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Title: Transoral Surgical Approach to Solitary Fibrous Tumors in Buccal Space with Infratemporal Tumor Extension: A Case Report

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Title of the article:

Transoral Surgical Approach to Solitary Fibrous Tumors in Buccal Space with Infratemporal Tumor Extension: A Case Report

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Running Title: Solitary Fibrous Tumor in Buccal Space

Abstract:

A solitary fibrous tumour (SFT) is a rare spindle-cell neoplasm of mesenchymal origin usually located in the pleura. It has been recently described as occurring in various head and neck sites, including the oral cavity. The purpose of this article is to report a case of SFT originating in the buccal space and extending into the infratemporal space treated by means of transoral approach surgery. A 25-year-old female patient reported to the Department of Oral and Maxillofacial Surgery, Universitas Airlangga Hospital, chiefly complaining of a painless lump in the left cheek which had been present for nine months. The diagnosis was arrived at on the basis of a combination of clinical investigation, imaging studies and histopathological examination (biopsy). The surgical approach involved transoral incision through the buccal mucosa. An SFT of buccal space may extend to nearby structures producing the anatomical challenge of removal through a transoral approach. Excisional biopsy involving a transoral approach is, nevertheless, considered appropriate because it produces an attractive aesthetic appearance, reduces morbidity from nerve/vascular/gland injury and promotes more effective healing.

Key-words: solitary fibrous tumor, buccal space, infratemporal space, transoral approach

Text of the Article

Introduction:

A solitary fibrous tumour (SFT) is an uncommon spindle-cell neoplasm of mesenchymal origin. An increasing incidence of the tumor has been found in the extrapleural sites, including the peritoneum, mediastinum, orbit, infratemporal fossa parapharyngeal space, upper airways and nose, salivary and thyroid glands and oral cavity. As documented in the literature, SFT accounts for 3% of all cases occurring in the oral region. When SFT develops in oral cavity it appears like a painless swelling, some of them may give rise to compression symptoms and with differential diagnosis lipomas, pleomorphic adenomas, schwannomas, fibrous histiocytomas, benign glandular tumors and dermoid cyst. Members of both sexes are equally affected, with patients being predominantly middle-aged or elderly. Symptoms depend on the site and depth of the tumour, but tend to be mostly asymptomatic slow growing masses and shows a high rate of positivity for CD34 on immunohistochemical staining(1-8).

The case reported here is one of an SFT in the left buccal space extending into the infratemporal region. The depth at which the tumor was located between the spaces rendered both distinguishing it from other soft tissue tumor and its removal challenging. An initial diagnosis was established through clinical examination, imaging and incisional biopsy. Definitive treatment ultimately consisted of excisional biopsy incorporating a transoral approach due to the benefits of an attractive aesthetic appearance, reduced morbidity from nerve, vascular and gland injury in addition to enhanced healing.

Case Report:

A 25-year old female reported to the Oral and Maxillofacial Surgery Department, Universitas Airlangga Hospital, Surabaya presenting the primary symptom of a painless lump in the left cheek she had first detected nine months earlier. The individual concerned did not complain of toothache or numbness and had no previous history of ill-health. Further oral examination revealed asymmetry of the left-hand side of the face without accompanying redness (Figure 1A). Palpation indicated a firm, fully mobile, non-tender mass 4cms in diameter located relatively deep in the buccal space and extending into the left malar below the zygomatic arch. Palpation also confirmed the absence of lymphadenopathy in the head or neck region. Intraoral examination revealed a bulge in the left buccal mucosa, while palpation confirmed the presence of a well-defined, firm and non-tender mass located deep in the buccal space.

An initial orthopantomogram showed no destruction of the mandible and maxilla, but a post-extraction socket existed in the 36 region due to the patient having undergone a tooth extraction two months before (Figure 1B). Ultrasonography (USG) of the left cheek indicated the presence of a discrete well-demarcated, hypoechoid solid mass 2.6x2 cm in size located on or overlapping with the left parotid gland. There was no increase in internal in vascular flow or calcification of the mass (Figure 1C).

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A head and neck CT scan with contrast detected a developing soft tissue tumor 2.69x3.73x4.0 cm in size in the left buccal space (Figure 1D) resulting in indentation and saucerization of the left posterior maxillary sinus wall, medial pterygoid, masseter and left temporal muscle which appeared as a slow growing tissue mass. The tumor originated in the buccal space and extended into the temporal space. The mass was fed by branches of the left buccal artery. The bone structure was normal.

A fine-needle aspiration biopsy was performed by puncturing an area from the extraoral to the intraoral site. Microscopic examination showed a hypocellular area consisting of a broad distribution of erythrocytes with a number of inflammatory cell histiocytes and neutrophils, but no signs of malignancy. An incisional biopsy was also performed through an intraoral incision on the left buccal which revealed tissue with wide fibroblast proliferation, extremely similar to a fibroma.

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Following the securing of patient consent, an excisional biopsy using an transoral approach was performed under general anesthesia. First, the left buccal was palpated to localize the mass area which was subsequently marked with methylene blue. The incision site was also marked in the middle of the mass. Prior to incision, 1:200.000 of pehacain was

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injected as a vasoconstrictor to reduce oozing during surgery (Figure 2A). A blunt dissection was performed post-incision as far as the mass area which was then completely separated (Figure 2B, 2C). The tumor was capsulated but, being brittle, it tended to rupture during removal. Therefore, it proved necessary to conduct dissection in several steps in order to achieve total removal. Since the mass extended deep into the temporal space, the most difficult aspect of the removal came during dissection through the temporal space which was located immediately below the zygomatic arch.

The gross pathology of the tumor confirmed a total of five grams of specimen material (Figure 2D). From the microscopic findings, it was evident that the tumor tissue was composed of fibroblast proliferation with spindle and flat nuclei arranged in a patternless structure within the collagen matrix. Partly dilated vasculars were also present in the tumor resulting in its ultimate pathological diagnosis as a solid fibrous tumor (Figure 2E).

No post-surgery complications of paraesthesia, paralysis or parotid injury were apparent and a routine follow-up conducted six months after the operation found no evidence of recurrence.

Discussion:

An SFT is a rare mesenchymal neoplasm generally considered to occur in ubiquitous interstitial stem cells situated within soft tissues. Although most appear in the parietal or visceral pleura or peritoneum, they can be present in other extrapleural sites, including: the mediastinum, lungs, liver, breasts, retroperitoneum, spine, meninges and extracranial head and neck regions. Symptoms depend on the site and depth of the tumour. Usually slow-growing and asymptomatic, it is described as having normal overlying skin and mucosa (1,2,9-11). The patient reported an unspecific slow-growing lump on her cheek which was not tender and had first appeared approximately nine months previously. Immunoreactivity for CD34, bcl-2, CD-44, CD99 and Vimentin but is negative to keratin, EMA, S-100, desmin, smooth muscle actin and muscle specific actin was helpful to confirm the diagnosis of SFT (12,16).

Although not pathognomonic, homogeneous or heterogeneous, attenuated enhancement is reported to be the most prominent feature of SFT revealed through CT and MR imaging. This characteristic is attributed to high vascularity because of the prominent vascular channels within the tumor. While remodeling of the adjacent bones may be observed in large, long-standing lesions, frank bone destruction represents an exceptional finding that should prompt suspicions of a malignant tumor (2,9). Extrapleural SFTs are almost always

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The authors would like to express their gratitude to Universitas Airlangga Hospital for providing data relating to the case and Danang Limanto for assistance with illustration.

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Figure Legends

Figure 1. (A) Extra oral picture showed an asymmetry on the left cheek (black arrow) and (B) Orthopantomogram revealed no bone destruction of maxilla and mandible, (C) USG showed a hypoechoid solid mass on the left parotid area. (D) CT scan with contrast enhancement from coronal, axial, and sagittal view revealed a slight enhancing soft tissue tumor with well border

Figure 2.(A,B,C) Injection of vasoconstrictor 1:200.000 and Blunt dissection to reach the mass; (D) Tumor specimen excised from buccal space. (E) Microscopic view of the mass using HE staining with 20x and 40x magnification

Figure 3. (A) Location of tumor showed in blue area, (B) Design of incision.