Case Report

Intraosseous Neurilemmoma of the Mandible: Report of a Rare Ancient Type

Gholamreza Jahanshahi[1], Abbas Haghighat[2], Faezeh Azmoodeh[3]

ABSTRACT

The neurilemmoma is a benign neoplasm of Schwann cell origin. One of the histopathologic subtypes of this tumor is ancient schwannoma which is characterized by degenerative alterations including cystic change, calcification, hemorrhage, and hyalinization.

Intraosseous schwannomas especially ancient ones are rare tumors. Here we present a case of intraosseous ancient schwannoma in the lower jaw of an 11-year-old girl which caused a non-tender expansion. Radiographic examination showed a well-circumscribed, unilocular radiolucent lesion with thin sclerotic borders in the mandibular body and the ramus. Histopathologic examination of the incisional biopsy showed areas of typical Antoni A with verocay bodies and Antoni B that was strongly suggestive of a schwannoma. Complete excision of the lesion was done under general anesthesia. The histopathologic examination confirmed the primary diagnosis and also degenerative changes such as hyalinization and calcification. Based on these findings, the diagnosis of ancient schwannoma was made. No recurrence was observed in the follow-up examination after 3 months.

***Keywords*:** Ancient schwannoma, Intraosseous neurilemmoma, Mandible.

Received: *October 2010*
Accepted: *January 2011*

Dent Res J 2011; 8(3): 150-153

Introduction

The neurilemmoma (Schwannoma) is a slow-growing, benign neoplasm of Schwann cell origin, that typically arises in association with nerve trunks.[1-5] These tumors most commonly arise in the soft tissues of the head and neck, as well as on the flexor surfaces of the upper and lower extremities.[4, 6, 7]. Intraoral lesions are not frequent and the tongue is the most common site of oral neurilemmomas. Intraosseous schwannomas are rare (44 true reported cases up to now) but when they occur, the mandible is the most common site, particularly in the posterior areas of the body and ramus.[5, 8]

Several histological variants of schwannoma, including the cellular, plexiform, epithelioid, ancient, and melanotic types have been described.[4, 9]

Ancient schwannomas tend to be large tumors of long duration and are characterized by degenerative alterations including cystic change, calcification, hemorrhage, and hyalinization. Macrophages and hemosiderin deposits are common. Although nuclear atypia may be striking, mitotic activity is absent.[3, 4, 9] However, only eleven cases of ancient schwannoma occurred in the oral cavity have been reported in the English literature that none of them were Intraosseous.[9]

The recommended treatment for schwannoma is surgical enucleation with periodic follow up; and recurrence is uncommon because of encapsulation.[4, 8] In this article, we report a case of an intraosseous ancient schwannoma located in the mandibular ramus.

Case Report

An 11-year-old girl referred to a head and neck

[1] Associate Professor, Department of Oral & Maxillofacial Pathology and Torabinejad Dental Research Center, School of Dentistry, Isfahan University of Medical Sciences, Isfahan, Iran.
[2] Assistant Professor, Department of Oral & Maxillofacial Surgery Torabinejad Dental Research Center, School of Dentistry, Isfahan University of Medical Sciences, Isfahan, Iran.
[3] Postgraduate Student, Department of Oral & Maxillofacial Pathology, School of Dentistry, Isfahan University of Medical Sciences, Isfahan, Iran.
Correspondence to: Gholamreza Jahanshahi, Email: Jahanshahi@dnt.mui.ac.ir

surgeon with a chief complaint of a swelling in her lower jaw with two month duration. The patient reported no history of pain and paresthesia.

The clinical examination revealed a right mandibular expansion without tenderness (Figure 1a). On panoramic radiograph, there was a well-circumscribed, unilocular radiolucent lesion with thin sclerotic borders extending from the distal aspect of impacted right second molar to the ramus (Figure 1b).

The differential diagnosis included odontogenic cysts (especially odontogenic keratocyst), odontogenic tumors (such as unicystic ameloblastoma and ameloblastic fibroma), non odontogenic central tumors (such as central giant cell granuloma), traumatic bone cyst and aneurysmal bone cyst. Because neurilemmoma of the jaw is a rare entity, it was not considered in our differential diagnosis list.

A CT-scan was performed. CT images showed a solitary well-defined central mass in the right mandibular ramus with 52 mm width × 39.8 mm length in dimension. The lingual cortex seemed to be eroded (Figure 1c).

An incisional biopsy was obtained under local anesthesia. Histopathologic examination of the H&E stained section of the incisional biopsy showed areas of Antoni A tissue including bundles of spindle shaped cells with long basophilic nuclei palisaded around an acellular eosinophilic areas known as Verocay bodies. Antoni B tissue with randomly arranged spindle cells, within a loose myxomatous stroma was also seen (Figure 2a).

Therefore a preliminary diagnosis of neurilemmoma was made. Subsequently the patient underwent total removal of the tumor under general anesthesia. Initially it was decided to resect the involved area of the mandible and reconstruct the region with bone plate. Using an extraoral incision in the right retromandibular area, the muscle dissection and exposure of the involved bone was done. Following osteotomy in the second molar zone, the detached segment was turned laterally around the head of condyle. The medial part of the ramus was widely destructed up to the pharynx and the base of skull. The mass found to be encapsulated, so it was separated from the medial portion of the ramus by finger dissection. The left inferior alveolar nerve was sacrificed due to its adhesion to the lesion. Despite lingual cortex destruction and buccal perforations of the ramus, the separated segment was returned to its place in order to maintain bone integrity and fixed by two miniplates. Interdental wiring was also done for three weeks.

Grossly, the specimen was lobulated & encapsulated mass, approximately 5.5cm length × 5cm width in dimension, with solid tan to grayish cut surfaces (Figure 2b). Histopathologic examination confirmed the primary diagnosis. In addition, degenerative changes such as hyalinization, calcification, hemorrhage and mild pleomorphism were also seen (Figure 2c). With all these features, the diagnosis of ancient schwannoma was made.

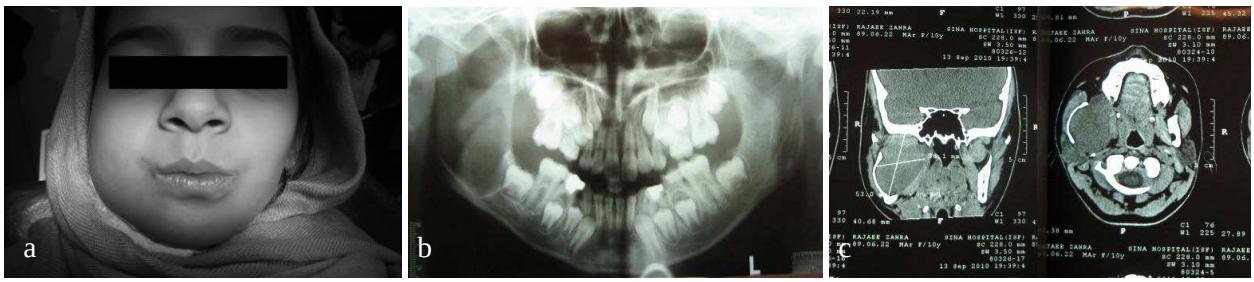

Figure 1. (a) large expansile mass of the right side. (b) Panoramic radiograph showing a well-defined unilocular radiolucent area in the right mandibular ramus. (c) Solitary well-defined expansile mass with cortical erosion.

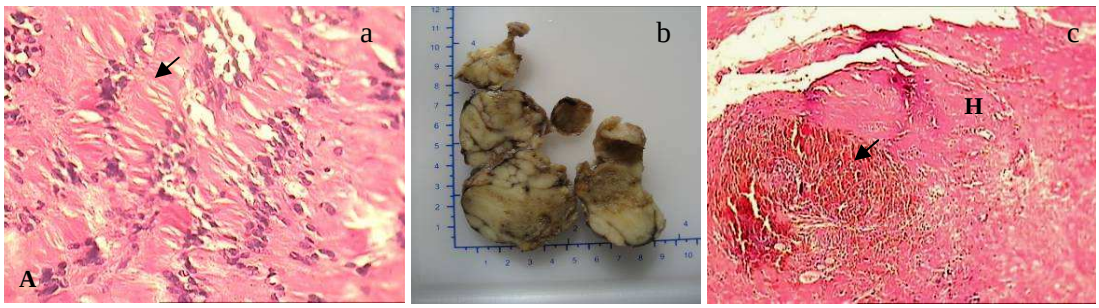

Figure 2. (a) Areas of Antoni A tissue (A) and Verocay bodies (arrow), H&E stain, magnification x100. (b) Gross specimen of the tumor. (c) Areas of hemorrhage (arrow) & Hyalinization (H), H&E stain, magnification x40.

The patient was followed up for three months without any evidence of infection, wound dehiscence or recurrence (Figure 3).

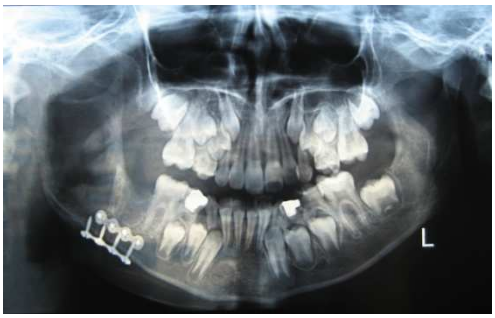

Figure 3. Panoramic radiograph 3 months after surgery.

Discussion

The neurilemmoma is a benign neoplasm of Schwann cell. These tumors commonly arise in the head and neck soft tissue, but intraoral lesions are unusual, especially centrally in the jaws.[1-5]

In the review of the literature, Chi et al.[4] reported 44 intraosseous schwannomas of the jaws including 39 mandibular and 5 maxillary tumors. Most of mandibular tumors were placed in the posterior areas of the body and ramus. It can be attributed to the long intraosseous pathway of the inferior alveolar nerve within the mandible.[5, 7, 8] The average age was 34 years, with peak prevalence in the second and third decades of life,[4] however our case was an 11-year-old girl which was younger than the mean age.

Intraosseous schwannoma is a slow–growing tumor that frequently produces expansion of the affected bone and causes swelling. Pain and paresthesia may be found in 50% of the patients.[8, 10] Our patient had a large swelling with no pain or paresthesia. She reported 2 months duration, but based on the tumors large size and histopathologic degenerative changes, true duration of the lesion seems to be more.

Radiographically, Intraosseous schwannoma is a well-circumscribed unilocular radiolucency, with thin sclerotic borders commonly found in the posterior areas of mandible that can be like the other benign cystic lesion such as OKC or a benign tumor, like ameloblastoma.[5, 11] In the present case, the lingual cortex was eroded which is not a common feature (reported in less than 11% of cases).[4]

Microscopically, the features were found to be characteristic of neurilemmomas including encapsulation, Verocay bodies, palisading nuclei, and Antoni A and B tissues. These tumors´ cells typically showed a diffuse positive immunoreactivity for S-100 protein.[1, 3]

In addition to the classic schwannoma, there are several histopathologic variants, including the cellular, plexiform, epithelioid, ancient, and melanotic types.[12]

Ackerman and Taylor[13] first found the schwannoma presented with clear areas of hypocellular tissues and attributed the changes to the long standing degenerative changes.

Ancient schwannomas tend to be large tumors of long duration and are characterized by degenerative alterations including cystic changes, calcification, hemorrhage, and hyalinization. Macrophages and hemosiderin deposits are common. Although nuclear atypia may be striking, mitotic activity is absent.[4,9] In this case, histopathologic examination showed areas of considerable hyalinization, hemorrhage, focal calcification and mild pleomorphism.

Reviewing the literature, only eleven cases of ancient schwannoma had been reported that were placed in the anterior portion of tongue, palate, mandibular and maxillary vestibule region, floor of the mouth and buccal mucosa.[9, 12] The most common site of this tumor among the cases seen in the literature was floor of the mouth, with mean age of 30.5 years and female sex predilection.[9]

Salehinezhad and colleagues[14] in 2009 reported the first case of central ancient schwannoma in a 23-year-old woman in Iran. So this case might be the second one.

Conclusion

Schwannoma treatment consists of surgical enucleation with periodic follow-up examinations. Recurrence is uncommon and in this case, the patient was followed up for three months with no clinical or radiographic signs of recurrence.

References

1. Neville B, Damm DD, Allen CM, Bouquot J. Oral and Maxillofacial Pathology. 3rd ed. Philadelphia: Saunders; 2008. p. 526-7.
2. Regezi JA, Sciubba JJ, Jordan RCK. Oral Pathology: Clinical Pathologic Correlations. 5th ed. Philadelphia: Saunders; 2007. p. 170.
3. Gnepp DR. Diagnostic Surgical Pathology of the Head and Neck: Expert Consult - Online and Print. 2nd ed. Philadelphia: Saunders; 2009. p. 245-6.
4. Chi AC, Carey J, Muller S. Intraosseous schwannoma of the mandible: a case report and review of the

literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2003; 96(1): 54-65.
5. Hsieh MY, Chen CM, Lin LM, Chen YK. Intraosseous Neurilemmoma (Schwannoma) of the Mandible - Report of a Case. Cases Journal Taomfr 2009; 1(2):1-6.
6. de Lacerda SA, Brentegani LG, Rosa AL, Vespucio MV, Salata LA. Intraosseous schwannoma of mandibular symphysis: case report. Braz Dent J 2006; 17(3): 255-8.
7. Gallego L, Junquera L, Rodriguez-Recio C, Fresno MF. Intraosseous mandibular schwannoma mimicking an odontogenic keratocyst, with a postsurgical pathological fracture. J Laryngol Otol 2009; 123(5): 560-2.
8. Nakasato T, Katoh K, Ehara S, Tamakawa Y, Hoshino M, Izumizawa M, et al. Intraosseous neurilemmoma of the mandible. AJNR Am J Neuroradiol 2000; 21(10): 1945-7.
9. Amirchaghmaghi A, Salehinejad J, Basirat M, Delavarian Z, Javadzade A, Forouzanfar A. Gingival Ancient Schwannoma: Review of Literature and a Case Report. Journal of Applied Sciences 2010; 10(23): 3137-40.
10. Ito S, Mandai T, Ishida K, Deguchi H, Hata T, Irei I, et al. Intraosseous schwannoma of the mandible: A case report. Kawasaki medical journal 2009; 35(3): 249-52.
11. Jang KY, Moon WS, Park HS. Intraosseous Neurilemmoma of the Mandible - A Case Report. The Korean Journal of Pathology 2009; 43(1): 88-91.
12. Subhashraj K, Balanand S, Pajaniammalle S. Ancient schwannoma arising from mental nerve. A case report and review. Med Oral Patol Oral Cir Bucal 2009; 14(1): E12-4.
13. Ackermann LV, Taylor FH. Neurogenous tumors within the thorax. A clinicopathological evaluation of 48 cases. Cancer 1951; 4: 869.
14. Salehinezhad J, Babazadeh F, Saghafi Sh, Zare M, Abadi R, Rajaei AR. Intra-Osseous Degenerated Neurilemmoma of the Mandible in a 23 Year-Old Woman. Journal of Mashhad Dental School 2010; 33(4): 253-360.