

Intramuscular Schwannoma Mimicking a Parotid Tumor: Case Report and Review of Literature

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Case report

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Abstract

Background

Schwannoma is a benign neoplasm originating from Schwann cells of peripheral nerves as well as the cranial and spinal roots. It often occurred in head and neck region involving cranial nerves VII, VIII, and XII. However, schwannoma located in intramasseteric region is extremely uncommon. There have been only ten reported cases documented in English literatures to date.

Case presentation

Here, we report a rare intramasseteric schwannoma case, who presented a painless mass in the left cheek with 2 years evolution and was treated with surgical excision through trans-parotid approach. An uneventful recovery without salivary fistula or facial paralysis was recorded after the surgery. Due to its rarity and clinical significance, we also review intramasseteric schwannoma regarding its clinical characteristics and surgical managements.

Conclusion

In a word, intramasseteric schwannoma is a benign lesion with favorable prognosis. MRI (magnetic resonance imaging) scanning is recommended for evaluation and differential diagnoses for this condition. Surgical excision of the tumor without parotidectomy is considered to be effective.

Background

Schwannoma is a slow growing, encapsulated tumor representing about 5% of benign soft tissue tumor (1). It was firstly described by Stout as a benign neurogenic tumor with sporadic malignant degeneration (2). Schwannoma could be originated either from cranial, peripheral or from autonomic nerve sheath cells. The signs and symptoms of this condition in different patients varied because their origin nerve and involved sites are diverse (1). The most common involved area for schwannoma is the head and neck region, which accounts for 25–45% of the whole body schwannoma (3). Schwannoma in head and neck region could arise either medially from glossopharyngeal, vagus, accessory and hypoglossal nerve, the sympathetic chain, or laterally from cutaneous or muscular branches of the cervical/brachial plexus and so on (4). It has been intensively reported with respect to the clinical, pathological and immunohistochemical characteristics of schwannoma in head and neck region in literature (3, 5–9). However, intramasseteric schwannomas was rarely reported and might be misdiagnosed as parotid tumors. To the best of our knowledge, there have only ten cases with intramasseteric schwannoma been reported in English literatures. Here, we update an uncommon case of schwannoma located in the intramasseteric muscle and describe its clinical findings. In addition, we also performe an extensive retrospective literature review of schwannoma presenting in the region of intramasseteric muscle to evaluate the clinical findings and treatments.

Case Presentation

A 15-year-old boy presenting with a painless slow-enlarging mass in his left cheek was referred to the Department of Oral and Maxillofacial Surgery, Hospital of Stomatology, Guanghua School of Stomatology, Sun Yat-sen University, China. There was no numb feeling during the 2 years evolution. No significant family history was noted. An asymmetry profile in the maxillofacial region was noted while performing facial examination. The mass was measured approximately 4.0 × 3.5 cm in diameter (Fig. 1A) on the left cheek. It was noted that the lump was palpated with poor mobility, hard elastic texture and smooth contour. The overlying skin was intact and discolored (Fig. 1A). No facial paralysis or cervical lymphadenopathy was detected. Intraorally, the buccal and oropharyngeal mucosa was normal. Computed tomography (CT) scanning showed that a well-circumscribed mass was located in parotidomasseteric region, which was hypodense to surrounding soft tissue. No involvement of bony destruction was found in the adjacent ramus of mandible (Fig. 1B). Based on these examinations, the initial impression was a tumor of parotid gland. Under general anesthesia, surgical removal of the tumor was carried out with a standard preauricular/cervical incision. However, we found that the tumor was located at the inside of muscoli masseter, superficial to the ramus of mandible (Fig. 1C, D). The tumor was then completely removed and subjected to pathological examination. The parotid gland with facial nerve was preserved, repositioned and wound was closed layer by layer (Fig. 1E, F). Macroscopically, the resected specimen was a well circumscribed mass with elastic texture (Fig. 2A). On sectioning, the mass demonstrated a white soft tissue with cystic degeneration and haemorrhage (Figs. 2B). The preliminary pathology report from frozen-section specimen was schwannoma, which was confirmed by examination of the tumor in paraffin section. As shown in Fig. 2C, the tumor was composed of well-organized Antoni A tissue with adjacent myxoid and less organized Antoni B tissue. The Schwann cells of the Antoni A tissue form a palisaded arrangement around acellular, eosinophilic zones known as Verocay bodies (Figs. 2D). The postoperative recover was uneventful. During the follow up period, there was no salivary fistula or facial paralysis occurred.

Discussion And Conclusions

A review of the English-language literature reveals only 10 reported cases of schwannomas located at the intramasseteric region(10–19). As shown in Table 1, a female-to-male ratio of 1:1 was observed and there is no significant gender predilection. The patient age was ranged from 12 to 61 years. The average size of tumor was 1.45 cm, with a range of 2 to 4 cm. All lesions were described as a painless and slow-growing mass. However, the mass showed various characteristics on palpation, including soft or hard elastic consistency, well or poor mobility. Notably, all fine-needle aspiration (FNA) biopsies were inconclusive but the final diagnosis can only achieved based on pathological examination. All of the ten reported tumors were surgically removed, among which seven cases were recorded with follow-up information. There were no recurrences observed within the follow-up period. Some of the lesions (6/10) were comprised with both Antoni A and B components. Two patients were diagnosed as Antoni A type schwannoma and only one was classified as Antoni B type schwannoma. The histological pattern of the case reported by Schreiber et al(13) was not available.

Table 1
Case reports of schwannoma in the intramasseteric region

Author, Year	Age(Yr), Gender	Symptom, History	Clinical manifestation	Tumor size(mm)	Imaging examination	Biopsy	Treatment	Pathc
Ishikawa et al, 1991	61, Female	Subcutaneous nodules with sudden growth for 3 months. Hemicrania presented.	The tumor was mobile, elastic, soft, and fluctuant; however, no fluid could be aspirated	Multiple foci One lump with 30 × 30 One lump with 15 × 25 The other were not described	No abnormal finding in sialography. CT suggested a well defined but heterogeneous tumor involved the right pterygopalatine fossa.	Biopsy was performed intraorally, supporting the diagnosis of schwannoma	Excision with modified preauricular incision and without parotidomandibulectomy	Anton and B
Nakamura et al, 2006	12, Male	a painless left cheek mass for 1 year	The tumor was elastic-hard, painless, non-tender and poorly Mobile.	20 × 10	CT revealed a well-circumscribed and heterogeneously enhanced tumor. MRI showed isointensity (T1) and high signal intensity (T2)	No	Excision	Anton and B
He et al, 2010	Middle aged female	a painless right cheek mass for 3 years	A soft non-tender, poorly mobile mass	40 × 30 × 20	CT scan showed a well-circumscribed hypodense mass within the masseter muscle	FNA inconclusive	Excision with submandibular incision	Anton and B
Schreiber et al, 2011	35, Male	an indolent, slow-growing mass in the right parotid-masseteric region for 1 year	A hard elastic consistency on palpation	25 × 28 × 26	MRI showed moderately hyperintense with a very hyperintense central portion. Peripheral enhancement with nonenhancing central (cystic) portion after contrast agent administration.	ultrasonography-guided FNA inconclusive	Tumor excision and superficial parotidectomy with preservation of the facial nerve.	Undes
Wang et al, 2016	33, Male	a slowly enlarging painless swelling in the left preauricular region for more than 1 year	A smooth contour, elastic, and well-demarcated mass located in the left preauricular region	Two foci with 36 × 28 and 20 × 11 mm respectively	In CT scanning, the two hypodense masses showed homogeneous, and well-defined. Ultrasound scanning revealed two low echo masses.	No	Excision	Anton

Author, Year	Age(Yr), Gender	Symptom, History	Clinical manifestation	Tumor size(mm)	Imaging examination	Biopsy	Treatment	Pathc
Endo et al, 2017	50, Female	a nodule that had been on her left cheek for 5 years	a soft, well contoured, round mass was palpated in the left cheek	25 × 20 mm	CT revealed a well-circumscribed tumor. MRI (T1) showed the tumor was isointense to the masseter muscle and hypointense to the parotid gland, however it was hyperintense to both in T2 images	No	Tumor excision with Endoscope-assisted Transoral Approach	Anton B
Khatib et al. 2018	30, Male	painless cheek mass	A soft, mobile and non-tender right facial mass	40 × 35 × 20	CT showed a 3.5 × 2.5 cm ring enhancing, necrotic lesion involving the masseter muscle without definite intra-parotid extension.	Fine needle aspiration (FNA) showed parotid benign acini and scanty cellular fluid, which are nonspecific but may represent a hypertrophic process involving the parotid or a cystic mass in the proximity of the parotid.	Tumor excision Intraoperatively the parotid gland was completely normal.	Anton A and
Aizawa, et al. 2019	37, Male	a painless mass in the right cheek region that had been slowly enlarging for 3 years	he had a firm, well-demarcated, nonpulsatile mass with poor mobility	20 × 20	intermediate signal intensity on T1WI and high signal intensity on T2WI	Fine needle aspiration cytology only detected blood cells. Incisional biopsy was scheduled, taking the possibility of malignancy into consideration	A 2.5 cm linear skin incision was made directly above the lesion. Total excision was performed with a narrow margin.	Anton A and
Matsumine et al. 2019	24, Female	gradually enlarging initially asymptomatic mass in the left parotidomasseteric region 1 year before	A soft, painless mass near the angle of the left mandible, and the mass was poorly mobile.	3.2 × 1.7 × 1.6 cm	MRI revealed an isolated neoplastic lesion within the body of the masseter muscle; low-signal intensity on coronal T1-weighted imaging and high-signal intensity on T2-weighted imaging.	No	Excision with trans-parotid approach. The tumor was enclosed within the masseter muscle, and continuous with a branch of the masseter nerve identified using intraoperative electrical stimulation.	Anton A
Hwang et al. 2019	42, Female	slowly growing, painless mass on her left cheek with 9 year evolution	hardly movable, and non-tender mass	2.8× 2.8 × 1.8 cm	In CT scan, a 2.8× 2.8 × 1.8 cm sized tumor was detected in the left masseteric muscle. It was hypodense compared to muscle and was heterogeneously enhanced.	No	A facelift incision with pretragal and occipital incisions was chosen over a modified Blair incision. The gland was elevated from the masseter muscle and branches of the facial nerve were retrogradely dissected and preserved.	Anton A and
The current case	15, Male	a painless left cheek mass for		30 × 35	CT	No	Excision with trans-parotid approach	Anton A and

FNA: fine-needle aspiration biopsy

Schwannoma, also known as neurilemmomas or neurilemmomas, was originated from schwann cells. It was reported that schwannoma account for as rare as 5% of all benign soft tissue tumors, and the malignant schwannoma represent only 5% of all soft tissue sarcomas (1). Head and neck region is the most common site affected by the extracranial schwannoma (20). However, intramasseteric schwannomas are extremely uncommon. Currently, there were only 10 cases with intramasseteric schwannoma reported in English literatures, indicating that intramasseteric region is an extremely rare site for schwannoma (Table 1) (10–14). The epidemiological characteristics of schwannoma in head and neck region were in agreement with those in other regions of the body, which was prone to affect patients in their second and third decade and an equal gender distribution was observed. [21–24]. Similar results were also observed in these case series of intra-masseteric schwannoma.

Clinically, intramasseteric schwannomas might be misdiagnosed as the tumor originating from parotid therefore it should be differentiated from parotid tumors. The differential diagnosis of these two kinds of tumors is even more tricky when the intra-masseteric schwannoma become soft due to their mucinous degeneration, bleeding and cystic changes [20]. It's still a great challenge to diagnose this palpable mass preoperatively for a junior head and neck surgeon (1). Medical history, physical examination, fine needle aspiration (FNA), ultrasound scanning, computed tomography (CT), magnetic resonance imaging (MRI) and positron emission tomography (PET) are used as diagnostic modalities. However definitive diagnosis and identification of the affected nerve are often difficult before surgery. (21, 22). The most common presentation of schwannoma is an asymptomatic slow-growing mass, which is similar with benign tumor of parotid gland. Generally, CT, MRI and FNA might provide helpful information in diagnosis of parotid tumors. As shown in Table 1, all cases received CT or MRI scanning before operation. Both of this two imaging examination can provide useful information for differential diagnosis and development of surgical plan. On CT scanning, schwannoma typically demonstrates a heterogeneous low density with only limited soft tissue definition. MRI imaging, in which schwannoma demonstrates isointense T1 signal relative to skeletal muscle and increased heterogeneous T2 signals, is preferred for visualizing soft tissue neoplasms.. The ultrasound scanning is another optional for preoperative imaging since vessels and blood flow within the tumor could be detected [16]. However, These radiographic findings provide good but often limited information in the diagnosis of schwannomas (23). As evident in Table 1, FNA biopsies were performed in 4 cases and all inconclusive, indicating that FNA has a low accuracy in the diagnosis of schwannoma. Therefore, the conclusive diagnosis is only achieved based on microscopic findings of the harvested tissues, characterizing with a specific cellular Antoni A areas and less organized hypocellular Antoni B areas [16–20].

It was suggested that both surgical excision and observational approach are applicable in management of schwannomas in head and neck region due to the noninvasive nature (24, 25). Intracapsular enucleation and resection are both recommended to manage schwannoma in head and neck region. During the operation, resection of tumor was performed if the lesion was originated from the terminal branch of trigeminal nerve and lack of postoperative neurological deficit. However, intracapsular enucleation was highly recommended when the tumor involved the important nerve, such as facial nerve or trunk of trigeminal nerve. For intramasseteric schwannoma, it is difficult to identify the original nerve of the tumor since the tumor always obliterate a small nerve in masseter muscle. All reported cases were treated with tumor excision through different approach, including extraoral or intraoral approach, which was mainly depended on the location and involvement of tumor (15, 17, 19). An extraoral incision such as a preauricular incision or a submandibular incision is often unavoidable for the intramasseteric schwannoma. Intraoral approach accompanied with endoscope is feasible for tumor removal when the tumor size is limited (15). Similar with the surgical approach for parotid tumor, facelift incision was also used to excise the intramasseteric schwannoma (19). Superficial or total parotidectomy is not required for management of intramasseteric schwannomas. In the present case, the tumor was excised with transparotid approach. The parotid gland and facial nerve were preserved. The prognosis of schwannoma is usually outstanding as reported in literatures [21]. No recurrence was seen in any cases till date in the present study.

Taken together, intramasseteric schwannoma is a benign lesion with favorable prognosis. MRI scanning is recommended for evaluation and differential diagnoses of schwannoma preoperatively, but FNA biopsy is always inconclusive and not necessary. For treatment of these lesions, surgical excision of the tumor is considered to be effective, which neighboring structures and major nerve should be protected and preserved. Superficial or total parotidectomy is not required for managing intramasseteric schwannomas.

Abbreviations

MRI magnetic resonance imaging

CT computed tomography

FNA fine-needle aspiration

PET positron emission tomography

Declarations

Ethics approval and consent to participate

The present research was approved by Ethical Review Committee, Guanghua School of Stomatology, Hospital of Stomatology, Institute of Stomatological Research, Sun Yat-set University. The ethical approval letter is as attached.

Consent for publication

The consent for publication was obtained from the patient as attached.

Availability of data and materials

Not applicable.

Competing interests

The authors declare that they have no competing interests.

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Authors' contributions

Wei-xin Cai: Wrote the manuscript and assisted in surgery.

Nan Xie: Perform pathological examination and helped in preparation of manuscript.

Jing Hu: Assisted in the surgery and reviewed the literature.

Cheng Wang: Designed the study, carried out the surgery, prepared and revised the manuscript.

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Figures

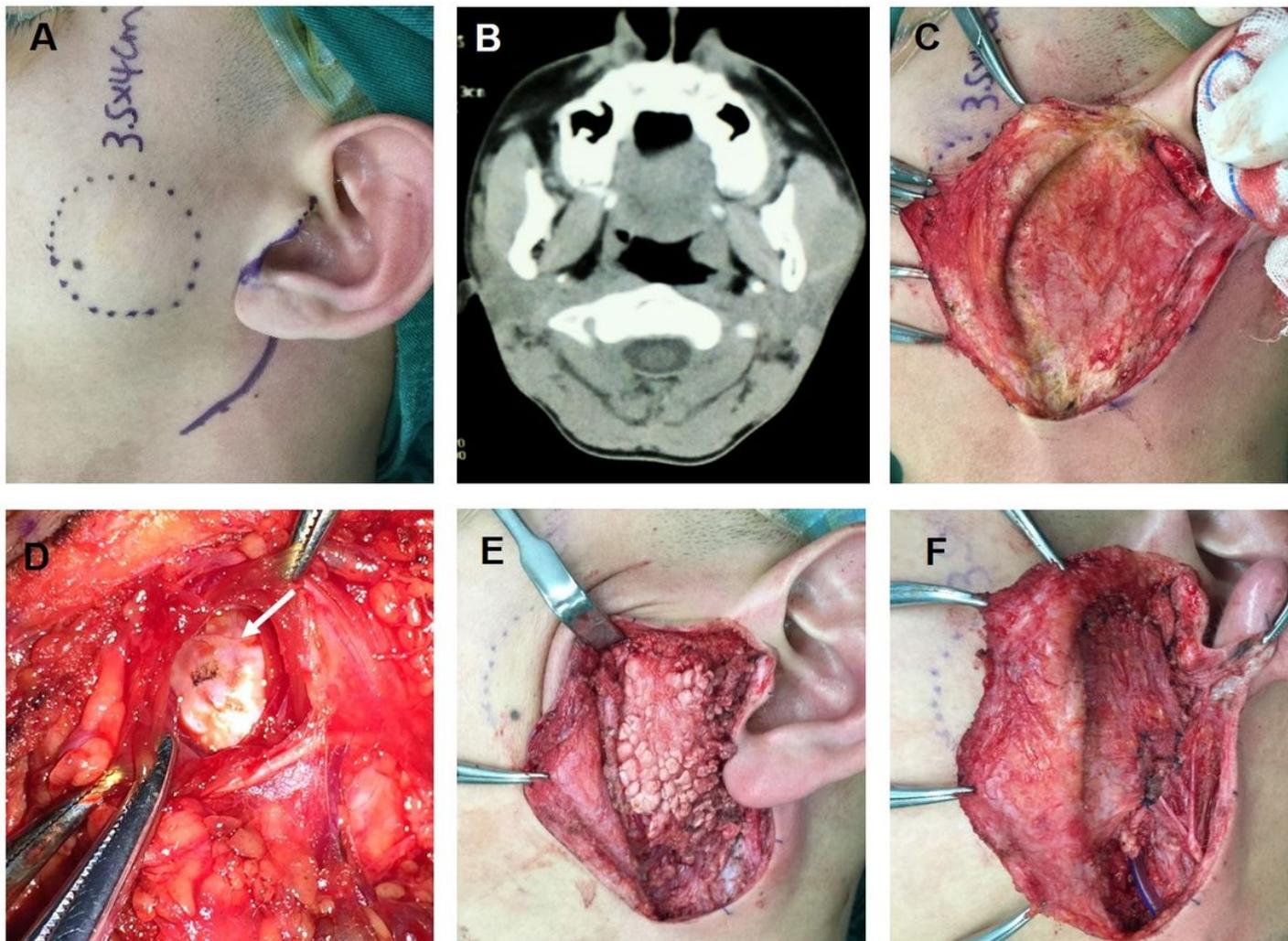


Figure 1

(A) Preoperative image of the tumor. (B) CT scanning showed a well-circumscribed mass. (C,D) Intraoperative image showed the tumor located within the musculi masseter. *indicated tumor, yellow arrow indicated facial nerve and black arrow indicated the masseteric muscle. (E) Repositioning of the parotid gland and parotidomasseteric fascia intraoperatively. (F) Closure of the operative site layer by layer.

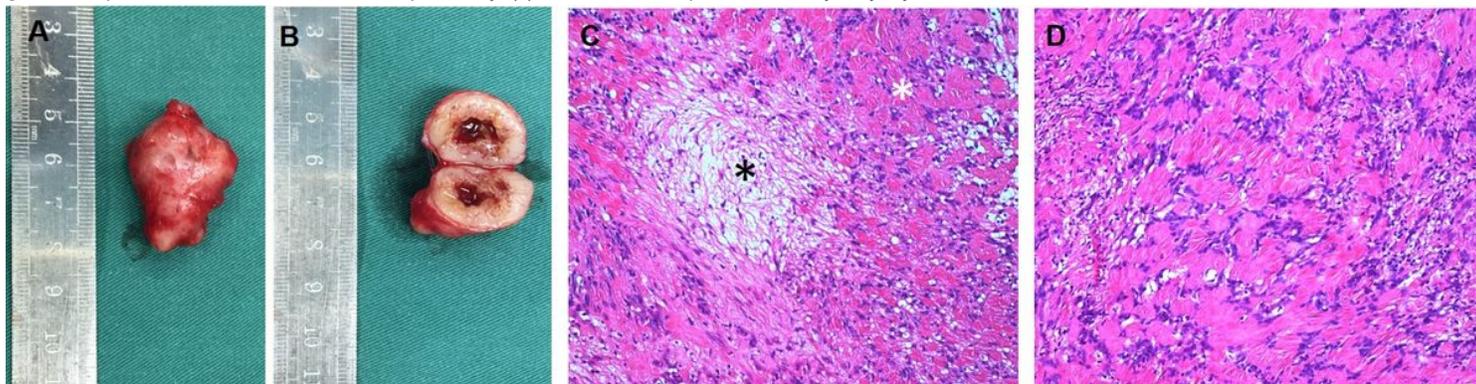


Figure 2

(A) Macroscopic view showed the intact mass separated from the masseteric. (B) On sectioning, the mass demonstrated a white soft tissue with cystic degeneration and haemorrhag. (C) Microscopic examination showed that the tumor was composed of well-organized Antoni A tissue with adjacent myxoid and less organized Antoni B tissue (Black asterisk: Antoni B tissue, white asterisk: Antoni A tissue). (D) The Schwann cells of the Antoni A tissue form a palisaded arrangement around acellular, eosinophilic zones known as Verocay bodies.