Journal of Craniomaxillofacial Research

Vol. 10, Issue. 4 Autumn 2023

Solitary Intraosseous Neurofibroma Of Mandible: A Case Report

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ARTICLE INFO

Article Type:
Case Report

Received: 10 August 2023
Revised: 21 September 2023
Accepted: 19 October 2023

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ABSTRACT

Neurofibroma is a benign tumor derived from the peripheral nerve. Most of these can be multifocal as a component of neurofibromatosis or present as a solitary tumor. Although the solitary neurofibroma is not an uncommon lesion, its intraosseous occurrence is rare, and less than 50 cases have been reported. We report a rare case of central neurofibroma, arising in the mandibular bone of 57-year-old edentulous female patient on the left side. Hemi mandibulectomy was performed and reconstructed with a reconstruction plate without any bone graft. Microscopic evaluation showed neoplastic tissue with haphazardly arranged and interlacing fascicular patterns composed of point-ended spindle-shaped tumoral cells. Immunohistochemical examination of the tumor showed positive staining for S-100 protein antibodies.

Keywords: Intraosseous neurofibroma; Mandible.

Please cite this Article as:
Abbasi AJ, Aminishakib P, Arashrad M. Solitary Intraosseous Neurofibroma Of Mandible: A Case Report. J Craniomaxillofac Res 2023; 10(4): 208-211. DOI: 10.18502/jcr.v10i4.15314

Introduction

Neurofibroma is a benign tumor originating from peripheral nerve sheath cells [1] and is the most common type of peripheral nerve neoplasm. It may be composed of various types of cells such as Schwann cells, perineural cells, fibroblasts and intermediate cells [2]. Neurofibroma can be multifocal as a component of neurofibromatosis (Von Recklinghausen's disease of the skin) or presents as a solitary tumor [3]. Solitary tumors are soft, painless, slow-growing lesions that usually involve skin tissue [3]. Solitary neurofibromas of the oral cavity are not uncommon and the most frequent sites are tongue and buccal mucosa [3-5]. Nerve sheath neoplasms rarely arise centrally within jaw bones. Since the first case of solitary neurofibroma of the oral cavity was described in 1954, 6 less than 50 cases have been documented in English literature up to now [7,8] and few of them are centrally located. When solitary neurofibroma occurs in the mandible, shows a 2:1 female-to-male ratio and occurs mostly in the posterior region of the mandible [3]. The tumor usually involves young adults with a mean age of 27.5 years old [9]. Here, we present a rare case of intra-osseous neurofibroma in a 57-year-old female and also discuss the diagnostic and therapeutic dilemmas of this lesion.

Case Report

A 57-year-old edentulous female patient was referred by her dentist to the Oral and Maxillofacial Surgery Department of Shariati Hospital with the chief complaint of denture irritation, during the past 2 months. Extraoral examination showed a mild facial swelling on the left side mandibular area. The overlying skin tissue was intact and there was no evidence of color change, scar or fistula. No sensory disturbance of the lower lip including pain or paresthesia was reported. Intraoral examination revealed buccal cortex expansion on the left side mandibular body, without crepitation or eggshell crackling. The overlying mucosa was rather tender in some areas which seemed because of long-lasting denture irritation. Also, the lesion showed a solid essence on the aspiration test. Then, the patient was referred for radiographic examination. On radiologic examination, the panoramic view revealed a well-defined multi-locular radiolucent lesion on the left side of the mandible, which extended from the angle of mandible to sigmoid notch anteroposteriorly, in which the anterior border of the ramus could not be recognized. The inferior alveolar canal could not be followed through the lesion. The lesion caused expansion of the body of mandible, ramus, sigmoid notch and condylar neck. The lesion contains coarse septa which resulted in a "SoapBubble" appearance (Figure 1). Also, computed tomography scan showed a lytic expansile lesion in the mandibular body and ramus with cortex disruption in some areas. Mandibular condyle was intact (Figure 2). Based on clinical and radiographic findings, Odontogenic Keratocyst, Ameloblastoma and were proposed as provisional diagnoses.

Therefore, to make a definite diagnosis an incisional biopsy was performed under local anesthesia. Microscopic evaluation of the specimen showed neoplastic tissue with haphazardly arranged and interlacing fascicular patterns composed of point-ended spindle shaped tumoral cells (Figure 3). A variable amount of hypo and hypercellular foci are distributed among delicate collagen bundles (Figure 4). No evidence of malignancy, including necrosis, atypical mitotic figures or invasion to surrounding normal tissue, was seen. After immunohistochemical positive staining of tumoral cells for Vimentin and S100 (Figure 5), the diagnosis of "Neurofibroma" was rendered. Consequently, the patient was examined to rule out/in any probable syndromic nature for the tumor, but no freckling, café-au-lait spots or subcutaneous nodules were found. The patient was admitted to the hospital and resection was performed from left side condyle to the left side parasymphis area through cervicotomy approach. After admitting the primary diagnosis and reporting free surgical margins by pathologist,the resected mandible was reconstructed by a 15 holes condylar reconstruction plate without any bone graft (Figure 6). Grossly a gray soft to firm mass measuring 5x3x2cm was observed one cm far from antrerior surgical margin as the closest margin. On cut sections homogenous white, gray solid surfaces were seen which significantly invaded the surrounding mandibular bone. Postsurgical follow-up after 18 months revealed no recurrence and no evidence of Neurofibromatosis.

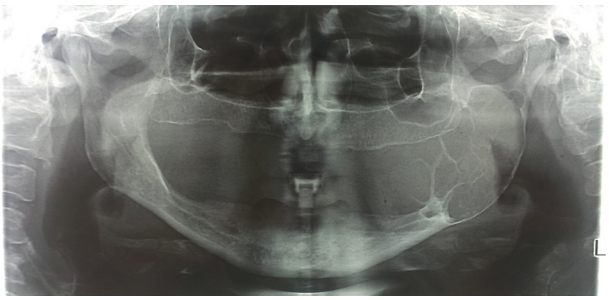

Figure 1. Preoperative panoramic radiogram of the patient.

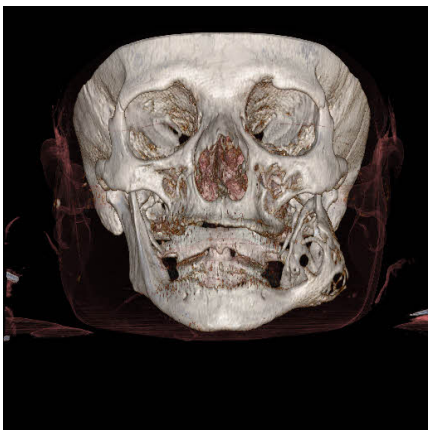

Figure 2. Three-Dimensional CT scan of the patient, showing an expansion.

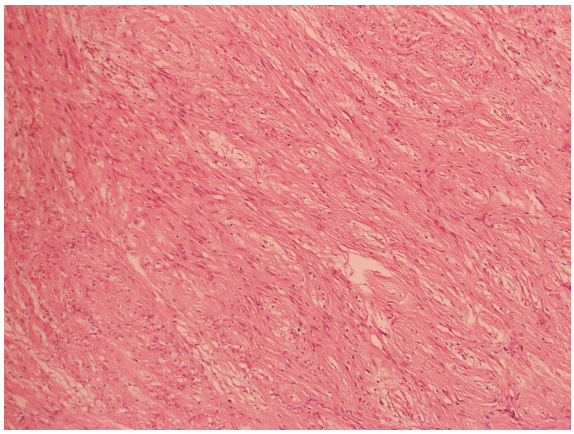

Figure 3. Photomicrograph shows moderately cellular neoplasm composed of interlacing bundles of spindle-shaped cells. (H&E staining, original magnification X40).

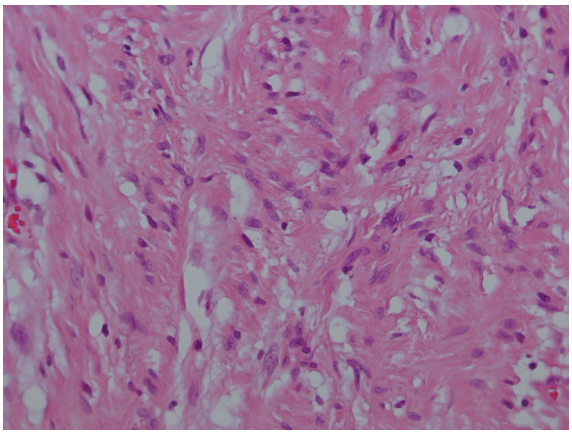

Figure 4. Photomicrograph shows spindle-shaped cells mostly exhibit wavy nuclei and are associated with delicate collagen bundles. No evidence of cellular atypia is seen. (H&E staining, original magnification X200.

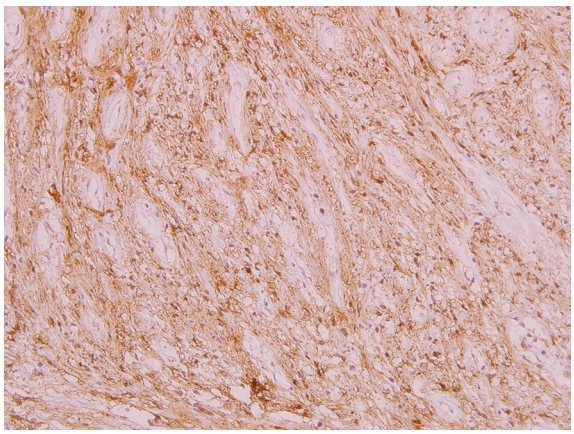

Figure 5. Positive immunoreactivity of tumoral cells with S100 antibody (×40).

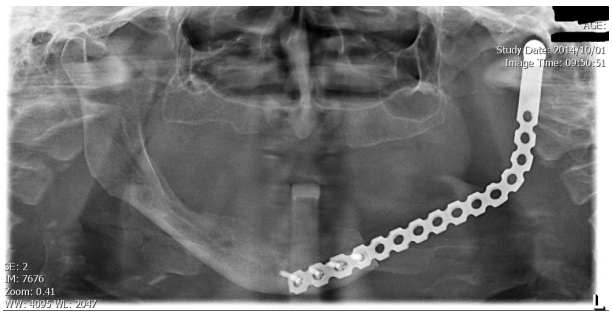

Figure 6. Postoperative panoramic radiogram of the patient.

Discussion

Intraosseous nerve sheath tumors are extremely rare 10 and only few cases of central neurofibroma of mandible have been published [11]. Solitary neurofibroma diagnosis requires thorough physical examination and family history to rule out/in Von Recklinghausens disease because about 90 percent of neurofibromas are associated with Neurofibromatosis (NF) type 1 [9,12-14]. Our Patient had no clinical signs of neurofibromatosis. Solitary neurofibroma is a slow-growing benign neoplasm originating within nerve and composed of complex proliferation of Schwann cells, perineural cells, endoneural fibroblasts and intermediate cells [2,15]. The neurofibroma is a nonencapsulated neoplasm, allowing the tumor to infiltrate the peripheral tissue, making complete surgical removal difficult especially in the peripheral type [16]. Difficulty of surgical removal probably accounts for some cases of recurrences [17]. However in central neurofibroma lack of encapsulation poses less difficulty. Neurofibromas can transform to malignant lesions especially when associated with neurofibromatosis and 8% to 13% of malignant transformation has been reported [16,18]. Since malignant changes and recurrence have been reported, it is necessary to follow the patient for long period.

DOI: 10.18502/jcr.v10i4.15314

Solitary neurofibroma is also proposed to be the first manifestation of neurofibromatosis that may develop at later stage [18,19]. As the patient did not agree with any bone graft harvest, mandible reconstruction was performed only by reconstruction plate with the condylar process and without any bone graft. Neurofibroma is extremely rare in the head and neck region, especially the jaws. Due to this rarity, the microscopic diagnosis of the lesion is rather challenging and most pathologists almost rely on immunohistochemical staining to make a definite diagnosis. The vast majority of intra osseous, nonodontogenic spindle cell lesions of the jaws are composed of interlacing wavy shaped benign looking cells with inconspicuous cytoplasm which makes it difficult to be differentiated from this entity. Therefore, additional molecular examination is necessary to achieve the final diagnosis.

Conflict of Interest

There is no conflict of interest to declare.

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