[症例報告]Neurilemmoma of the upper lip : literature review

Neurilemmoma of the upper lip : literature review

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(Received on February 13, 2003, accepted on May 26, 2003)

ABSTRACT

Neurilemmoma occurs in all parts of the body, but occurrence in the oral and maxillofacial regions is relatively rare. In the oral and maxillofacial region, tumors usually arise on the tongue and rarely on the lip. The upper lip especially is a rare site of occurrence. Recently, we treated a case of neurilemmoma located in the upper lip. The patient, a 48-year-old woman, visited our hospital with a complaint of a mass formation on the left side of the upper lip. Excision of the tumor was done under local anesthesia. Up on examination under the light microscopy, it was found to be Antoni A type neurilemmoma. In addition, immunohistochemical study using S-100 protein antibody, showed a positive reactivity in the tumor cell. There was no evidence of recurrence 6 months after the operation. *Ryukyu Med. J., 22(1,2) 59～62, 2003*

Key words: Neurilemmoma, upper lip

INTRODUCTION

Neurilemmoma is a benign tumor arising from Schwann cells. In the oral and maxillofacial region, tumors usually arise on the tongue and rarely on the lip. A literature search revealed that 5 cases of neurilemmoma of the upper lip have been previously been reported in Japan[1,2]. We present a case of neurilemmoma of the left side of the upper lip.

CASE REPORTA

48-year-old Japanese woman was referred to the Oral and Maxillofacial Surgery at the University of Ryukyus Hospital for the evaluation of a painless mass on the upper lip. She had noticed the mass about 2 months prior but had not sought medical treatment because there were no symptoms.

Oral examination disclosed a painless mass of the upper lip measuring 10 × 10 mm. The mass was lobular with an elastic consistency. The overlying mucosal surface appeared normal in color and texture (Fig. 1). The clinical impression was a salivary gland tumor. An examination of the head and neck was otherwise normal.

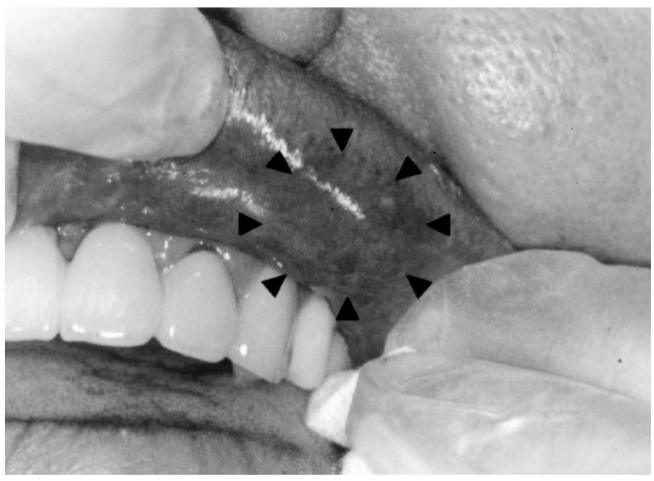

Fig. 1 Nobule on upper lip. Approximatery 1 cm in diameter.

Ultrasonography (US) was performed with a linear scanner 5.0 Mhz (RT-2800: Yokogawa Medical, Tokyo). Imaging showed a heterogeneous mass with hypoechoic features, well-delineated margins,

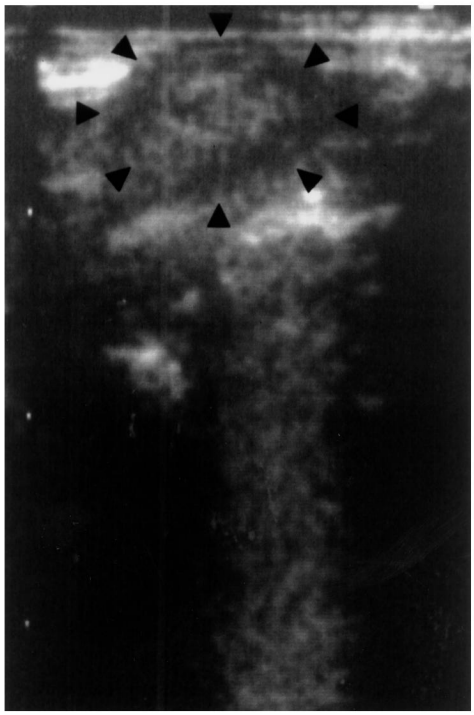

Fig. 2 Ultrasonography showsing a heterogeneous mass with hypoechoic features, well-delineated margins, and an enhancement of posterior echo.

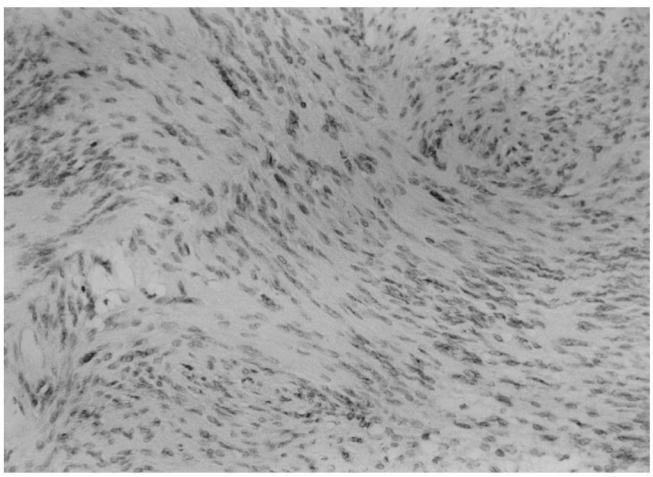

Fig. 3 Tumor cells aligned in places to form a characteristic palisading patten (Hematoxyline eosin stain. Original magnification ×200).

and an enhancement of the posterior echo (Fig. 2).

Excision was performed under local anesthesia. The excised tissue was friable and of a grayish yellow color. Sutures were removed 1 week after the procedure. No post operative complications occurred, and there has been no sign of recurrence at

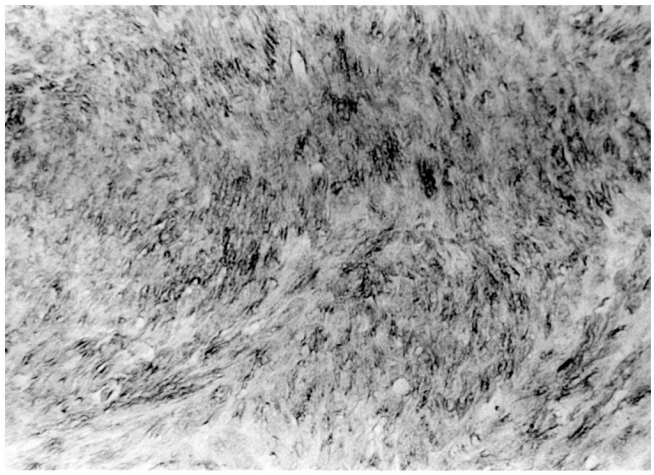

Fig. 4 Tumor cells showing strong S-100 reactivity (×200).

6 months post surgery.

Microscopic examination showed compactly arranged spindle cells oriented parallel to one another, forming interlacing bundles. There was nuclear palisading of Antoni A type (Fig. 3). The nuclei were not malignant. A confirmatory S-100 immunoperoxidase stain was positive (Fig. 4). The tumor had a thin connective tissue capsule. The final diagnosis was neurilemmoma.

DISCUSSION

The neurilemmoma, also called schwannoma or neurinoma, is a benign tumor of the peripheral nerve sheath believed to originate from Schwann cells. Schwannoma was first established as a pathologic entity in 1910 by Verocay [3]. Embryologically, Schwann cells arise during the fourth week of development from a specialized population of ectomesenchymal cells of the neural crest, which then detach from the neural tube and migrate into the embryo. Schwann cells form a thin barrier around each extracranial nerve fiber and wrap larger fibers with an insulating membrane, the myelin sheath, to enhance nerve conductance. As nerves exist in the brain and spinal cord, there is a change in the myelination by Schwann cells. Neurilemmoma arise when proliferating Schwann cells form a tumor mass encompassing motor and sensory peripheral nerves[4].

In a study of soft tissue tumors by Enjoji *et al*[5], there were 825 neurilemmomas out of 8,806 cases, a frequency close to that of hemangioma and lipoma. Das Gupta *et al*[6], reported that 20%

of a series of 136 tumors of the head and neck occurred either within or around the oral cavity. Neurilemmoma is a relatively rare benign oral tumor.

It is generally agreed that in the oral cavity, the tongue is involved most frequently. As for other oral locations, the palate and buccal mucosa are the next most common sites[7]. Slow growing, painful or painless lumps in and around the mouth are frequently found. A literature search revealed that 5 cases of neurilemmoma of the upper lip have been previously reported in Japan[1,2]. Neurilemmoma in the lower lip exceeds that of the upper lip by a ratio of 3.4～5.0:1[1,2,8]. The reason for this difference is thought to be due to the differences in embryonic development of the upper and lower lips. It has also been postulated that potential tumor cells are destroyed by the continuous presence of inflammatory cells in the lower lip induced by frequent traumatic episodes[9]. No clear reason for this difference has been proven. Neurilemmoma has been reported to be 2 to 4 times more common in women than in men[10]. It has also been reported however by Williams *et al*, that 83% of a group of 12 paitents were male[11]. Intraoral neurilemmomas in soft tissue appear as smooth submucosal swellings, thus resembling other lesions like mucocoeles, fibroepithelial polyps, fibromas, lipomas and benign salivary gland tumors.

Histologically, neurilemmoma is an unilocular mass surrounded by a capsule of epineurium and residual nerve fibers, often with the peripheral nerve attached to the edge of the neoplasm[11]. On histologic examination the neurilemmoma appears as a well-defined fibrous lesion that may present two different patterns. One pattern is the Antoni A type, which is characterized by cells that have aligned nuclei, simulating a palisade, and surround an anamorphous homogenous substance called Verocay body. The other pattern is the Antoni B type, which is characterized by cells that are arranged in a random fashion and are surrounded by collagenous fibers[12]. S-100 protein is useful in confiring the diagnosis of neurilemmoma. This acidic protein, common to supporting cells of the central and peripheral nervous system, can be consistently demonstrated in neurilemmomas as the majority of cells within neurilemmomas have the antigenic phenotype of Schwann cells. In our case, immunostaining was particularly strong in the Antoni A areas while less so in the Antoni B areas, a consistent feature in the neurilemmoma.

As a diagnostic and treatment tool, US, CT, or MRI may be helpful for the estimation of tumor margins and determination of whether there is infiltration into the surrounding structures. However, because most tumors of the upper lip present as relatively small lesions, a differential diagnosis is established using CT, MRI should not be considered as routine or necessary. We believe, therefore, that using US is useful in examining the upper lip. Few reports have described the characteristics of neurilemmoma on US[13-16]. Chinn *et al*[14] were the first to report the ultrasonographic characteristics of neurilemmoma. The US imaging showed heterogeneous, hypoechoic features and posterior acoustic enhancement consistent with previous reports[15,16]. Our findings were similar to these previous reports reported.

Treatment is always surgical, and complete excision results in no recurrence[4,12]. To our knowledge, no other reports have described malignant transformation of neurilemmoma in the upper lip[1,2,8,13].

ACKNOWLEGEMENT

We are grateful to Prof. N.Yoshimi of the Department of Pathology, School of Medicine, University of the Ryukyus for his help in evaluating the histological material.

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