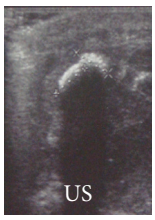

CASE

Obstructive Submandibular Sialadenitis Complicated with Severe Neck Phlegmon: Transcutaneous Removal of Sialolith with a Flow of Pus: Literature Review of Extraoral Complications

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SUMMARY

Sialolithiasis, as the most common reason (60–85 percent) of obstructive salivary gland disease, in the rare cases, may be complicated by soft tissues abscess or/and fistula. The purpose of this report is to present a rare case of submandibular gland sialolithiasis complicated with severe neck phlegmon in a 47-year-old Caucasian male. Ultrasonography and transcutaneous removal of sialolith upon the purulent locus lancing are highlighted. Our literature review based on the existed ones with a total 24 complication cases is presented. Wakoh et al`s classification of submandibular gland sialolith-associated fistulas types is analyzed. The ultrasound imaging is still underestimated and not adequately popularized among head and neck and oral and maxillofacial surgeons. Presented case and published reports show the usefulness of this constantly developing diagnostic technique in a combination with knowledge of possible extraoral purulent complications' and its management.

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Please cite this article as: Savchuk LA, Fesenko II. Obstructive submandibular sialadenitis complicated with severe neck phlegmon: transcutaneous removal of sialolith with a flow of pus: literature review of extraoral complications. J Diagn Treat Oral Maxillofac Pathol 2021;5(11):125–34.

Abbreviation 'US' at the upper right icon means that article contains ultrasound image.

Paper received 25 October 2021
Accepted 07 November 2021
Available online 30 November 2021

https://dx.doi.org/10.23999/j.dtomp.2021.11.1

INTRODUCTION

Sialolithiasis, as the most common reason (60–85 percent)[1] of obstructive salivary gland disease, in the rare cases, may be complicated by soft tissues abscess[2,3] or/and fistula.[4,5] A clinical study by Kishore Kumar et al (2012) of the 200 cases of cutaneous sinuses revealed that only in 0.5 percent of cases was noted cutaneous fistula as a result of submandibular gland calculus.[6] Such fistula usually is named by authors as *orocutaneous* or *sialo-cutaneous fistula*.[5,7]

According to our literature search, submandibular gland calculus-associated abscesses/phlegmons are even rarer comparing with sialo-cutaneous fistulae. Purulent floor of the mouth processes as a complication of obstructive salivary stone usually were diagnosed by multi-slice computed tomography (MSCT).[3,8] Despite ultrasound (US) are widely described as a diagnostic tool for submandibular sialolithiasis[1,9,10] its application in case of calculus-associated cervical fistulae and abscess/phlegmon to our knowledge is presented in very limited publications.[11,12]

The purpose of this report is to present a rare case of submandibular gland sialolithiasis complicated with severe neck phlegmon in a 47-year-old male. Ultrasound and transcutaneous removal of sialolith upon the purulent locus lancing are highlighted. Our own literature review based on the existed ones with a total 24 complication cases is presented.

CASE

A 47-year-old Caucasian gentleman was referred to the Kyiv Regional Clinical Hospital with a left neck and submandibular swelling (Fig 1), fever, difficulty of swallowing. The complaints had been initiated with salivary colics several weeks before. Previously, during the last years, patient noted a painful chewing–associated submandibular swelling at the left side.

US was performed by an experienced doctor of ultrasound diagnostics (L.A.S., her experience – 31 years). Submandibular and neck regions were examined bilaterally according to the protocol (ie, swelled and healthy side). Using linear transducer (12-5MHz) in a longitudinal submandibular position, the gray scale US (also known as B-mode [brightness mode]) showed hyperechoic semilunar structure (ie, sialolith, measured 0.76 × 0.78 cm) with an artifact of acoustic shadowing behind (Fig 2). Also, in this left submandibular region, the submandibular gland with a hypoechoic parenchyma and the collection of hypoechoic heterogenic content (ie, pus) were visualized. The diagnosis of obstructive sialadenitis caused by sialolith and complicated with submandibular and left neck phlegmon was established.

The phlegmon lancing was done under the local infiltration anesthesia using 8.0 ml of 4 percent Ultracain® D-S solution (Sanofi-Aventis Deutschland GmbH, Frankfurt am Main, Germany) mixed with 12.0 ml of 0.9% saline solution in a 20-ml syringe. Intravenous sedation was done by anesthesiologist`s team. An oblique 3.5-cm incision was planned and done inside the prominent upper neck skin crease (Fig 1C). A 1.2-cm salivary calculus (ie, sialolith) was removed with a flow of pus (~22 ml) upon the finger examination (ie, revision) of the purulent wound (Fig 3). Tubular double drainage was installed after rinsing with an antiseptic solution. Post-operative period associated with infusion and antibacterial therapy and wound rinsing showed gradual decrease of soft tissues inflammatory changes and complaints.

DISCUSSION

Digestion-associated swelling[9] (also known as "mealtime syndrome"[13]) in the submandibular region is a common disorder at the in- and out-patient oral and maxillofacial units. And typically, this is a manifestation of obstructive sialadenitis caused by sialoliths. Such complications of obstructive sialoliths as pus collection in the cellular spaces and fistulae are not common and its diagnostic features and management strategies are to be analyzed. In a literature review we analyze all purulent extraoral complications' manifestations (abscesses, phlegmons, sialo-cutaneous fistulae, and tender cutaneous nodules).

Table 1 is presenting our literature review of submandibular sialolithiasis complications and its management strategies based on the reviews of Ha et al (2020),[14] Wakoh et al (2021)[7].

Among all 24 published cases (Table 1) of submandibular sialolithiasis with extraoral complications in the majority of cases (n = 17) there were noted the cutaneous fistulous tracts (in 69.56 percent), collection of pus in cellular space – in 5 cases (in 21.74 percent), and tender cutaneous nodule – in 2 cases (in 8.7 percent).

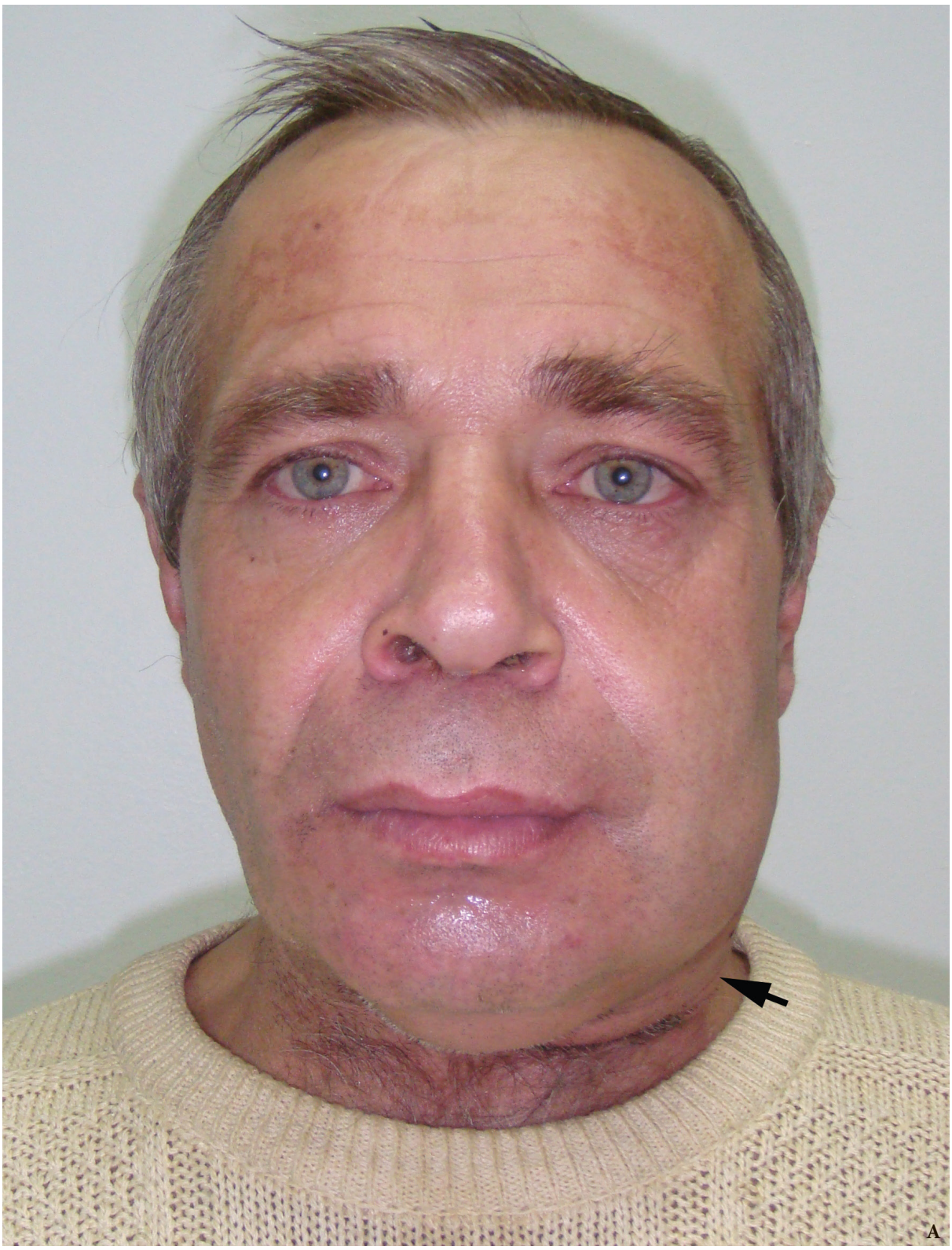

FIGURE 1. Preoperative anterior view (**A**). Notes perspiration on the lower face, soft tissue swelling (*arrow*) in submental, left submandibular, and left neck region. Skin erythema visualized at the area of swelling and reached the level of left clavicle and suprasternal notch. **(FIGURE 1 continued on next page.)**

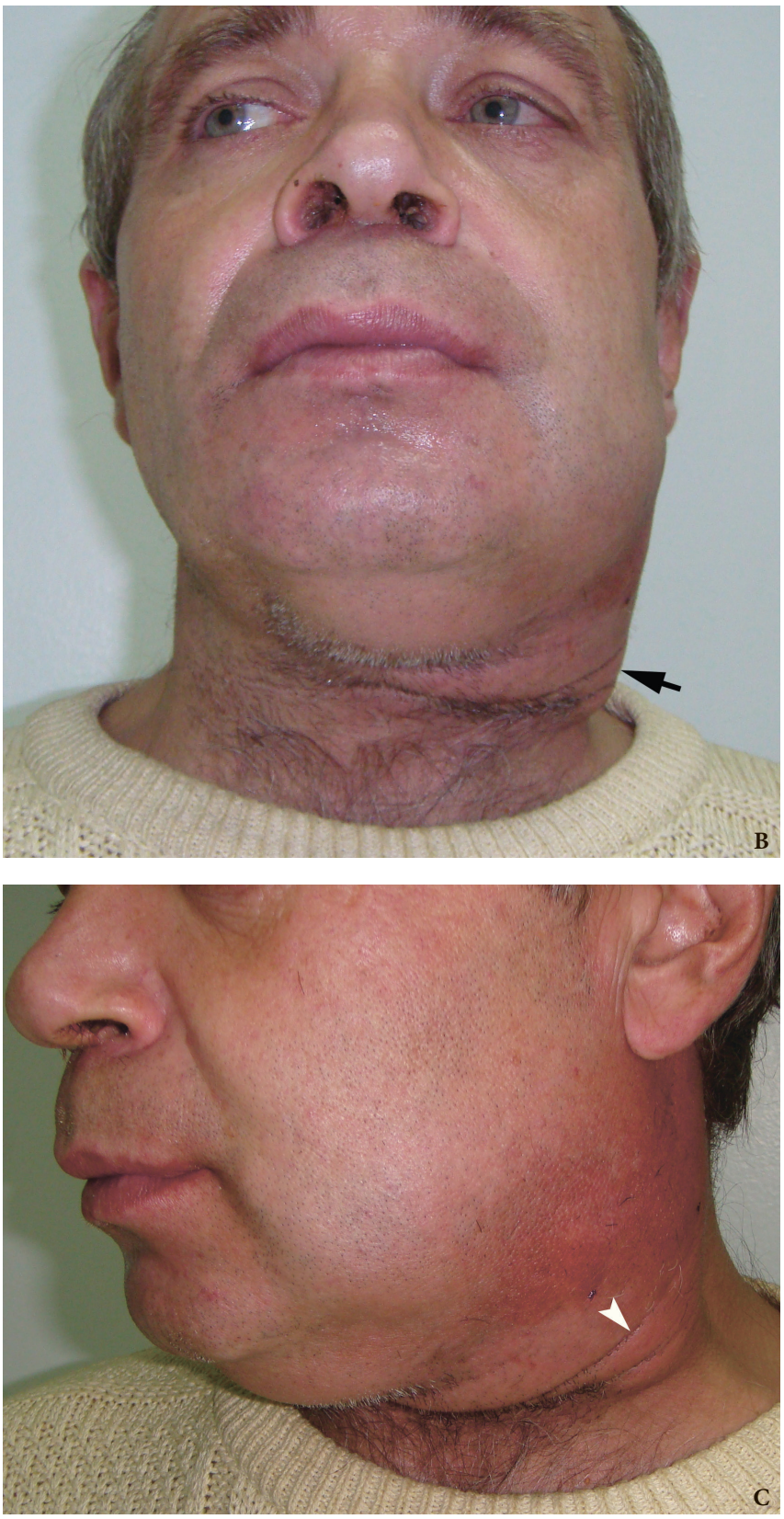

FIGURE 1 (continued). Preoperative 45 degree angled (**B**) and lateral view (**C**). An oblique 3.5-cm incision was planned and done inside the prominent upper neck skin crease (*arrowhead*).

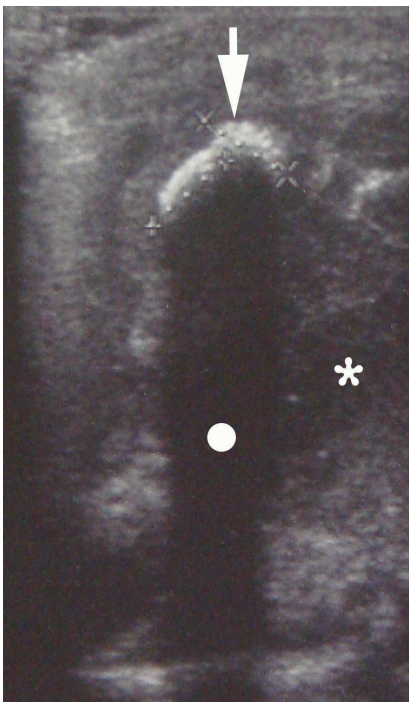

FIGURE 2. Preoperative gray scale ultrasound shows hyperechoic semilunar structure (ie, sialolith) (indicated by '+' and '×' calipers and *arrow*) with an artifact of acoustic shadowing behind (*dot*) and the collection of hypoechoic heterogenic content (pus, indicated by *asterisk*).

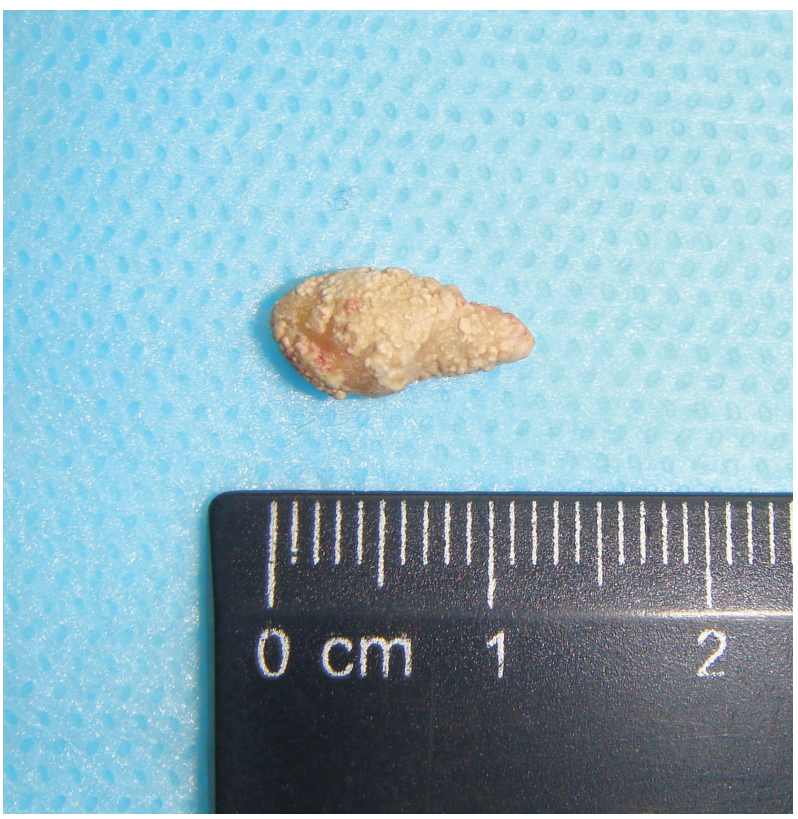

FIGURE 3. A cropped view of a 1.2-cm sialolith after its removal upon the phlegmon lancing. The sialolith was removed with a flow of pus upon the finger revision (ie, exploration) of the purulent wound.

TABLE 1. Literature Review of Submandibular Sialolithiasis with Purulent Extraoral Complications' Manifestations and Its Management. **(TABLE 1 continued on next page.)**

#	Authors	Sex/Age (Yrs)	Salivary Gland	Extraoral Complication	Imaging	Surgical Management
1	Present case (treated in 2015, published in 2021)	M/47	Left submandibular	Submandibular and upper neck phlegmon	US	Transcutaneous removal of sialolith during the stage of finger examination of the purulent wound upon the phlegmon lancing
2	Wakoh et al (2021)[7]	M/49	Ectopic right submandibular	Abscess with discharging sialo-cutaneous fistula in submandibular region	OPG, MSCT (non-contrast and contrast enhanced)	Abscess lancing and calcified mass removal
3	Bridwell et al (2020)[3]	F/64	Right submandibular	Mouth floor purulent process in cellular spaces (abscess, according to authors)	MSCT	Ductotomy with removal of sialolith and drain placement
4	Hoffman (2018)[8]	M/Adult	Left submandibular	Submandibular abscess	MSCT	First surgery: Abscess lancing, draining. Second surgery: Sialadenectomy with a stone removal
5	Stegmann et al (2018)[11]	F/6	Left submandibular	Submandibular cutaneous fistula	US, sialendoscopy	Endoscopic removal of the sialolith and excision of the fistula
6	Kusunoki et al (2017)[4]	M/72	Left submandibular	Poor left upper neck granuloma in a skin defect with a fistulous tract	MSCT	Sialadenectomy with sialolith removal and fistula excision
7	Ballal et al (2016)[15]	M/80	Left submandibular	Cutaneous fistulous opening at the left neck	CT (No data it was CBCT or MSCT)	Excision of fistulous tract and submandibular gland with removal of calculus
8	Singh et al (2015)[16]	M/53	Left submandibular	Fistulous opening in submandibular region	Contrast-enhanced MSCT, X-ray fistulogram	Excision of the fistulous tract and submandibular gland
9	Rangappa et al (2014)[17]	F/55	Left submandibular	Cutaneous fistula in the left neck	Lateral X-ray view	Excision of the fistulous opening, tract and submandibular gland with sialolith removal
10	Saha et al (2012)[5]	M/54	Right submandibular	Cervical fistula	X-ray fistulography	Excision of the submandibular gland with the fistulous tract
11	Kishore Kumar et al (2012)[6]	No data	Submandibular	Cutaneous sinus	No data	No data
12	Jayachandran et al (2011)[18]	M/52	Right submandibular	Both, sialo-oral and sialo-cutaneous fistula	Lateral oblique X-ray view, MSCT, MRI	Excision of the gland with removal of sialolith and surgical fistula closure.
13	Salikumar et al (2006)[19]	F/56	Right submandibular	Discharging sinus in the right submandibular region	US, MSCT, fistulogram	Submandibular sialadenectomy with sialolith removal and fistulectomy

TABLE 1 (continued). Literature Review of Submandibular Sialolithiasis with Purulent Extraoral Complications' Manifestations and Its Management.

#	Authors	Sex/Age (Yrs)	Salivary Gland	Extraoral Complication	Imaging	Surgical Management
14	Almasri (2005)[20]	M/70	Left submandibular	Discharging fistula in the left neck	MSCT, OPG	Excision of submandibular gland and the stone
15	Drage et al (2005)[13]	F/45	Left submandibular	Sinus at the lower third of left neck	CT, fluoroscopy	Transcutaneous removal of the migrated stone from the superficial soft tissues
		M/73	Left submandibular	Discharging sinus in the middle third of left neck	Plain radiographs	
16	Karengera et al (1998)[21]	M/64	Left submandibular	Cutaneous fistula in a submandibular region	–	First surgery: Widening the fistula and drainage placement. Second surgery: Transcutaneous removal of the stone with a drain and large amount of necrotic tissue. Third surgery: Sialadenectomy do to the further calculi
17	Paul and Chauhan (1995)[22]	M/45	Right submandibular	Both, sialo-oral and sialo-cutaneous fistula	Lateral X-ray of the neck	Intraoral removal of the calculus and excision of the cutaneous fistula
18	Abe et al (1990)[12]	M/25	Right submandibular	Cervical fistula	X-ray sialogram, OPG, US	Excision of the submandibular gland and salivary fistula
19	Asfar et al (1989)[23,14]	M/55	Submandibular	Orocervical fistula	No data	Excision
		M/43	Submandibular	Tender cutaneous nodule	No data	Excision
		M/75	Submandibular	Tender cutaneous nodule	No data	Excision
20	Cartwright and Hardingham (1983)[2]	F/79	Left submandibular	Left neck abscess	Plain X-ray of the neck	First management: Pus aspiration and prescription of ampicillin. Second management: At day 10 after aspiration – partial excision of the submandibular gland with a sialolith
21	Druez et al (1982)[24]	No data	Submandibular	Cutaneous fistula	No data	No data

US, ultrasound.
OPG, orthopantomography.
MSCT, multi-slice computed tomography.
CT, computed tomography.
X-ray, radiography.
MRI, magnetic resonance imaging.

Among 16 fistulous cases, in two cases both sialo-oral and sialocutaneous fistulae were noted from one calculus, and in one case – the fistula located in a tissues with a resolution of the submandibular abscess.

Among five abscess/phlegmon cases, the abscess with discharging fistula was noted in one case (Wakoh et al)[7].

Bridwell et al presented the description and computed tomography of an obstructed submandibular sialolith with abscess mimicking Ludwig's angina.[3]

Henry Hoffman presented an adult patient with submandibular region abscess due to the submandibular gland sialolithiasis, obstruction disease, and acute infection.[8] In the reported institution`s protocol, the step by step management strategy was perfectly highlighted with a purpose to be a helpful guidelines for other practitioners designed to bridge the gap between procedural concepts and their implementation.[25]

Among five calculus-associated abscesses presentations and descriptions in three cases (Cartwright and Hardingham, Hoffman, and Wakoh et al)[2,8,7] a limited collection of pus was noted, and in two cases (Bridwell et al and our case)[3] the purulent process was diffuse and localized in two or more anatomical regions.

Worth of attention is the classification proposed by the authors from Japan who classified all submandibular gland sialolith-associated fistulas into four types[7]:

1. Sialo-oral fistula.[26]
2. Sialo-cutaneous fistula.[4–6,11–13,15–17,19–21,23,24]
3. Sialo-pharyngeal fistula (synonym: sialo-parapharyngeal fistula).[27]
4. Sialo-oro-cutaneous fistula.[18,22]

Wakoh et al presented the case of cutaneous fistula related with ectopic submandibular gland sialolith.[7] Ha et al (2020) exhibited a sialo-cutaneous fistula but with no evidence of submandibular gland sialolith. In their cases the diagnosis of submandibular sialadenitis was established and the conservative treatment helped to eliminate the symptoms.[14]

Cases with intraoral complications of submandibular sialolithiasis (like submandibular duct fistula, etc.) were excluded from this discussion and can be analyzed in further studies. Despite the fact that in the report of Chandak et al (2012) the case with diagnosis of acute submandibular sialadenitis complicated with abscess was established there was no radiological evidence of sialoliths in submandibular gland.[28]

In the reported cases, some authors combined multiple diagnostic tools, like Jayachandran et al (X-ray, MSCT, MRI),[18] others used a single one, like Hoffman (MSCT)[8]. Among 24 complication cases presented in the Table 1, computed tomography (non-contrast/contrast enhanced) was used in 10 cases, OPG – in three cases, X-ray – in five cases, X-ray fistulography – in three cases, fluoroscopy – in one case, US – in four cases, MRI – in one case, sialoendoscopy – in one case, and X-ray sialography – in one case. Thus, despite the fact that in the majority of cases (n = 10) the teams had chosen CT, ultrasonography (n = 4) has an undiscovered potential among surgeons.

US became a vital diagnostic tool for pathology detection in different glands – thyroid,[29] lymphatic (ie, lymph nodes),[30] and salivary ones[31]. Katz et al are more than right pointing out that a clinical US is to be indicated as soon as the first symptoms of sialadenitis occur.[32] US is more than useful in the detection of structural changes of the salivary gland parenchyma, vascularization, condition of the duct system, sialoliths[9,10] and even mucous plugs[31]. The authors note that approximately 20 to 40% of the salivary stones are not opaque on plain radiography, but most of these sialoliths are visible upon sialography.[32] 2.0-mm and longer sialoliths can be detected on gray scale US.[32]

Practitioners must keep in mind the possible calcifications in the area of major salivary glands as potential mimickers of sialoliths. These can be: healed tuberculous lymphadenitis, phleboliths, tonsilloliths, segmental ossification of the stylohyoid ligament.[33] So, analysis of the anamnesis data, complaints and radiological features can significantly help in differential diagnostics.

In different countries, the purulent process presented in our case can be described by different terms. In English-speaking countries the term *abscess*[34] is usually used, and in some of East European countries (Ukraine, etc.) the term *phlegmon* can be used for the similar condition. In general, *phlegmon* is an acute, clearly not limited purulent inflammation of cellular tissue.[35] And *abscess* is a cavity filled with pus and delimited from the surrounding tissues by a pyogenic membrane.[35] In this particular case the

term *phlegmon* was used to describe the spread of the purulent process up to two anatomical regions.

In general, based on our literature search, the transcutaneous removal of submandibular gland sialoliths was reported in 4 cases. And our case became the fifth one. More than rare case of cutaneous exfoliation of the submandibular gland sialolith was recorded Karengera et al (1998).[21] Very unusual two cases of the submandibular salivary stones migration in the cutaneous direction was described by Drage et al (2005).[13] In 2021, Wakoh et al also highlighted the submandibular gland calculus migration to subcutaneous tissue.[7] Calculi removal in those three cases[7,13] was similar with our case in which the gland was preserved.

CONCLUSIONS

Ultrasound imaging is still underestimated and not enough popularized among head and neck and oral and maxillofacial surgeons. Presented case and published reports show the usefulness of this constantly developing diagnostic technique in a combination with knowledge of possible extraoral purulent complications' and its management.

PATIENT CONSENT

The patient provided written consent for the use of his images.

AUTHOR CONTRIBUTIONS

Conceptualization: Fesenko II. Data acquisition: Savchuk LA. Data analysis, interpretation, and drafting of the manuscript: Fesenko II. Critical revision of the manuscript: Savchuk LA, Fesenko II. Approval of the final version of the manuscript: both authors.

ACKNOWLEDGEMENTS

We would like to thank Professor Henry T. Hoffman, MD, Iowa City, Iowa, United States for the kind support in preparing the manuscript.

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