# Asymptomatic subcutaneous swelling of lower face

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## CLINICAL PRESENTATION

A 17-year-old male presented at the School of Dentistry, complaining of an asymptomatic subcutaneous swelling on the right cheek lasting around 1 year. The patient referred to a history of extraction of the right mandibular first molar (for treatment of caries) 6 months after the swelling appeared, combined with systemic antibiotic treatment for a suspected dental abscess. Trauma or previous lesions in the region were not recalled by the patient, who had no remarkable past medical or family history and no smoking or drinking habits. No other contributing information was obtained from the anamnesis. The physical examination revealed a 3 × 3 cm skin-colored solitary subcutaneous nodule, located in the right submandibular area (Figure 1). The overlying skin presented serosanguinous drainage on palpation, but there was no lymphadenopathy. No intraoral signs were visible, and panoramic radiography did not indicate any noteworthy alterations.

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

## DIFFERENTIAL DIAGNOSIS

The clinical aspect and the absence of radiographic alterations indicated a benign condition, and therefore, the preliminary differential diagnosis comprised residual dental abscess, foreign body granuloma (FBG), epidermoid cyst, dermoid cyst, epidermal inclusion cyst, pleomorphic adenoma, and poroma.

An *abscess* is defined as a cavity containing pus, resulting from tissue disintegration. A periapical abscess develops when the dental pulp and root canal become infected and necrotic, usually because of dental caries. Although drainage through the alveolar bone into the oral cavity is more common, when the abscess is located in the mandible, the infection may spread below the mylohyoid muscle, reach the sublingual and/or submandibular space, and drain through the skin. If the microorganism is able to evade the host response, a chronic infection is established, with formation of granulation tissue to confine the bacterial agent, often presenting with little or no symptoms as in the case presented here.[1] Considering the presence of the lesion for 1 year, a radiographic radiolucent area was expected as a result of bone destruction caused by the infection of the right inferior molar. However, the panoramic radiograph showed no signs of bone involvement, and the drainage did not contain suppuration. In addition, antibiotic therapy did not lead to resolution.

An FBG may develop anywhere in the body after penetration of external substances into skin. The list of substances includes tattoo pigments; cosmetic fillers; drugs and medications; metallic or mineral particles; other particles, such as food, glass, fibers and sutures; and endogenous materials, such as calcium deposits and keratin.[2] The clinical presentation of an FBG varies widely, from a classic inflammatory reaction with erythema,

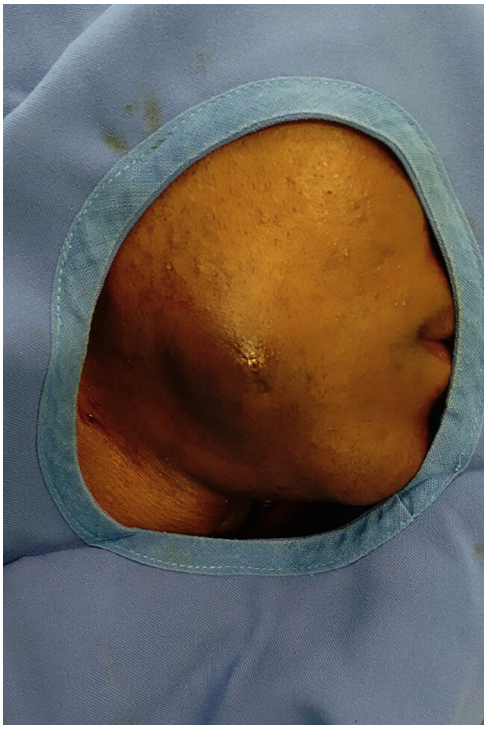

Fig. 1. Solitary subcutaneous nodule, located on the right submandibular area, measuring 3.0 × 3 cm.

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swelling, and pain to a chronic granulomatous inflammatory reaction appearing as an asymptomatic solid mass, with gradually increasing volume.[3] The latter features are compatible with the present case, justifying FBG as an important differential diagnosis.

Epidermoid and dermoid cysts are rare lesions that may grow anywhere in the body, presenting in the head and neck in 7% of cases, including the submandibular cutaneous region, most commonly in the midline for dermoid cyst.[4,5] These developmental cysts are benign, slow growing, and well circumscribed and usually present as asymptomatic lesions. However, if secondary infection occurs, it may become acutely symptomatic and present serous fluid.[5] Most commonly, these cysts affect males in the first 4 decades of life, as in the present case. Histologically, the epidermoid cyst is lined by stratified squamous epithelium, without ectodermal appendages. If skin appendages are present, the lesion is regarded as a dermoid cyst.[6] In addition, the epidermal inclusion cyst, which is an acquired epidermoid cyst that develops after traumatic or surgical implantation of epithelium into mesenchymal structures, presents with similar clinical features.[7] Therefore, although the patient did not present an obvious etiology, the epidermal inclusion cyst was part of the differential diagnosis.

Although less probable because of the patient's age, pleomorphic adenoma (PA) of the parotid gland was also considered. PA is the most common benign tumor of parotid gland that frequently occurs in middle-aged individuals (mean age 52.8 years),[8] as a slow-growing, asymptomatic mass with a slight predilection for women. About 80% of cases develop in the tail of superficial lobe of the parotid gland, similar to the reported case.[8] Considering the exposed features, it is important to emphasize that the lesion described in the present case showed discharge, which would not be expected for PA.

Poromas are benign sweat gland neoplasms most commonly known to arise on hairless regions of the body.[9] However, a series of 353 cases revealed that 30% were located on the face, 10% on the scalp 14% on the trunk, 15% on the feet, and 5% on the hands.[10] Poromas present as well-delineated papules or nodules that are skin colored or pigmented because of the presence of melanin and are usually solitary. The peak of incidence is the seventh decade of life, although the ages of the patients at diagnosis ranged from 15 to 80 years in a series of head and neck cases, with no gender predilection.[10] In addition, some lesions may present symptoms, such as pain, bleeding, and clear discharge.[9,10]

Although malignancy was less favored because of the clinical characteristics of the present case, for the completeness the differential diagnosis, we also included cutaneous leiomyosarcoma or rhabdomyosarcoma (RMS). Leiomyosarcoma is a smooth muscle–derived malignant neoplasm that generally follows an aggressive course, accounting for 1% to 4% of soft tissue sarcomas in the head and neck region. The lesion most commonly presents cutaneously, developing superficially in the subcutaneous layer, or it may present in deeper tissues. The average age at presentation ranges from 50 to 64 years.[11] RMS is a rare neoplasm of skeletal muscle differentiation, which most often occurs in deep tissues, with less than 1% developing as a primary cutaneous neoplasm. The lesion normally affects individuals under age 10 years, with an additional peak occurring in adolescence, as in our patient.[12]

## DIAGNOSIS

Total excision of the lesion was performed via submandibular incision. The sample was placed in 10% formalin and submitted for histopathologic diagnosis. The gross specimen showed a well-defined firm cystic lesion, with a fibrous capsule and multiple intracystic yellowish villous-like projections, measuring 35 × 33 × 18 mm (Figure 2).

The histopathologic examination revealed a cystic cavity with multiple intraluminal villous and papillary projections, lined by a partially hyalinized membrane containing flat cells that resembled synoviocytes. The fibrous

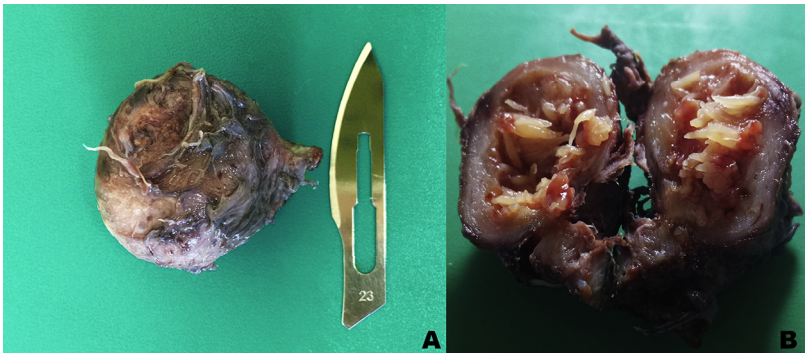


Fig. 2. Macroscopic examination. **A,** Well-defined gross specimen, measuring 35 × 33 × 18 mm, and of firm consistency. **B,** Resected cystic lesion, revealing multiple intracystic yellowish villous-like projections.

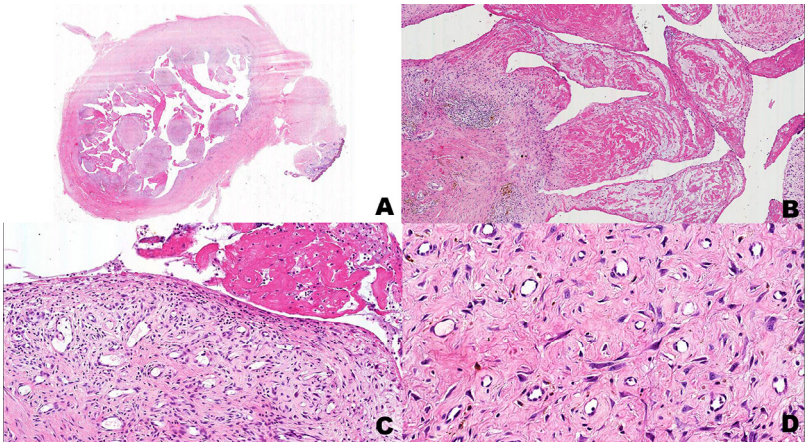


Fig. 3. **A,** Cystic structure, with multiple intraluminal villous projections, surrounded by a fibrous capsule (hematoxylin and eosin [H&E], Fit zoom). **B,** Villous and papillary structures, exhibiting areas of hyalinization and fibrin deposition (H&E; original magnification ×40). **C,** Membrane containing flat cells that resembled synoviocytes, lining the cystic cavity and villi, along with hyalinized areas and fibrous connective tissue (H&E; original magnification ×100). **D,** Fibrous connective tissue of villi and capsule, composed of varying amounts of spindle and stellate shaped fibroblasts (H&E; original magnification ×200). *A high-resolution version of this slide for use with the Virtual Microscope is available as eSlide: VM04835.*

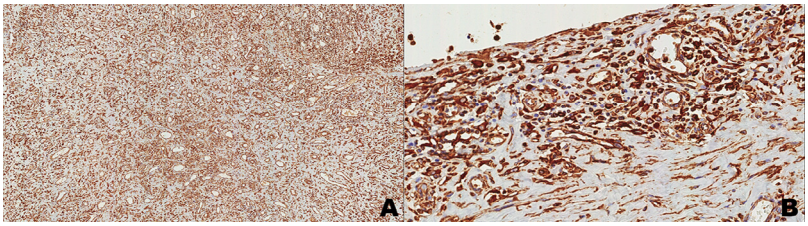


Fig. 4. Positive immunohistochemistry for vimentin. **A,** Uniform positivity (original magnification ×40). **B,** Immunopositive staining of lining membrane (original magnification ×200). *A high-resolution version of this slide for use with the Virtual Microscope is available as eSlide: VM04836.*

connective tissue of both the capsule and the villi presented areas of hyalinization, along with fibrin deposits and varying amounts of spindle and stellate shaped fibroblasts. Chronic inflammatory cells were also visible (Figure 3). Immunohistochemistry staining was positive for vimentin (Figure 4) and negative for S100 and CD34. On the basis of the microscopic characteristics, a diagnosis of cutaneous metaplastic synovial cyst (CMSC) was established.

## MANAGEMENT

The postoperative course was uneventful, and 16 months later, the patient is symptom free and has no evidence of recurrence.

## DISCUSSION

Gonzalez et al.[13] reported the first case of CMSC, 3 decades after Selye[14] experimentally demonstrated the induction of hyperplastic synovium by air injection in the connective tissue of rats in locations not related to the joint or tendon sheath areas. CMSC is a rare lesion that occurs in skin as opposed to the synovial cysts that develop in bone and joints after orthopedic surgery. Up to now, the English language literature contains no more than 31 cases of this lesion, only 8 of them located in the head and neck region, including the present case.[15-17]

In most cases, the lesions present at sites of previous skin trauma or surgical intervention,[16] and it has been suggested that the synovial metaplasia may represent an uncommon reactive process elicited by surgical trauma, although some of the reported cases do not present any association with history of skin damage.[18] Moreover, the etiology remains unclear and could be linked to relative tissue fragility secondary to certain conditions, such as Ehlers-Danlos syndrome, rheumatoid arthritis, basal cell carcinoma, and long-term systemic corticosteroid administration.[16,19] Embryologically, the normal synovium develops from the epithelioid cells derived from the histiocytes of bone marrow and from the fibroblasts from local mesenchymal cells, which differentiate after local mechanical disruption of connective tissue that occurs as consequence of movement of the embryo in vivo. Thus,

it is hypothesized that metaplastic synovial-like structures could originate from the differentiation of multipotential surrounding connective tissue cells, during the wound healing process in response to tissue disruption, as seen in most of the reported cases of CMSC.[15,16] In the present case, the patient did not recall any previous lesion, traumatic injury, history of systemic corticosteroid use, or insertion of a foreign body.

In addition to CMSC, other synovial-related conditions might occur in the head and neck region. Synovial chondromatosis is an uncommon pathology that affects joints, most likely as a result of metaplasia of mesenchymal tissue rests within the synovial membrane causing production and secretion of cartilaginous bodies into the joint space. Although larger joints, such as the hip, knee, and shoulder, are more commonly involved, approximately 100 cases in the temporomandibular joint have been reported. This condition can occur primarily, with no known predisposing factor, or may be secondary to trauma, arthritis, or internal derangement.[20] Pigmented villonodular synovitis is a rare disorder that may also affect the temporomandibular joint, which accounts for about 50 cases reported in the literature. It is characterized by benign and locally aggressive proliferation of synovium, most probably caused by chronic inflammatory changes of the synovial tissue.[21]

CMSC can occur in any site of the body and usually presents as a solitary erythematous subcutaneous nodule, varying from 0.4 up to 6.0 cm, with a tender consistency, related to a scar in most cases. However, multiple or skin-colored lesions have also been reported.[16,22] The literature reports no gender predilection and a wide age range of occurrence (7-82 years), although in most cases, patients aged greater than 40 years were affected. In addition, serosanguinous fluid may drain or be aspirated from the lesion, as occurred in the case reported here.[16]

The normal synovial tissue is composed of an outer layer, which contains blood and lymphatic vessels, and an inner layer comprising a sheet of flat synovial cells.[15] Likewise, CMSC presents a thin layer of flat cells that resemble a hyperplastic synovial membrane, surrounding a cystic cavity with multiple villous projections toward the center. Despite the cystic architecture, CMSC is not a true cyst because it lacks an epithelial lining. The fibroconnective tissue of the pseudocyst wall contains a mixture of epithelioid, fibroblastic, mononuclear inflammatory, and multinucleated giant cells. In addition, hypocellular areas are a common feature, appearing as extensive zones of hyalinization with fibrin deposition. The surrounding dermis or subcutaneous tissue may also show reactive changes, such as fibrosis.[16,23]

With regard to immunohistochemical features, patients with CMSC are positive for vimentin and negative for CD34, S100, carcinoembryogenic antigen, epithelial membrane antigen, and keratin, supporting its mesenchymal origin. The immunoreactivity for CD68, a positive marker in normal synovium, is variable—some cases showing negativity and others positivity. This variability might only indicate a structural similarity between the lesion and normal synovial tissue and not the same origin and nature for them.[15,16] Nevertheless, an immunohistochemical study is not mandatory for diagnosis because CMSC can be diagnosed by using morphologic characteristics.

CMSC presents a low recurrence rate and a benign behavior, indicating excisional biopsy as the preferred treatment. Additionally, elimination of possible local trauma to the involved areas is recommended. So far, 2 recurrent cases have been reported in the literature; in one case the recurrence was caused by persistence of a traumatic factor and in the other for unknown reasons.[16,24]

## CONCLUSIONS

We have reported here a case of oral CMSC in a male patient. Considering the rare occurrence of this lesion, it is extremely important to develop consistent and well-documented reports for better understanding of the lesion.

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