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THE CLINICAL ENTITY OF SIALOLITHIASIS OF THE MINOR SALIVARY GLANDS

by

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INTRODUCTION

Sialolithiasis of the minor salivary glands seems to be a rather rare entity, as judged by the small number of publications on this subject.

In 1955 *Ligtherman* reported the first case of the disease. In subsequent years individual cases appeared in the literature (*Chaudry et al.*, 1960; *Erickson & Hale*, 1962; *Wussov*, 1963; *Moskow et al.*, 1964; *Alexander & Andrews*, 1965; *Fourestier et al.*, 1966). The present author has published three further cases, *Holst*, 1968, and in 1969 three more cases have been reported, (*Allan et al.*, *Crawford & Guernsey*). So far 13 cases of sialolithiasis of the minor salivary glands have been published; these cases being listed in Table I, according to author, sex, age of the patients, and the location of the sialoliths.

The present report contributes 10 additional cases and presents the first published cases of sialolithiasis of the minor salivary glands of the tongue. Furthermore the first cases of the clinical appearence of multiple sialoliths with different locations in the same patient is presented.

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Table I.

Distribution of sialolithiasis in the minor salivary glands according to sex, age, and location of the lesion in previously published cases

Sex	Age	Location	Author	Year
1. f	51	cheek	Ligtherman	1955
2. f	70	cheek	Chaudry et al.	1960
3. m	40	cheek	Erickson & Hale	1962
4. m	63	cheek	Wussow	1963
5. f	69	cheek	Moskow et al.	1964
6. m	52	cheek	Alexander & Andrews	1965
7. m	72	cheek	Fourestier et al.	1966
8. m	49	cheek	Holst	1968
9. m	71	upper lip	Holst	1968
10. m	45	cheek	Holst	1968
11. m	63	upper lip	Allan et al.	1969
12. m	56	cheek	Allan et al.	1969
13. m	29	upper lip	Crawford & Guernsey	1969

REPORT OF CASES AND RESULTS

Case 1. A 50-year old woman referred to the Dental Department, University Hospital of Copenhagen because of recurrent swellings of the upper lip. The patient stated that the swelling had persisted during the last six years. The intra-oral examination revealed two pea-sized, hard, mobile nodules in the labial mucosa in the region of the upper right canine and the upper left canine. A fistula was observed in both regions, from where a thin liquid could be expressed. Under local anesthesia the nodules were excised. In both lesions concrements were seen during the operation. The microscopical examination of both specimens confirmed the diagnosis of sialolithiasis. The specimen from the rigth side contained even several concrements in a dilated duct (Fig. 1).

Case 2. A 20-year old woman was referred to the Dental Department, University Hospital of Copenhagen because of a nodule in the buccal sulcus of the right lower jaw in the premolar region. The lesion was detected during a routine dental examination by the family dentist. Intra-oral inspection revealed a pea-sized, hard, mobile nodule in the region mentioned. The covering mucous membrane appeared normal in the affeced area. Under local anesthesia the lesion was excised and a concrement was found in the removed tissue which microscopically showed a chronic sialoadenitis in the mucous salivary glands and remnants of hard concrements in the tissue.

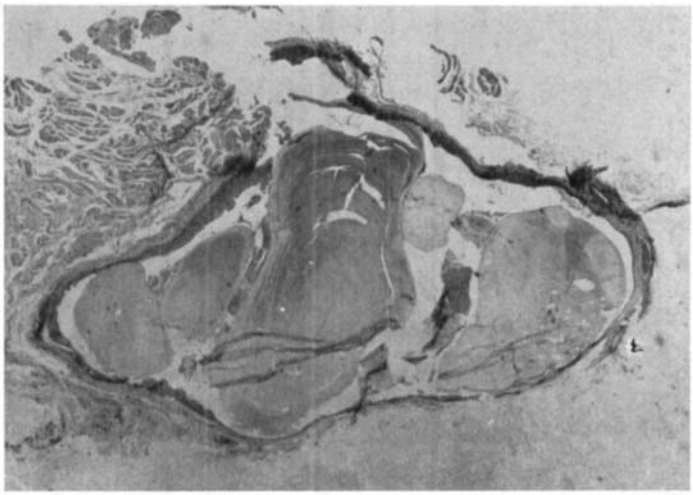

Fig. 1. The histological specimen from a swelling in the upper lip in a 50-year old woman. Several concrements are seen in a dilated salivary duct.

Case 3. A 64-year old man was referred to the Dental Department, University Hospital of Copenhagen, because of a tumor in the right margin of the tongue; the tumor was observed by the patient himself. The intra-oral examination showed a yellow, nodule-like, hard mass located just anteriorly to the papillae foliatae, (Fig. 2). The mass was excised under local anesthesia and the histological examination revealed a mineralized concrement with accumulations of bacteriae in the surrounding tissue.

Case 4. In a 67-year old woman a biopsy was taken from a normally appearing mucous membrane of the right buccal mucosa in a study comprising the histopathology of normal oral mucosa carried out in the Dental Department, University Hospital of Copenhagen. The microscopical examination revealed a specimen covered by a normal nonkeratinized stratified squamous epithelium with normal differentiation. In the subepithelial connective tissue a cross-sectioned salivary duct was seen, surprisingly, containing a sialolith. Surrounding the duct a sligth infiltration of lymfocytes and plasma cells was observed.

Case 5. A 66-year old man was referred to the Department of Oral Surgery, Royal Dental College, Copenhagen, because of a hard mobile nodule in the right upper lip. The covering mucous membrane appeared normal in the area. The lesion was excised under local anesthesia and a stone-like concrement was seen during the operation. The microscopical examination showed a tissue dominated by mucous salivary glands and ducts. In one of

Fig. 2. Sialolithiasis of the minor salivary glands of the tongue in a 64-year old man. A well circumscribed nodule is seen in the right margin of the tongue.

the ducts, which was dilated, three eosinophilic concrements were found, and a squamous metaplasia of the ductal epithelium had occurred. Almost two years after the first examination the patient was referred again because of a recurring of the swelling of the right upper lip. The intraoral examination revealed a nodule-like swelling both in the right and the left upper lip. Under local anesthesia the lesions were excised. During the operation three stone formations were observed in the rigth side. The histological examination of the excised tissue from both locations confirmed the diagnosis of sialolithiasis.

Case 6. A 69-year old woman was referred to the Department of Oral Surgery, Royal Dental College, Copenhagen, for treatment of a swelling in the right cheek in the region of the upper second premolar. A fistula was seen in the central part of the lesion, which had a soft consistency. The patient stated that she had noted the presence of the swelling during the last six or seven years. No symptoms from the swelling was recorded. The affected area was excised under local anesthesia. Microscopical examination showed in the connective tissue of the specimen, a cystic formation covered by an unkeratinized squamous epithelium with two cell-layers. Centrally in the lumen, an accumulation of mucine was observed in connection with a lamellated stone formation. A dilated duct with squamous metaplasia was seen.

Case 7. A 24-year old man was referred to the Department of Oral Surgery, Royal Dental College, Copenhagen, because of a hard, indolent nodule in the left side of the upper lip. In the central part of the lesion a fistula was observed. The duration of the affection was not known. The mass was excised

under local anesthesia and a stone formation was seen during the operation. The microscopical examination showed the presence of a sialolith.

Case 8. A 39-year old man was treated in the Department of Periodontology, Royal Dental College, Copenhagen. During the routine dental examination a hard, indolent swelling of the right upper labial mucosa, close to the midline, was detected. The covering oral mucosa was normal. The lesion was excised under local anesthesia and during the operation a hard concrement was found. The microscopical diagnosis was sialolithiasis.

Case 9. A 49-year old man was referred to an oral surgeon because of an indolent swelling of the labial mucosa in the region of the upper right lateral incisor. A hard nodule was observed in the affected area. Under local anesthesia the lesion was excised and the microscopical examination revealed a sialolith in a dilated salivary duct.

Nine additionel cases of sialolithiasis in the minor salivary glands have been presented. Together with the three cases previously published by the author, the material comprises a total of 12 patients. The mean age of series was 51 years (range 20—71). There were eigth males aged 24—71 years (mean age 51 years) and four females aged 20—69 years (mean age 52 years). Five cases were located to the buccal mucosa and six were located to the mucosa of the upper lip. One case occurred in the lateral margin of the tongue. In two patients two separate locations of sialoliths were observed in each side of the upper lip. In one patient the entity recurred almost two years after the first operation. The new cases in the material are listed in Table II, according to sex, age, and location of the lesion.

The clinical appearence of the entity was mostly that of a hard, mobile, indolent nodule, or a firm, well defined swelling. However, in one case the

Table II.

Distribution of sialolithiasis in the minor salivary glands according to sex, age, and location of the lesion in the present material

Sex	Age	Location
1. f	50	upper lip (right and left side)
2. f	20	cheek
3. m	64	tongue
4. f	67	cheek
5. m	66	upper lip (right and left side)
6. f	69	cheek
7. m	24	upper lip
8. m	39	upper lip
9. m	49	upper lip

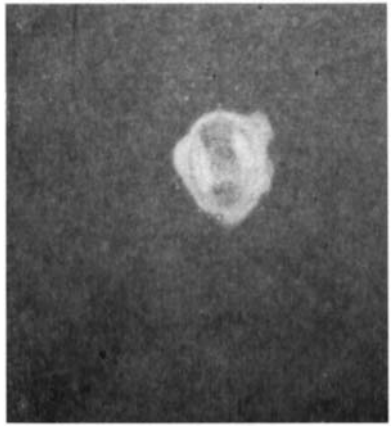

Fig. 3. A radiograph of the right upper lip in a 71-year old man revealing a lamellated stone formation in the soft tissue. From: Holst, E., J. Oral Surg. *26:* 354—356, 1968. Reprinted by permission of the American Dental Association.

swelling was soft of consistency. The mucous membrane covering the affected area was either normal or penetrated by a fistula, most often situated in the central part of the lesion. If secondary inflammation had occurred, pus could be expressed through the fistula. In one case from the previous material a radiograph of the soft tissue showed a radiopacity and the lamellated structure of the stone was distinctly seen (Fig. 3). In some other cases radiographs were taken but did not show any radiographical alterations.

Microscopically, the sialoliths usually were found in a dilated salivary duct as a large, lamellated, well organised stone that filled the ductal lumen

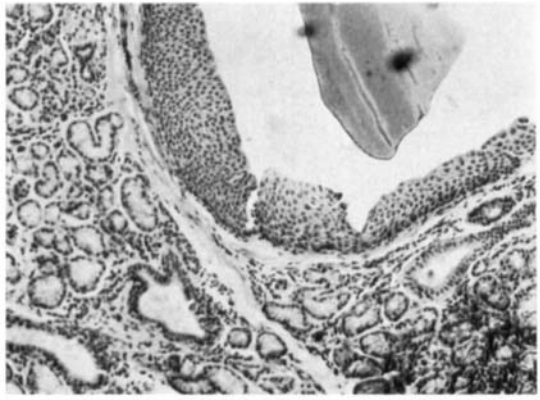

Fig. 4. Squamous metaplasia in the ductal epithelium. The central part of the salivary duct contains a stone formation.

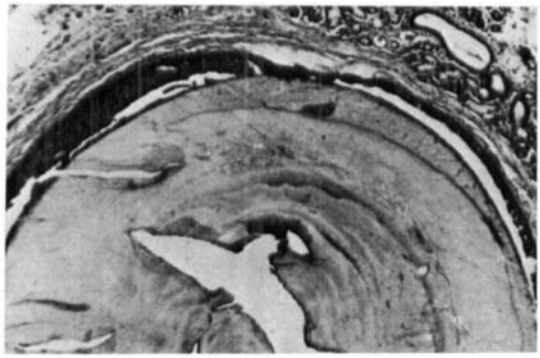

Fig. 5. Respiratory ciliated epithelium in a salivary duct containing a sialolith. From: Holst, E., J. Oral Surg. *26:* 354—356, 1968. Reprinted by permission of the American Dental Association.

(Fig. 4). Also numerous small well circumscribed concretions in different stages of calcifications were obserbed (Fig. 1). Squamous metaplasia of the ductal epithelium was a common finding (Fig. 4) and even metaplasia into respiratory ciliated epithelium was encountered in one of the cases from the authors previous material (Fig. 5).

In all cases the treatment was surgical excision under local anesthesia and no complications were observed in the postoperative period.

DISCUSSION

A total of 15 cases of sialolithiasis in the minor salivary glands in 12 patients have been collected by the author. 12 of these have been found within the last four years during which the interest of the author has been focused on this disease. Summarizing the results of other investigators no more than 10 cases have been reported by nine different authors. Consequently, these authors have characterized the entity as extremely rare. However, with regard to the present results, this type of sialolithiasis seems not to be an unusual finding in the mucous membrane of the oral cavity, and it should be considered as a possible diagnosis when swelling of the oral tissues is encountered.

The most frequent location of the disease is the buccal mucosa, but the lip should also be regarded as a potential site of sialolithiasis of the minor salivary glands. Probably, sialolithiasis may develop in any location of the oral mucosa where salivary glands are found. The condition is maybe even more frequent than the reported cases indicate, which is stressed by the

accidental finding in a biopsy from a clinical normally oral mucous membrane. The cases of sialolithiasis in more than one location in the same patient and even recurrence of the entity as reported here in one case may also support this statement.

Sialolithiasis in the major salivary glands may occur at any age but is most common among the middle-aged persons (Husted, 1953). The same tendency is also seen from the published cases of minor salivary gland sialolithiasis. From Tables I and II it appears that the average age of the latter entity is 54 years, ranging from 20—72 years. The female/male distribution was found to be almost 1:2 (7 females and 15 males).

Wooten (1969) has published a case of the entity in the premaxilla in connection with supernumerary teeth and a follicular cyst in a patient with congenital alveolar ridge cleft. This case has not been included in this discussion because the description of the case does not clearly prove that the calcification is a sialolith in the minor salivary glands. The possibility of a rhinolith can not be excluded, which also is stressed by the author. Moreover, a calcification in the follicular cyst should be taken into consideration.

Squamous metaplasia of the ductal epithelium is a common finding associated with this entity. The phenomenon is probably caused by a long-standing chronic irritation and was found in several of the published cases. Even metaplasia into pseudostratified columnar epithelium and respiratory ciliated epithelium has been observed (*Chaudry et al.*, 1960, *Holst*, 1968).

The etiology of the disease is not known. It was stated in a previous paper (*Holst*, 1968) that trauma may constitute part of the pathogenesis. Thus, all published cases are apparently located in close connection with the teeth. Therefore, traumatizing of the mucous membrane by the teeth during mastication or speech may be a possible pathogenesis. This trauma may again cause desquamation of epithelial cells and in connection with bacteriae a nucleus may be formed in the salivary duct with subsequent calcification.

The finding in one of the reported cases of a stone formation within a cystic cavity may rise the question of sialolithiasis as etiology in some of the cysts encountered in the oral mucous membrane.

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SUMMARY

Nine additionel cases of sialolithiasis of the minor salivary glands were presented. A total of 12 cases of this disease has then been collected by the

author. Ten of these have been found within the last four years. Compared with previous publications of the entity, ten cases by nine different authors, this suggests that the disease is not so rare as previously stated. Five of the presented cases were located to the upper lip, three to the cheek and one to the lateral margin of the tongue. In two patients two separate sialoliths were found in different sites of the oral cavity. The clinical, histological and etiological aspects of the disease were finally discussed.

RÉSUMÉ

ENTITÉ CLINIQUE DE LA LITHIASE DES GLANDES SALIVAIRES MINEURES

Neuf cas supplémentaires de lithiase des glandes salivaires mineures ont été présentés. Un total de 12 cas de cette affection a ensuite été recueilli par l'auteur. Dix de ces cas ont été constatés pendant les 4 dernières années. Par comparaison avec les travaux publiés antérieurement sur cette entité, lo cas présentés par 9 auteurs différents, il semblerait que cette affection ne soit pas aussi rare qu 'on l' a indiqué antérieurement. Parmi les cas présentés, il y avait 5 cas de localisation a la lèvre supérieure, 3 à la joue et un au bord latéral de la langue. Chez deux patients, on a trouvé deux sialolithes séparés à des endroits différents de la cavité buccale. L'article se termine par une discussion sur les aspects cliniques, histologiques et étiologiques de cette affection.

ZUZAMMENFASSUNG

DIE KLINISCHE GESAMTHEIT VON SPEICHELSTEINE DER KLEINEN SPEICHELDRÜSEN

Neun neue Fälle von Speichelsteine der kleinen Speicheldrüsen wurden präsentiert. Der Verfasser hat in Total 12 Fälle dieser krankheit gesammelt; zehn von diesen seit vier Jahren. Im Vergleichung mit frühen Arbeiten über das Krankheitsbild, zehn Fälle die von neun verscheidenen Verfassern publiciert geworden sind, könnte bedeuten, dass die Krankheit nicht so selten wie früher postuliert ist. Fünf der aktueller Fälle waren in der Oberlippe lokalisiert, drei in der Wangenschleimhaut und einer zum Zungenrand. Zwei Speichelsteine wurden an verschiedenen Stelle der Mundhöhle in zwei patienten gefunden. Die klinische, histologishe und ätiologische Verhältnisse der Krankheit wurden diskutiert.

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