

# A case report of coexistence of a sialolith and an adenoid cystic carcinoma in the submandibular gland

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## ABSTRACT

The occurrence of sialoliths in the submandibular gland is 80% due to the specific anatomy of both the gland and its duct. The diagnosis is rather easy because of the obvious clinical signs of the entity. Imaging studies are always necessary in order to treat the patient as effectively as possible. The stones do not tend to occur within the gland as frequently as in the respective duct. The coexistence of sialoliths and malignant tumors is extremely rare. A 70-year-old woman with intraparenchymal stone was operated in our ENT department. In addition to the sialolith the pathological examination revealed the existence of an adenoid cystic carcinoma (ACC), that extended to the neighboring skeletal muscle. This is the reason why we believe it would be useful to report this case of a large stone (14 mm in diameter) located in the submandibular gland coexisting with ACC. This case report is a very good example illustrating that all available means should be used prior to reaching a conclusion and making a health professional decision.

**Key words:** Sialolithiasis, adenoid cystic carcinoma, submandibular gland.

## INTRODUCTION

Sialolithiasis is a disease characterized by the occurrence of sialoliths in the different parts of the salivary glands. Sialoliths are hard formations, with round or oval shape and a variety of sizes. Their colour ranges from white to brown and have nodular surface with prominences, the smaller ones looking like coral. Sialoliths are usually composed of a round organic core, intensely calcified, that is surrounded by alternative layers of organic and inorganic substances. The organic layers are composed of condensed mucus, mucopolysaccharides, glucoproteins, cellular elements and lipids, while the inorganic material is composed of calcium phosphate, calcium carbonate and trace elements (1). The etiology of the disease is assumed to be related to the specific anatomic and physiological factors of the affected gland. Salivary gland calculi occur in the submandibular gland in 80 per cent of the cases, far more commonly than in the parotid gland (20%)(2). The incidence of sialolithiasis in the sublingual gland is very rare (3) and in some studies absent (4).

The incidence of adenoid cystic carcinoma varies in bibliography. According to some studies it constitutes the second most frequent malignant neoplasm of salivary glands (10,0%), second only to mucoepidermoid carcinoma (15,7%) (5), whereas according to other authors it is the most frequent tumor of the salivary glands (27%) (6). Others report that it is the fifth frequent tumor after mucoepidermoid carcinoma, adenocarcinoma, acinic cell carcinoma, and polymorphous low grade adenocarcinoma. It is the most frequent malignant tumor of epithelial origin in the submandibular and the minor salivary glands constituting 30% to 40% of malignant tumors of these glands (5,7). However, in other series this tumor has not been detected in the submandibular gland (8). Adenoid cystic carcinoma increases slowly and insidiously. It is characterized by a high tendency of recurrence, early nerve infiltrations and hematogenous metastasis. The prevailing patients' complaint is of a painless swelling rather tender in palpation. The aim of this study is to report the coexistence of sial

thiasis and adenoid cystic carcinoma of the submandibular gland. The possibility of a malignancy in the submandibular gland in the presence of sialolithiasis has not been described before.

### CASE REPORT

A 70-year-old woman presented herself in the emergencies of our ENT department complaining of intermittent episodes of moderate to severe pain and swelling in the submandibular region for a week. The intensity of the pain increased during mealtimes. The swelling was described as minimal in the morning on awaking but varied in size throughout the day. Her medical history was negative. Upon inspection, the patient's face appeared asymmetric. A 5 cm hard tender swelling in the right submandibular region was palpated. Oral examination revealed negligible salivary flow from the duct of the right submandibular gland. No cervical lymph nodes could be detected. There was no evidence of inflammation in the mouth, especially along the Wharton's duct, and attempts to milk the left submandibular gland produced only minimal amounts of saliva. The ultra-sound examination revealed a 1,4 cm size oval shaped stone, inside the right submandibular gland's substance with an irregular arrangement of gland parenchyma and without swelling of local lymph nodes. For calculi that lie in the proximal duct or gland the treatment of choice has been sialoadenectomy (9) A critical stone size has been reported (less than 7 mm) above which lithotripsy is unsuccessful (9). According to these guidelines we did not try lithotripsy, intraoral removal or basket retrieval. On the contrary the patient was scheduled and finally underwent surgical removal. The gland was solid and strongly adherent to the neighbouring tissues and the stone was yellow with nodular surface.

In addition to the sialolith, the pathological examination revealed the existence of an adenoid cystic carcinoma that extended to the neighboring skeletal muscle.

The patient underwent computed tomography of thorax and abdominal area that did not reveal any pathological findings. Computed tomography of head and neck however showed a small uneven fibrosis below the point of surgical incision. The following day, the patient was submitted to FNA (Fine Needle Aspiration Biopsy) that revealed the existence of malignant cells.

A second operating procedure was performed. The patient underwent functional neck dissection on the right side and the region was carefully controlled with the help of microscope. There were no signs of contamination of the lingual, the hypoglossal or the marginalis mandibulae nerve. The removed tissue proved to be fibrous. The histopathological examination confirmed the existence of adenoid cystic carcinoma. The patient underwent postoperative radiotherapy for 3 weeks after the second incision. 36 months later, the laboratory results showed no signs of recurrence.

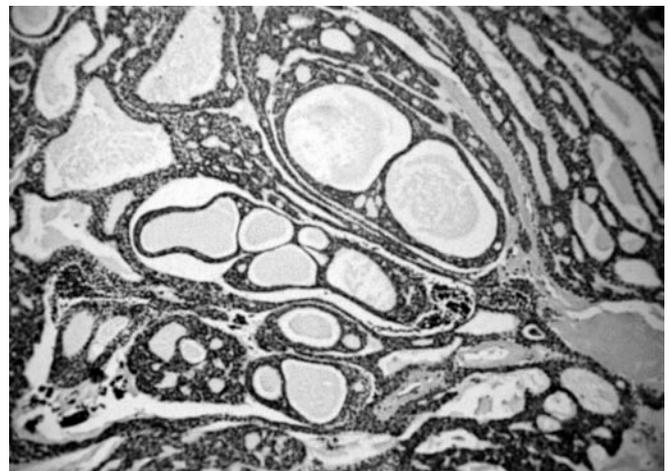


Fig. 1. Adenoid-cystic carcinoma with variation in size and shape of the cystic spaces and arrangement of the tumor cells in "Swiss cheese" configuration (cribriform pattern).

### DISCUSSION

The sialolithiasis is a very common disease, which can be asymptomatic for a long time. Microcalcifications occur frequently in the submandibular glands. In a study of normal submandibular glands at autopsy Scott (10) found that solid-appearing calcified and noncalcified micro deposits in small ducts increased with age. Seifert and associates (11) stated that the earliest changes of sialolithiasis result from electrolyte imbalance in the saliva and the filling and distension of small ducts with viscous mucoid secretions. Lamellar microcalcifications would form later by precipitation of additional secretions and appositional growth. In a study by Levy (12) and associates, 5% of patients with submandibular stones had asymptomatic, discrete masses, which in a few instances were suggestive of a neoplasm on physical examination.

Salivary gland neoplasms, as well as mesenchymal neoplasms (tumors) located in the salivary gland regions, are common and may also be confused with sialolithiasis. Neoplasms may cause pain and paresthesia in the affected regions. Any lesion in question should be evaluated. Sialolithiasis should be differentiated from a tumor of the salivary gland (2). However, the accuracy of sonography in assessment of sialolithiasis is approximately 90% (13). A tumor was ruled out for this patient because the swelling and pain were not constant and a radiopaque nodule was detected in the simple X-ray of the submandibular gland, as well as in ultrasonography. On the other hand, sialolithiasis most commonly occurs in the submandibular gland at a rate reportedly fluctuating between 80% and 95% (10). Most stones are found in the submandibular gland because of the viscous consistency and mineral content of its saliva and the long, irregular length of its duct (11).

The coexistence of sialoliths and malignancies has been reported for parotid but not for submandibular gland. Multiple myeloma (14) and lymphoid tissue lymphoma (13) have

been reported previously in relation to sialoliths in parotid gland. Adenoid cystic carcinoma in the submandibular gland simultaneously with sialolithiasis in the same site has not been described before.

CT is widely used and has the advantage of being very sensitive in the detection of calcifications (15); however, its disadvantages include a relatively high radiation dose, and high cost. Some authors (16) state that CT does not add substantial new information of ductal changes and they used sialography with US for the diagnosis of salivary calculi. These two methods demonstrate the morphologic features of the ducts proximal and distal to the obstruction and provide information on aspects of obstructive salivary gland disease that affect outcome - that is, the location and possible mobility of the stones. In diagnosis of tumors, computed tomography shows the submandibular gland as a structure of similar density to muscle, so evaluation of extension of disease around the gland is sometimes difficult (17). Yasumoto et al (18) reported that 27 from 35 submandibular gland tumors could not be seen clearly on precontrast CT scans. Magnetic resonance (MR) imaging is less sensitive than radiography and CT in detecting sialolithiasis. However, MR imaging can delineate various kinds of soft tissues clearly with high contrast resolution. Takashi et al (19) reported in one study that all patients with sialadenitis had diffusely abnormal signal intensity in the submandibular gland, which was different from focal abnormalities of neoplasms. On the contrary, MR imaging could not differentiate between benign and malignant tumors, and was not useful in histopathologic diagnosis in the same study.

The probability of coexistence of sialolithiasis with malignancy cannot be overemphasized. However, there are cases where the physician should be cautious. In this patient the diagnosis of sialolithiasis was performed by: a) the patient's history of an intermittent pain and swelling of the submandibular region during mealtime; b) inspecting the face for asymmetry; c) palpating the gland for enlargement and consistency; and d) after making an ultra-sound examination that revealed a 1,4 cm size, oval shaped stone. If a CT had been performed, probably, we could have seen the stone but not the carcinoma. If we had performed an MRI, we should differentiate between infection and tumor and should exist alertness for carcinoma occurrence. The ultrasound control alone is not enough to decide the kind of operation in a patient with sialolithiasis. We would suggest an MRI when the suspicion index is high, since this may save the patient from a second surgery. That is when:

The stone is intraparenchymal.

When the stone is large (>1cm)

When in ultrasound there are signs of irregular arrangement of gland parenchyma.

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