulp vitality tests on the teeth involved in the lesion can also help identify neurofibromas. It should be noted that the clinical manifestations of non-chief complaint diseases in the adjacent teeth may affect judgments regarding the nature of the tumor, highlighting the need for improved accuracy in the clinical judgment of the chief complaint.

The imaging characteristics of solitary intraosseous neurofibromas typically show low-density unicystic or multicystic images with clear or unclear borders that lack specificity. However, these images may be closely related to those of the mandibular nerve canal. We have compiled a list of mandibular diseases that can easily be mistaken for solitary intraosseous neurofibromas (Table 2). The radiological appearance of this patient closely resembled periapical cysts,

Table 1 Summary of reports of solitary intraosseous neurofibromas of the mandible

Ref.	Age (year)/sex	Site	Clinical features	Radiographic features	Root resorption	Size	Immunohistochemistry features	Surgical approach	Prognosis
Present case	37/F	Body	Numbness and swelling of the gum in the right lower molar area	Unilocular expansile radiolucency involving the inferior alveolar nerve canal	Yes	4.7 cm × 2.3 cm × 1.6 cm	Positive staining for S100, CD34, and H3K27me3, negative staining for SMA, NF, and EMA	Excision of the tumor and nerve	No recurrence observed nine months post-surgery
Kamalakaran <i>et al</i> [5]	5/M	Body-ramus	Limited mouth opening and enlarged submandibular lymph nodes	Expansile lesion with a mixture of radiolucent and radiopaque components	No	3.9 cm × 3.8 cm × 3.1 cm	Not reported	Segmental resection of the mandible	No recurrence observed one-year post-surgery
Sarkar <i>et al</i> [6]	2.5/M	Body-ramus	Swelling in the posterior right part of the mandible. Displacement of deciduous teeth	Well-defined expansile solid-cystic lesion with evidence of cortical erosion	No	3.7 cm × 3.5 cm × 3.4 cm	S-100 multifocal and moderately positive, strong CD34 positivity EMA, cytokeratin, CD31, SMA, STAT-6, TLE-1, and HMB-45 negative MIB-1 index low	Preservation of the nerve while removing the mass	Not reported
Behrad <i>et al</i> [13]	32/F	Body	Swelling and intermittent dull pain in the left mandible	Unilocular expansile radiolucency involving the inferior alveolar nerve canal	No	3 cm × 1.5 cm × 2.4 cm	Not reported	Simple excision of the mass	Not reported
Iqbal <i>et al</i> [14]	13/M	Body-angle	Swelling in the posterior area of the right mandible	Ill-defined radiolucency	No	2.0 cm × 3.0 cm × 2.0 cm	S-100 positive	Simple excision of the mass	No recurrence
Inoue <i>et al</i> [15]	27/M	Ramus to infratemporal fossa	Progressive numbness on the lower right jaw	A nonenhanced mass in the infratemporal region, with an enlarged inferior alveolar nerve canal and foramen ovale	No	Not reported	S-100 positive	Excision of the tumor and nerve	No recurrence
Narang <i>et al</i> [16]	45/F	Left mandibular angle to right mental foramen	Swelling in the posterior area of the left mandible	The radiographic irregularity in the region of the inferior lacrimal canal	Yes	Not reported	S-100 positive, MIB-1 negative	Segmental resection of the mandible	Not reported
Fortier[17]	70/F	Body	Osteolytic lesion of the right posterior mandible	The clear single-eyed translucent area. Involvement of the inferior alveolar nerve canal	No	Not reported	S-100 positive	Preservation of the nerve while removing the mass	No recurrence
Gujjar <i>et al</i> [18]	28/F	Body	Swelling and	Uniform radiopacity.	No	3.0 cm × 4.0 cm	S-100 positive	Segmental resection	Not reported



			intermittent dull pain in the left mandible	Involvement of the inferior alveolar nerve canal				of the mandible	
Saravani <i>et al</i> [19]	39/F	Body	Severe pain in the right posterior mandible	Relatively clear single-cyst projection shadow	No	Not reported	S-100 positive	Simple excision of the mass	No recurrence
Jangam <i>et al</i> [20]	62/F	Right body-left body	The lower jaw is significantly swollen. Occasionally, there is severe pain	Radiographically transparent image with clear borders, accompanied by thinning of the lower boundary	Edentulous jaw	Not reported	S-100 positive	Segmental resection of the mandible. Repair using a free fibular graft	No recurrence
Deichler <i>et al</i> [21]	14/M	Ramus	No clinical symptoms, discovered incidentally	Unilocular radiolucency	No	4.0 cm × 1.5 cm × 0.5 cm	Tumor cells: Vimentin positive, neurospecific enolase (NSE) positive and anti S-100 negative. Residual nerve fibres: S-100 positive; NSE positive	Simple excision of the mass	Not reported
Tao <i>et al</i> [22]	16/F	Ramus	Limited mouth opening, numbness in the lower left lip	Multilocular radiolucency with irregular edges	No	3.5 cm × 2.0 cm	S-100 positive	Segmental resection of the mandible. Repair using a free iliac bone graft	Not reported
Vivek <i>et al</i> [7]	39/F	Symphysis-parasymphysis	Spontaneous tooth loss, persistent tingling sensation in the lower lip	A relatively well-defined radiolucent area	No	Not reported	S-100 positive	Segmental resection of the mandible	No recurrence observed one-year post-surgery
Apostolidis <i>et al</i> <sup>[8]</sup>	67/F	Body-ramus	Abnormal sensation in the lower left lip	Involvement of the inferior alveolar nerve canal	No	2.5 cm × 2 cm × 0.7 cm	Not reported	Excision of the tumor and nerve	No recurrence observed three-year post-surgery
Alatli <i>et al</i> [23]	37/F	Body	Abnormal sensation in the lower right lip	No abnormalities detected	No	Diameter of 1.3 centimeters	Not reported	Excision of the tumor and nerve	No recurrence observed two years post-surgery
Ueda <i>et al</i> [24]	37/M	Body	No clinical symptoms, discovered incidentally	Well-defined unilocular radiolucency. Involvement of the inferior alveolar nerve canal	No	Not reported	S-100 positive	Segmental resection of the mandible. Repair using a free scapular flap	No recurrence observed three years post-surgery
Papageorge <i>et al</i> [10]	4.5/M	Ramus-infratemporal fossa	Expansile lesion in the right mandible. Limited mouth opening, chin deviated to the left	Well-defined unilocular radiolucency. Involvement of the inferior alveolar nerve canal	No	Not reported	S-100 protein and vimentin positive	Segmental resection of the mandible. Reconstruction using autologous rib cartilage graft	No recurrence

Weaver <i>et al</i> [25]	22/F	Body	Swelling on the left side of the face	Well-defined unilocular radiolucency	No	5.0 cm × 3.0 cm	S-100 positive	Excision of the tumor and nerve.	No recurrence observed six months post-surgery
Polak <i>et al</i> [26]	60/M	Body	No clinical symptoms discovered incidentally. When palpated, a crackling sensation can be felt, similar to the sound of breaking eggshells	Unilocular radiolucency involving the mandibular canal	No	Not reported	S-100 and anti-Leu positive	Excision of the tumor and nerve	No recurrence observed six years post-surgery
Papadopoulos <i>et al</i> [27]	15/M	Body	Swelling and mild pain in the left mandible	Unilocular radiolucency near mental foramen	No	0.5 cm × 0.5 cm × 0.5 cm	Not reported	Excision of the tumor and nerve	No recurrence
Larsson <i>et al</i> [11]	25/F	Body-ramus	No clinical symptoms, discovered incidentally	Well-defined large radiolucency. Involvement of the inferior alveolar nerve canal	No	10 cm × 2.0 cm	Not reported	Excision of the tumor and nerve, with nerve end-to-end anastomosis	No recurrence observed two years post-surgery
Larsson <i>et al</i> [11]	46/M	Body	Swelling and slow, intermittent pain in the left mandible	Rounded, slightly radiopaque, well-circumscribed radiolucency	No	Diameter of 1 cm	Not reported	Remove all visibly abnormal tissues within the bone	No recurrence observed five months post-surgery
Ellis <i>et al</i> [28]	41/F	Body-ramus	Swelling in the right mandible	Poorly defined multilocular radiolucent lesion	No	Not reported	Not reported	Not reported	Not reported
Ellis <i>et al</i> [28]	4/F	Body	A firm lump on the right mandible	Well demarcated radiolucent lesion. Involvement of the inferior alveolar nerve canal	No	2.5 cm × 1.6 cm	Not reported	Not reported	No recurrence observed one-year post-surgery
Ellis <i>et al</i> [28]	8/M	Body-angle	Not reported	Well demarcated radiolucent lesion with sclerotic borders	No	6.0 cm × 4.0 cm	Not reported	Not reported	No recurrence observed one-year post-surgery
Ellis <i>et al</i> [28]	23/F	Body-ramus	Swelling in the posterior area of the right mandible	Radiolucent-radiopaque lesion with indistinct borders	No	Not reported	Not reported	Not reported	Recurrence observed after 3 yr. Partial mandibulectomy performed
Ellis <i>et al</i> [28]	4/M	Body	Swelling in the right mandible	Multilocular radiolucent lesion	No	Not reported	Not reported	Not reported	Not reported
Cundy and Matukas [29]	55/F	Body-angle	Pain and swelling in the left mandible, accompanied by discomfort while chewing	Unilocular radiolucency	No	Not reported	Not reported	Segmental resection of the mandible. Repair using a free iliac bone graft	Not reported

Cassalia and Miller [30]	16/F	Angle	No clinical symptoms, discovered incidentally	Multilocular radiolucency involving mandibular canal	No	Not reported	Not reported	Preservation of the nerve while removing the mass	No recurrence observed six months post-surgery
Sharawy and Springer [31]	22/F	Body-angle	Intermittent numbness in the left mandible, with slight swelling on the cheek side	Multilocular expansile radiolucency	No	Not reported	Not reported	Excision of the tumor and nerve.	No recurrence observed thirteen months post-surgery
Gutman <i>et al</i> [32]	5/F	Body	Painless swelling in the left mandible	Unilocular expansile radiolucency	No	2 cm × 4 cm	Not reported	Excision of the tumor and nerve	No recurrence observed sixteen months post-surgery
Gohel <i>et al</i> [12]	17/F	Body	A swollen lump on the gums of the lower right mandible	Osteolytic radiolucent lesion	No	4 cm × 1 cm	Not reported	Not reported	Not reported
Johnson <i>et al</i> [9]	34/F	Body	Abnormal sensation on the left side of the lower lip and tenderness in the lower left jaw during biting	Unilocular radiolucency	No	2.5 cm × 1.5 cm	Not reported	Not reported	Skin pigmentation observed six months later, followed by local recurrence two years post-surgery
CORNELL and VARGAS [33]	65/F	Body	Oral discomfort	Ill-defined unilocular lesion with unclear borders	Edentulous jaw	Not reported	Not reported	Excision of the tumor and nerve	No recurrence
BRUCE [34]	36/M	Body	A swelling on the alveolar ridge of the edentulous lower left jaw	Well-defined radiolucency involving mandibular canal	No	3 cm × 2 cm × 2 cm	Not reported	Not reported	Not reported
Blackwood and Lucas [35]	41/M	Body	Swelling in the posterior area of the left mandible	Unilocular radiolucency	Yes	2 cm × 1.5 cm × 1 cm	Not reported	Simple excision of the mass	Not reported

SMA: Smooth muscle actin; NF: Neurofilaments protein; EMA: Epithelial membrane antigen; STAT6: Signal transducer and activator of transcription 6; TLE: Temporal lobe epilepsy; HMB-45: Human melanoma black-45.

unicystic ameloblastoma (UA), and odontogenic keratocysts. Periapical cysts are inflammatory odontogenic cysts and are generally the most common type of jaw cysts. They are associated with the apex of non-vital teeth [9]. Radiographically, periapical cysts show well-defined unilocular radiolucency with sclerotic borders in close proximity to the adjacent teeth [36]. UA is a variant of ameloblastoma that presents as a cyst and shares clinical and radiological characteristics with odontogenic cysts. They appear as well-defined unilocular radiolucencies that often surround the crown of an impacted tooth [37]. In the present case, the impacted tooth crown was located outside the lesion. Compared to ameloblastomas, odontogenic keratocysts exhibit less prominent buccolingual expansion and less frequent and severe adjacent tooth root resorption. The presence of pasty fluid in odontogenic keratocysts can lead to areas of attenuation within the cystic cavity, resulting in uneven internal density on CT [38]. Papageorge *et al* [10] and Larsson *et al* [11] observed radiographic evidence of calcification in solitary intraosseous neurofibromas and proposed that this might be the result of high collagen content

**Table 2 Differential diagnosis of benign lesions in the mandible**

Disease	Age (year)	Sites	Radiographic Findings
Radicular (periapical) cysts	In the 3 <sup>rd</sup> to 5 <sup>th</sup> decades of life	Located at the apices of dead pulp teeth, caused by inflammation of the apical tissue due to caries or trauma	Circular, unilocular low-density images at the apex of the tooth. Massive periapical cysts may cause root resorption, displacement of adjacent structures, and expansion
Dentigerous cysts	In adolescents and young adults	The upper canine and lower third molar	Clear radiolucent image around the tooth crown with cortical border causing a significant displacement. Expansion and root resorption may be present. The cortical border is usually preserved
Simple bone cysts	In the 2 <sup>nd</sup> decades of life	The posterior part of the mandible	A radiolucency scalloping between the roots of the teeth
Odontogenic keratocysts	In the 3 <sup>rd</sup> decades of life	The posterior part of the mandible	Unilocular/multilocular lesions with scalloped margins. May present as radiolucent around the tooth crown. May lead to cortical thinning, tooth displacement, and root resorption
Ameloblastoma	In the 3 <sup>rd</sup> to 5 <sup>th</sup> decades of life	The posterior part of the mandible	Well-defined unilocular/multilocular radiolucency. May appear as a pericoronal radiolucency. Extensive root resorption. Has a typical “soap bubble” appearance
Aneurysmal bone cyst	In young adults	The posterior part of the mandible	A unilocular or multilocular radiolucency with cortical expansion
Central giant cell granuloma	In adolescents and young adults	The anterior part of the mandible	Mandibular border is clearly defined. Radiolucent with granular calcifications. May cause tooth displacement, root absorption, destruction of cortical plates, and invasion into surrounding soft tissues. Early-stage lesions: Small unilocular areas (similar to odontogenic cysts). Progressive stage: Multilocularity with sparse internal septa and bone expansion
Schwannomas	Most often in the 4 <sup>th</sup> and 5 <sup>th</sup> decades	The posterior part of the mandible	Radiolucent, unilocular, homogeneous, and well-defined lesions with cortical expansion
Neurofibromas	In adolescents and young adults	The posterior part of the mandible	Low-density unicystic or multicystic images with clear or unclear borders. They can expand and penetrate the boundary of the dermis. Tooth root absorption and tooth displacement may also occur. Fusiform enlargement of the mandibular canal
Central hemangioma	In the 2 <sup>nd</sup> decades of life	It mainly occurs in the spine. The mandible is a very rare location, with the posterior part of the mandible being slightly more common	Unilocular or multilocular lesion, with large bone marrow spaces and rough trabeculae formation. Accompanied by a typical honeycomb or soap-bubble appearance. When it occurs within the inferior alveolar canal, the canal was wider than normal
Arteriovenous malformation	In the 1 <sup>st</sup> decades of life	They are uncommon lesions of the head and neck. The majority of jaw lesions occur in the mandibular ramus and body	May appear cystic due to adjacent bone resorption. May appear multilocular. When located within the inferior alveolar canal, the canal can enlarge throughout the entire course
Lymphoma	Most patients aged between 40 and 60 yr	Mandibular body	Radiolucent pattern with non-transmitted radiation particles and reactive bone attachment. May cause “moth-eaten” appearance, lamellar bone formation in periosteum, widening of mandibular canal, irregular increase in periodontal ligament width, and tooth spacing

rather than genuine calcification. Odontogenic lesions in the mandible originate above the mandibular canal. Neural and vascular lesions mostly arise within the mandibular canal, whereas lesions centered below the mandibular canal are usually of non-odontogenic origin[12]. Therefore, it is crucial to carefully analyze the correlation between lesions and the inferior alveolar nerve canal. If the lesion involves the inferior alveolar nerve canal, it is essential to consider that the swelling may have a neural origin.

Histologically, solitary neurofibromas originate from the nerves and are composed of Schwann cells, perineural cells, endoneurial fibroblasts, and intermediate cells[39,40]. They have well-defined boundaries with the surrounding connective tissue, distinguishing them from the multiple neurofibromas observed in cases of neurofibromatosis[3]. It is crucial to perform S-100 and CD34 immunostaining on biopsy samples. In neurofibromas, tumor cells are loosely arranged and fragile, often with wavy or snake-shaped nuclei, and S-100 protein-positive cells are less common than in schwannomas[41]. The final pathological diagnosis should be based on the presence of CD34, which is located in the cell membrane and cytoplasm. CD34 positivity is observed in neurofibromas but not in schwannomas[42].

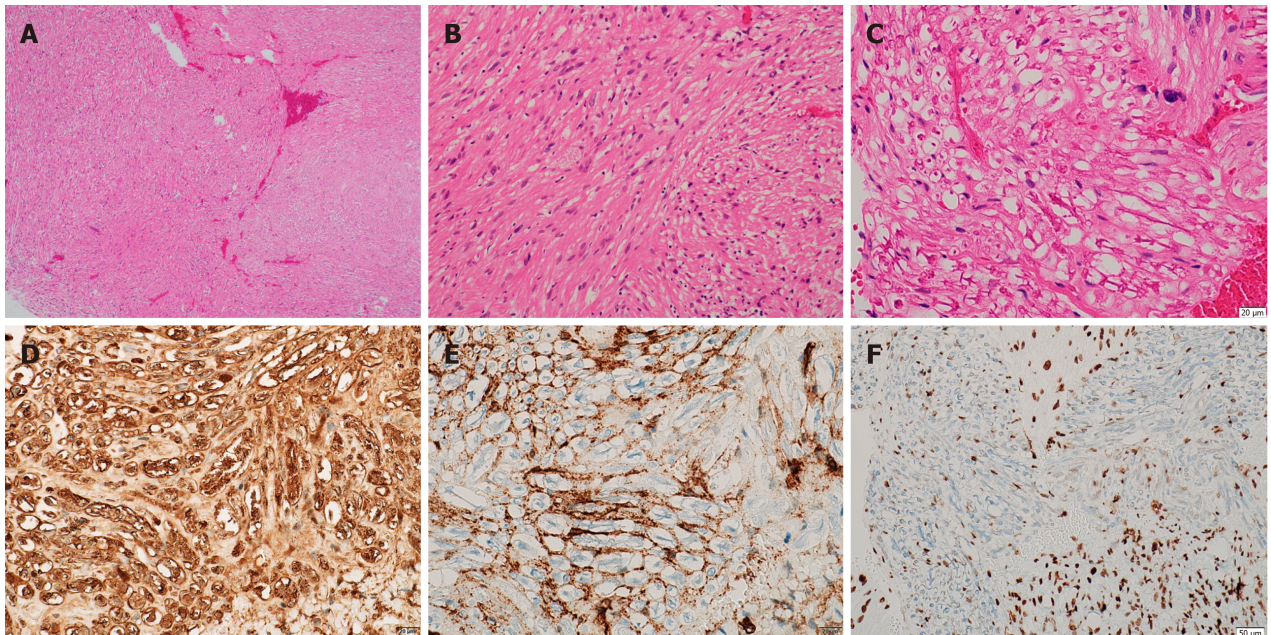
Surgical resection is currently the primary treatment for solitary intramedullary intraosseous neurofibromas. However, there have been no reports of adjuvant therapy, and the local recurrence rate of neurofibromas is higher than that of schwannomas, possibly because of the lack of an envelope[43]. This makes complete tumor removal more challenging, and the affected nerve is sacrificed during radical resection of neurofibromas[8]. While neurofibromas may progress to neurofibromatosis or undergo malignant transformation as the primary disease[44,45], the likelihood of solitary neurofibromas becoming malignant is quite low compared to that of neurofibromatosis[46]. The possible development of this disease emphasizes the importance of regular monitoring for patients with early neurofibroma symptoms. Our





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**Figure 1 Imaging examination.** A: The oral cone-beam computed tomography (CT) revealed a hypodense image with a size of approximately 4.7 cm × 2.3 cm in the body of the right mandible. A white line is observed around the lesion. The lesion affected the apex of tooth 47 with visible root resorption. The area affected by the inferior alveolar nerve canal was locally dilated without displacement; B: CT (axial section) showing an oval-shaped, low-density area of the right mandible accompanied by cortical bone thinning.



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**Figure 2 Microscopic features of neurofibroma.** A-C: Histopathological examination with toluidine blue and hematoxylin and eosin staining showing spindle fiber and fibroblast-like cell proliferation with slight nuclear enlargement. The nuclei were short and spindle-shaped, the cytoplasm was richly stained red, and axons were locally visible; D: Immunohistochemistry positive for S-100 protein; E: Immunohistochemistry positive for CD34; F: Immunohistochemistry positive for H3K27ME3.

patient showed no signs of recurrence or progression to neurofibromatosis nine months after surgery. In the future, we will continue to monitor these patients.

## CONCLUSION

In this report, we detail the case of a 37-year-old female with tooth root resorption and local sensory abnormalities in the right mandible, indicative of a solitary intraosseous neurofibroma. While pathological examination remains the primary diagnostic method for intraosseous neurofibroma, clinicians must carefully examine the patient's symptoms and observe the relationship between the lesion and inferior alveolar nerve canal. An accurate distinction between cystic lesions and neurogenic tumors in the mandible is crucial for early diagnosis and appropriate treatment. Following surgical intervention, patients with solitary neurofibromas of the mandible should undergo long-term follow-up due to the potential for local recurrence and malignant transformation of these tumors.

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## FOOTNOTES

**Author contributions:** Zhang Z reviewed the literature and drafted the manuscript; Yin Y provided professional opinions related to pathology; Hong X, Wang F, Ye X, Yao YD, and Yang HY were responsible for revising the manuscript for intellectual content; All authors issued final approval for the version to be submitted.

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