Case Report

Unilateral swelling of cheek

ABSTRACT

A 55 year old male patient reported in the department of oral and maxillofacial surgery with a chief complaint of swelling on right side of face from last 2 months. He reported a progressive increase in the swelling. Pain and discomfort was present from last 7 days with increased swelling, and reduced mouth opening. On examination, the face appeared asymmetrical with a swelling in the right cheek which was small, smooth, dome shaped, present approximately 1.5 cm anterior to tragus. Overlying skin was of normal colour. The patient was afebrile. Mouth opening was 25 mm.

Keywords: Cheek, swelling, unilateral

INTRODUCTION

A 55-year-old male reported in the Department of Oral and Maxillofacial Surgery with a chief complaint of swelling on the right side of his face for the past 2 months. He reported a progressive increase in the swelling. There was no history of fever, dysphagia, or difficulty in breathing. Pain and discomfort was present for the last 7 days with increased swelling and reduced mouth opening. There was no history of trauma; no other skin lesion was present.

On examination, the face appeared asymmetrical with a swelling in the right cheek which was small, smooth, dome-shaped, present approximately 1.5 cm anterior to tragus [Figure 1]. Overlying skin was of normal color. The patient was afebrile. Mouth opening was 25 mm. Good oral hygiene with no abnormality on oropharyngeal examination was present. No evidence of inflammation was eminent around the swelling. The oral soft tissues were normal. Bimanual palpation showed a diffuse, painful lesion in the right masseter muscle region and the overlying skin was free. Mass was soft to firm in consistency and smooth. There was no associated thrill or fluctuancy. No neurologic deficits were present. Salivary flow was normal from the right Stensen's duct. All hematologic parameters were within normal limits. There were no masses or adenopathy on neck examination.

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Aspiration was negative. The patient had undergone orthopantomography (OPG). No odontogenic or nonodontogenic maxillary or mandibular lesions were appreciated on OPG. Ultrasonography revealed a hypoechoic lesion present in the cheek. Magnetic resonance imaging (MRI) with contrast was done which showed well-defined enhancing soft tissue mass lesion located anterior to the right mandibular ramus and lower temporalis muscle with mild surrounding inflammatory and edematous changes [Figure 2].

DIFFERENTIAL DIAGNOSIS

The most common swelling on cheek lesions might include the diagnosis of abscess, lipomas, salivary gland tumors, lymphadenopathy, and cysts. Tumors of muscular origin are also a consideration due to close approximation and

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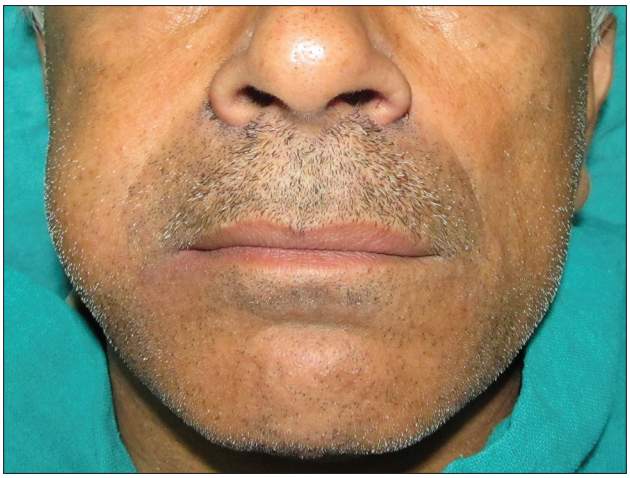

Figure 1: Swelling on the right cheek region

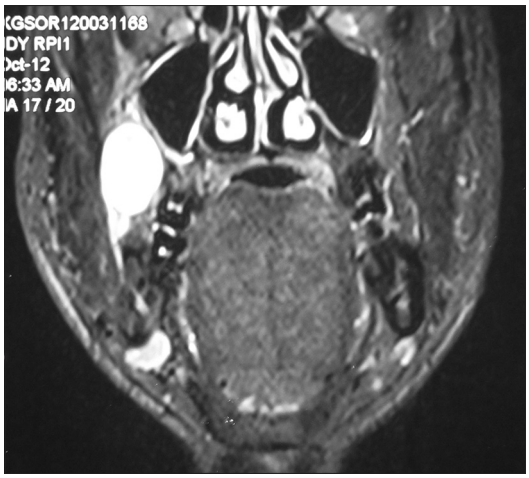

Figure 2: Coronal short T1 inversion recovery/T2-weighted image showing a well-defined mass on the right side between medial and lateral pterygoid muscles (along the course of mandibular nerve) which is hyperintense

involvement of masticatory muscles. Because this lesion was slow-growing, it was thought to be more likely a benign lesion.

Odontogenic infection and abscess becomes the first diagnosis due to acute onset and symptoms such as pain, swelling, and reduced mouth opening. Clinically, patient's intraoral examination was negative for carious lesion and pulpal pathosis and revealed a good oral hygiene. The panoramic radiography carried out was inconclusive. The patient was afebrile and had no signs of malaise. Patient's history revealed that swelling was slow growing. Thus, an abscess which was placed first on the differential diagnosis list was shifted down if not ruled out.

Lipomas are benign tumor of mature adipose tissue. It is the most common tumor of mesenchymal origin in head and neck region. These are commonly soft and superficial, but when they are infiltrative, they can exist entirely intramuscularly.[1] MRI excluded this diagnosis because lipomas had high signal intensity on both T1- and T2-weighted images.[2] In our patient, MRI showed low signal intensity on T1-weighted images and a high signal on T2-weighted images.

Any acute disease with swelling in the cheek region, pain, and limitation of motion also leads to the diagnosis of inflammatory lesions of the salivary glands, such as mumps or acute bacterial parotitis secondary to sialolithiasis. The 2-month history of swelling and normal hematologic values disagrees with the likelihood of an inflammatory lesion of the parotid gland.

The accessory parotid gland is salivary tissue separated from the main parotid gland. The accessory parotid gland exists in 21–61% of individuals. However, the appearance of an accessory parotid tumor is rare, with a reported frequency of 1–7.7% of all parotid gland tumors.[3] Pleomorphic adenoma is the most common benign neoplasm and mucoepidermoid carcinoma the most common malignancy.[4] Clinically, pleomorphic adenoma most commonly presents as an asymptomatic swelling, and this was consistent with the patient's first history, but falls short as a focus of inflammation noted in the examination.

Reactive hyperplasia of lymph nodes is often seen in the buccal or facial lymph nodes present at the anterior border of the masseter muscle.[5] Lymphadenopathy may progress into a submasseteric abscess which causes symptoms such as trismus and tenderness of cheek. It presents as high signal intensity on the T2-weighted images.

Progressively increasing swelling in the region of lymph node raises the suspicion of lymphoma. Hodgkin lymphoma has a bimodal presentation with one peak around the age of 15–35 years and another peak after the age of 50.[6,7] Clinically, it can present as an asymptomatic slowly enlarging mass and may be associated with systemic signs such as weight loss, early fatigue, and fever. Non-Hodgkin lymphoma commonly presents with swelling in the lateral neck. Its incidence increases with age.[6-8] The patient's hematological investigations and the presentation on MRI did not show lymph node connection. Thus, this was placed lower in our differential list.

Cyst-like cysticercosis (encysted larvae) may involve muscle and subcutaneous tissue.[9] In general, they remain asymptomatic for years but may cause pain due to inflammatory response produced when the larvae die. In general, these patients have eosinophilia due to parasitic infection. These features were absent in our patient.

Muscular tumors such as leiomyoma and leiomyosarcoma are rare in the oral cavity. These tumors arise mostly in the muscular layer of the abdominal cavity. In a recent review of the literature, 139 leiomyomas and 68 leiomyosarcomas of the oral cavity and pharynx were reported.[10]

Leiomyosarcoma presents with pain and swelling. The peak incidence was 40–49 years for benign tumors and 50–59 years for malignant lesions with males predominating over females. Neurogenic tumors such as neurofibroma and schwannoma were also included in the differential as they have been described in almost all anatomic sites in the head and neck region. But these are generally seen in second and third decades of life.

DIAGNOSIS AND MANAGEMENT

The patient was consented for excisional biopsy under general anesthesia. The proposed treatment included aspiration of the lesion with an 18-gauge needle and excisional biopsy under general anesthesia via a transoral approach. The lesion was removed without difficulty. The follow-up period (1 year) was uneventful [Figures 3 and 4].

PATHOLOGIC DIAGNOSIS

The submitted tissue showed hypercellular stroma with areas having parallel arrangement of intercellular fibers. Tumor cells in the stroma were spindle-shaped interspersed with fibers forming palisaded arrangement. Numerous areas show hyaline-like structures suggestive of Verocay bodies [Figure 5].

DISCUSSION

Schwannoma was first reported by Verocay in 1910 and called this benign neurogenic tumor as neurinoma.[11] These are also known as neurilemmoma, neurinoma, perineural fibroblastoma[12-14] and are slow-growing, encapsulated benign neoplasms derived from the sheath cells that encompass myelinated nerve fibers.

Both males and females are affected by neurilemomas. Quintarelli believes that males are more commonly affected than females. There are other authors who state that females are more prone to develop these tumors. These tumors can occur at any age but are frequently seen in the second and third decade of life.[13-17] Our patient was male and in the fifth decade of his life. These tumors are generally well defined and firm in consistency. Large lesions may present as soft and fluctuant mass. They are typically asymptomatic and slowly enlarge until causing functional or esthetic limitations. In the present case, the swelling was soft to firm in consistency and was slowly increasing in size and led to functional and esthetic restraint.

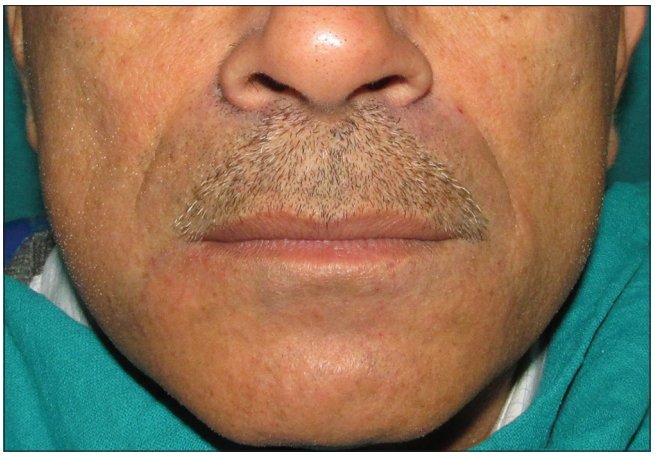

Figure 3: One-year follow-up photograph showing no swelling on face

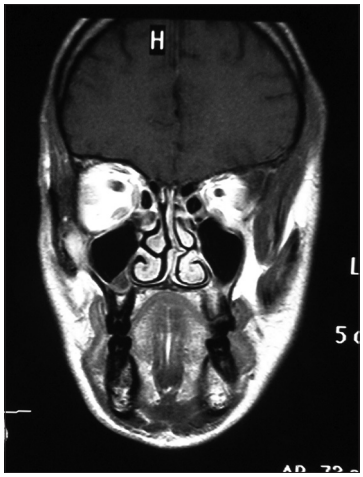

Figure 4: Coronal T1-weighted contrast-enhanced image 1 year after surgery showing no mass and normal musculature

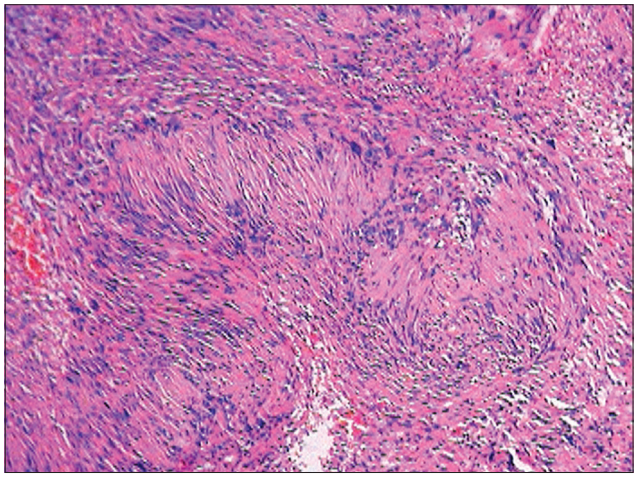

Figure 5: Spindle-shaped tumor cells in palisading arrangement with the presence of Verocay bodies (H and E, ×10)

Nerves which are most commonly involved in schwannomas of the head and neck are the vagus and the cervical sympathetic chain.[18] Trigeminal schwannomas are rarely seen and usually present as large masses due to the delay in the diagnosis and therefore in their surgical treatment.[19] We believe that in our case tumor arised from the terminal trigeminal branch as no facial nerve paresthesia was present and after excision of the tumor no motor or sensory functional disability was present.

Histologically, many schwannoma variants have been described, including common, glandular, plexiform, cellular, epithelioid, melanotic, and ancient schwannomas.[20] The microscopic portrait of schwannoma is distinct and can hardly ever be confused with that of other lesions. Schwannomas are unilobular masses enclosed by a capsule of epineurium and residual nerve fibers often with the edge of the neoplasm attached to the peripheral nerve. The core of the tumor is composed of a mixture of two cellular patterns, Antoni A and Antoni B[11-17,21-24] Antoni A areas are collection of compact spindle cells with twisted nuclei arranged in bundles or fascicles. In extremely differentiated areas there may be nuclear palisading and formation of Verocay bodies, which are formed by the arrangement of two rows of nuclei and cell processes which assume oval shape. Antoni B variant is less cellular and less organized, representing degenerated Antoni A areas composed of randomly arranged spindle or oval cells within myxoid, loosely textured, hypocellular matrix punctuated by microcyst, inflammatory cells, and delicate collagen fibers.[11-17,21-24]

An immunohistochemical examination of the tumor may show positive results with S100 antigen.[13,21-23] In the present case, the histologic analysis revealed a majority of Antoni A pattern with prominent nuclear palisading and formation of Verocay bodies, however few scanty areas showed Antoni B pattern also.

Ancient neurilemmoma exhibits benign degenerative changes in a classic neurilemmoma, taking place over a period of time. This change comprises cystic, myxoid, edematous and fibrotic areas, vascular abnormalities, and atypical cells with pleomorphic nuclei. Ancient neurilemmoma behaves much like a benign neural neoplasm.[21,24]

Cellular schwannoma is classified based on microscopic examination. Support for a microscopic diagnosis of cellular schwannoma can be obtained by immunostaining. It differs from classic schwannoma by its increased cellularity, nuclear pleomorphism and hyperchromatism, lack of Verocay bodies, and frequently higher mitotic activity.[25] The present case was of classical schwannoma.

Asaumi *et al*.[26] in their study described ultrasonography, computed tomography, and MRI may be helpful in diagnostic and treatment tools, for the estimation of tumor margins and the determination of infiltration to surrounding structures. MRI was particularly helpful in showing the internal characteristic of the encapsulated mass. Even though soft tissue schwannomas have no useful radiographic findings, in the rare case of intrabony (central) schwannoma, the role of plain film radiography in verifying locations and determining extent should be appreciated.[17,27]

Surgical excision is the treatment of choice. The proper surgical approach is decided by the site of the tumor and the extent of its spread. The nonencapsulated form requires a margin of normal tissue and careful separation from the involved nerve is also necessary to preserve normal function.[14,17,24,28] Recurrence[11-17,21-24] and malignant transformation of a benign schwannoma is rare.[29,30] In the present case, connection with the nerve could not be seen, the mass was well encapsulated and could be totally excised along with excellent prognosis. Swelling in the cheek region is one of the common symptoms given by the patient and include wide range of pathologies. The key to the success of this treatment is locating the specific area of origin of the tumor, defining its extent, and completely removing all diseased tissue trigeminal schwannoma although rare in this region must be kept in the differential diagnosis.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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