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CASE REPORT

Central Neurilemmoma of Mandible

Raghavendra Byakodi, Vaishali Keluskar, Anjana Bagewadi, Arvind Shetti

ABSTRACT

Neurilemmomas involving bones are rare tumors constituting less than 1% of central benign tumors of bone. Only few cases of neurilemmomas occurring in the mandible have been reported in the medical literature so far. Here, we report a case of neurilemmoma involving the posterior aspect of mandible in a 15 years old male child. This case was a diagnostic challenge as the clinical features and radiographic features mimicked that of an odontogenic cyst but histologically turned out to be a neurilemmoma. Discussion on the incidence, clinical presentation, radiographic appearance, histopathological features and treatment of this benign neurogenic tumor is presented here.

Keywords: Neurilemmoma, Schwannoma, Tumors.

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INTRODUCTION

Neurilemmoma (schwannoma) is a rare benign tumor originating from the Schwann cells of the neural sheath, of a myelinated nerve fiber.[1-9] Neurilemmomas involving the soft tissues are more common than intraosseous lesions and either of them is rarely seen in the oral cavity. Neurilemmomas involving bones are rare tumors constituting less than 1% of central benign tumors of bone.[3] The jaws especially the posterior mandible seems to be a site of predilection of central neurilemmomas arising from inferior alveolar nerve, a branch of mandibular nerve.[3,4] Other sites reported include the sacrum, vertebra, skull, maxilla, clavicle, scapula, sternum, ribs, humerus, radius, ulna, ilium, pubic bone, femur, patella, fibula, tibia and bones of hand and feet.[5]

Neurilemmomas can occur at any age group, but are most commonly reported between 8 and 72 years with a peak prevalence in the second and third decades of life.[3,5-7] They have a slight female predilection, with a 1.5:1 female to male ratio.[5]

Chi (2003) et al reviewed the literature on intraosseous neurilemmomas, with a special attention to cases arising in the mandible and maxilla. Their review confirmed the mandible to be the most common site for the occurrence of intraosseous neurilemmomas, with only 43 acceptable gnathic cases previously reported and added one case of their own.[5]

CASE REPORT

A 15 years old male patient presented to Outpatient Department of KLE Society's VK Institute of Dental Sciences, Belgaum, India with a chief complaint of swelling on the left side of the lower jaw since 6 months (Fig. 1). The child's parents were concerned about his facial disfigurement and desired cosmetic improvement. There was no history of pain, ulceration or discharge associated with the swelling, so also there was no positive history of trauma. Past history revealed that swelling was initially small and gradually increased to the present size of 4 × 4 cm. Patient's medical history and family history were noncontributory. Patient was in good general health and systems review was unremarkable.

Extraoral examination revealed a diffuse swelling measuring about 4 × 4 cm in size, present on the left side of the mandible (Fig. 1). Skin overlying and surrounding the swelling was normal in color and no pulsation noted. On palpation swelling was nontender and bony hard in consistency. There was no rise in temperature nor paresthesia or anesthesia present. Borders of the swelling were distinct and round. It was extending anteriorly till the left commisures, posteriorly till posterior border of the mandible, superiorly till ala-tragus line and inferiorly extending 1 cm below the lower border of the mandible. The skin overlying the swelling was pinchable. The swelling was noncompressible, nonfluctuant and nonpulsatile in nature. Submental, submandibular and cervical lymph nodes were unremarkable.

Temporomandibular joint examination did not reveal any association with the swelling nor did the swelling have any affect on the normal functioning of the joint.

Intraoral examination revealed a solitary swelling in the left retromolar region extending anteriorly till first premolar

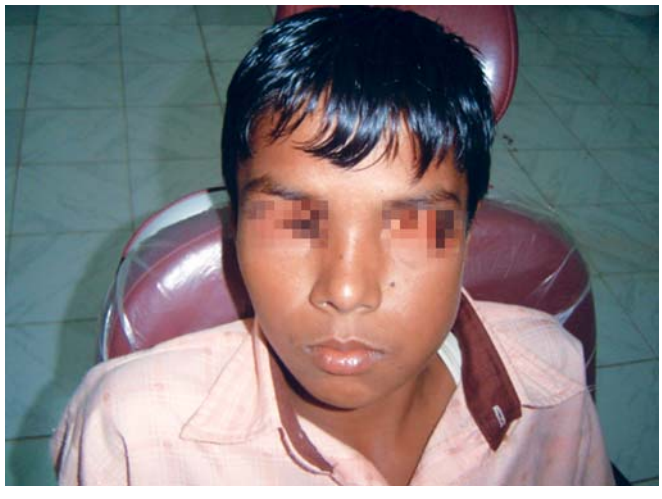

Fig. 1: Clinical photograph of the patient showing a diffuse swelling with left side of the mandible

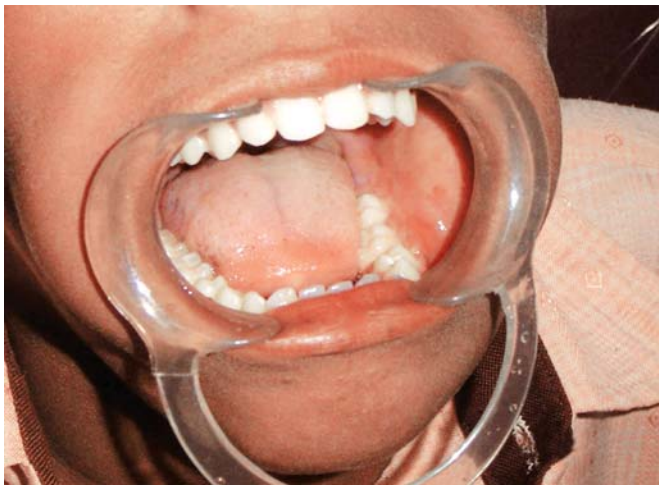

Fig. 2: Intraoral photograph showing swelling with left buccal vestibule and lingual tilt of 36 and 37

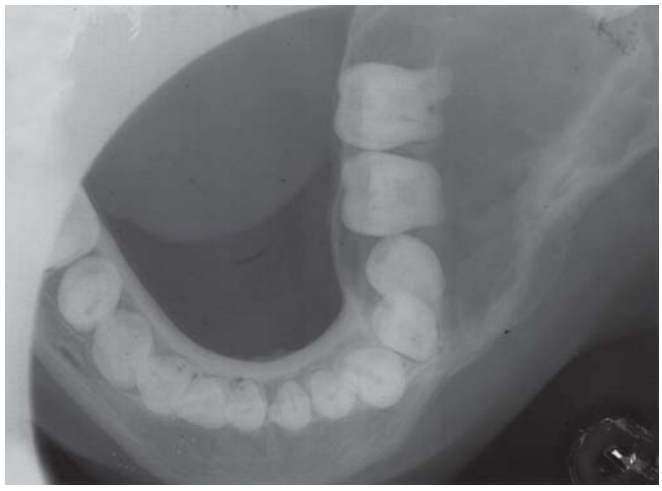

Fig. 3: Mandibular occlusal radiograph showing buccal cortical plate expansion and multilocular radiolucency with left side

region and obliterating the left upper and lower buccal vestibules (Fig. 2). On palpation the swelling was nontender and bony hard in consistency. The buccal cortical plate expansion was noted from left first premolar region to retromolar region. Overlying mucosa was normal in color with no fixity to the underlying swelling. There was no ulceration or pus discharge in relation to the swelling and it was noncompressible, nonfluctuant and nonpulsatile in nature.

Dental Findings

Teeth in the region of swelling, i.e. left mandibular first molar and second molar had a slight lingual tilt with grade I mobility. They were nontender on percussion. Other teeth showed no carious involvement nor were periodontally compromised. Occlusion on the right side was class I molar relation and left side occlusion was deranged due to the lingual tilt of mandibular first molar and second molar. All the third molars were clinically missing. Salivary gland examination was nonsignificant. Thus depending upon the history and clinical examination a provisional diagnosis of dentigerous cyst associated with impacted mandibular third molar was made. Odontogenic keratocyst, ameloblastoma, calcifying epithelial odontogenic tumor, odontogenic myxoma, ameloblastic fibroma and monostotic fibrous dysplasia were considered in the clinical differential diagnosis.

INVESTIGATIONS

Patient was advised the following investigations.

1. Complete hemogram was within normal limits except a decrease in Hb gm% to 11.4 gm% and a raised ESR to 88 mm after 1 hour.

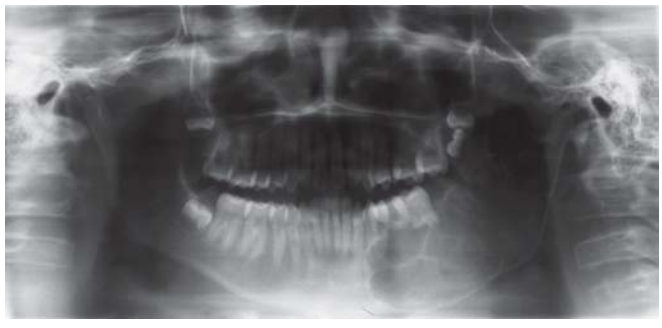

Fig. 4: Orthopantomogram showing multilocular radiolucency with left side of mandible involving ramus and body region giving a typical soap-bubble appearance

2. Random blood glucose and serum urea were within normal range.
3. Radiographs
   a. Mandibular occlusal radiograph (Fig. 3) revealed a buccal cortical plate expansion on left side extending from first premolar region till ramus region with irregular borders surrounding a multilocular radiolucency. Internal structure of lesion shows faint septa's extending into the radiolucency from the cortex. Localized expansion of lingual cortical plate is also evident adjacent to premolars on the left side. First molar and second molar are lingually displaced by the lesion.
   b. Orthopantomogram (Fig. 4) reveals a multilocular radiolucency in the left side of the mandible involving the ramus and body region extending from distal to first premolar and superiorly extending into the neck of condyle and coronoid process. Internal structure shows radiopaque septa's which are coarser in the ramus region and finer in the body region. These septa intermingle within this multilocular

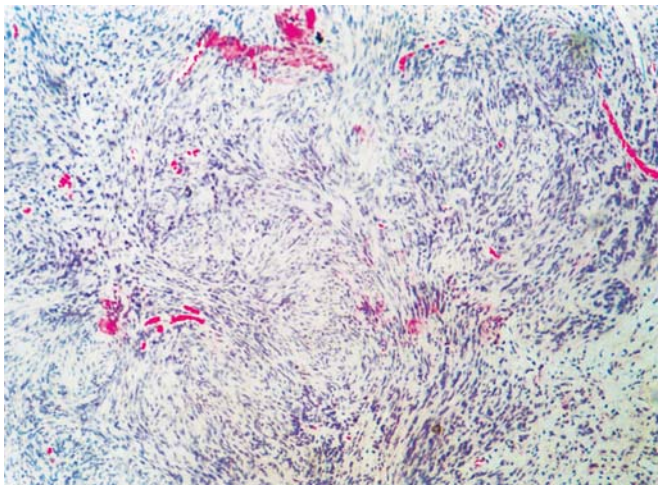

Fig. 5: Photomicrograph showing whorls of cells in nuclear palisading arrangement of Antoni A tissue and one area of less organized Antoni type B tissue (hematoxylin-eosin stain)

radiolucency giving a typical soap bubble appearance to the lesion. There is thinning of the lower border of the mandible. External root resorption is evident with first molar and second molar. The third molar is displaced posterosuperiorly into the coronoid process. Mandibular canal, mandibular foramen and mental foramen are not appreciable on the orthopantomogram on left side.

Radiographic features ruled out the dentigerous cyst, calcifying epithelial odontogenic tumor, monostotic fibrous dysplasia from the differential diagnosis list. But radiographic features were similar to that of odontogenic keratocyst, ameloblastoma and odontogenic myxoma.

4. Incisional biopsy was taken from the left retromolar region. The microscopic section (Fig. 5) showed, cells with elongated and wavy nuclei, which were arranged in palisading pattern giving a characteristic Antoni type A arrangement of neurilemmoma. The fibers in some planes gave the impression of occurring in whorls. Antoni type B tissue arrangement with disorderly arrangement of cells and fibers were also appreciated. Interspersed were blood vessels, Verocay bodies, small hyaline structures and areas of hyalinization and myxoid degeneration. Thus, the histopathological picture was consistent with neurilemmoma.

Final diagnosis of neurilemmoma involving the mandible was made and patient was advised surgical excision and regular follow-up.

DISCUSSION

Neurilemmoma was initially termed as schwannoma as it is thought to be derived from Schwann cells of the peripheral nerve sheath. Schwann cells derived their name from Theodor Schwann[5] (1880-1882), cofounder of cell theory. In 1908, Jose Verocay[5] provided first microscopic description of this tumor and named it as neurinoma. In 1935, Arthur Purdy Stout[4] described the histopathologic aspect of this disease and proposed the term neurilemma. In 1940, Isadore Max Tarlov[5] proposed these tumors to be of fibroblastic origin and coined the term peripheral fibroblastoma. Today most pathologists consider the term schwannoma, neurilemmoma, neurinoma and peripheral fibroblastoma to be synonymous.

There are three mechanisms by which neurilemmoma may involve bone: (1) a tumor may arise centrally with in the bone, (2) a tumor may arise with in the nutrient canal and produce canal enlargement and (3) a soft tissue or periosteal tumor may cause secondary erosion and penetration.[5]

The clinical presentation of the current case was largely atypical. The tumor presented as a painless swelling of the mandible with bony expansion in a 15-year-old male child in his second decade. Clinically, the central lesions are often asymptomatic but can cause bony expansion, pain and occasionally paresthesia. The radiographic appearance of central neurilemmomas is nonspecific.[3,4,6,7,9] They can mimic as many odontogenic and nonodontogenic lesions of the jaws. Unilocular radiolucency in relation to the mandibular canal is the usual manifestation. But this is noticed only when lesion is of a small size. Once the lesions increases to a large size with expansion of the cortex, a multilocular appearance is more evident.[6,7] Multilocular radiolucency mimicking soap bubble appearance was characteristic radiographic feature noticed in our case. Previously described radiological features include osteolysis with sclerotic borders, trabeculated contours, cortical erosions and occasional central calcification. Pathological fractures are rare. Erosion of the adjacent roots is a common finding and dystrophic calcification within the radiolucency has also been described.

Neurilemmoma has a few, if any defining features, consequently the differential diagnosis include odontogenic cysts and tumors, pseudocysts and intrabony nonodontogenic tumors.[3-5] Thus, in our case we initially thought it to be a dentigerous cyst depending on the history and clinical examination and radiographic features mimicked as of an odontogenic cyst but histopathologically it was diagnosed as a neurilemmoma. The microscopic picture of neurilemmomas is characteristic and can seldom be confused. The tumor is classically described as being composed of two types of tissues. Antoni type A is made up of cells with wavy nuclei aligned to form characteristic palisading pattern, while the intercellular fibers are arranged in parallel fashion between rows of nuclei. Some of them will be arranged in whorls or swirls. Antoni type B tissue

does not exhibit this characteristic palisading pattern, rather show a disorderly arrangement of cells and fibers with areas of edema fluid along with microcysts, Verocay bodies, and small hyaline structures. And most of the tumors will be encapsulated.[1,3,4]

In the present case, Antoni type A and Antoni type B cellular pattern were seen along with hyalinization and myxoid degeneration. Cells showed elongated and wavy nuclei and eosinophilic.

Treatment of neurilemmomas is complete surgical enucleation with periodic follow-up. Recurrence is rare but has been reported after incomplete excision.[3] Periodic recall is therefore recommended.

CONCLUSION

Rarities would be clinically extensive, radiographically destructive but histologically benign as reported in our case. Intraosseous neurilemmomas provide no definitive clinical or radiographic signs or symptoms that are diagnostic of the condition. They may resemble and mimic as other more common intraosseous conditions like cysts, ameloblastoma, odontogenic myxoma or any intraosseous nonodontogenic tumors.

Histopathology is the gold standard of diagnosis; yes this is very well revealed in present case, where even after thorough case history taking, cautiously doing clinical examination our provisional diagnosis was disapproved. Thus rarities are a must to be considered in provisional diagnosis.

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REFERENCES

1. Shafer WG, Hine MK, Levy BM. Benign and malignant tumors of the oral cavity. In: A textbook of oral pathology (4th ed). USA: A Prism Indian Edition of WB Saunders Company 1993:208-10.
2. Greenberg MS, Glick M. Benign tumors of the oral cavity. In: Burket's oral medicine diagnosis and treatment (10th ed). Hamilton: BC Decker Inc 2003:149.
3. Hietanen J, Mattila K, Calonius PEB, Ankkuriniemi O, Suonpaa J, Happonen RP. Central neurilemmomas of the mandible. Report of a Case. Int J Oral Surg 1984;13:166-71.
4. Sadeghi EM, Koenig LJ, Clark D. Intrabony neurilemmoma: Diagnosis and management. JADA 1998;129:729-32.
5. Chi AC, Carey J, Muller S, et al. Intraosseous schwannoma of the mandible. A case report and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2003;96:54-65.
6. Langlais RP, Langland OE, Nortje CJ. Circumscribed radiolucencies. In: Diagnostic imaging of the jaws: Baltimore, MD: Williams & Wilkins 1995:256-60.
7. White SC, Phroah MJ. Benign tumors of the jaws. In: Oral radiology principles and interpretation (4th ed). St Louis: Mosby 2000:405.
8. Gallego L, Junquera L, Rodríguez-Recio C, Fresno MF. Intraosseous mandibular schwannoma mimicking an odontogenic keratocyst, with a postsurgical pathological fracture. J Laryngol Otol 2009 May;123(5):560-62.
9. Buric N, Jovanovic G, Pesic Z, et al. Mandible schwannoma (neurilemmoma) presenting as periapical lesion. Dentomaxillofac Radiol 2009 Mar;38(3):178-81.

ABOUT THE AUTHORS

Raghavendra Byakodi (Corresponding Author)

Reader, Department of Oral Medicine and Radiology, Vasantdada Patil Dental College and Hospital, Kavalapur, Sangli, Maharashtra India, e-mail: raghubyakodi@hotmail.com

Vaishali Keluskar

Professor, Department of Oral Medicine and Radiology, KLE Society's VK Institute of Dental Sciences, Belgaum, Karnataka, India

Anjana Bagewadi

Professor and Head, Department of Oral Medicine and Radiology KLE Society's VK Institute of Dental Sciences, Belgaum, Karnataka India

Arvind Shetti

Professor, Department of Oral Medicine and Radiology, KLE Society's VK Institute of Dental Sciences, Belgaum, Karnataka, India