

Review Article

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Foreign bodies of submandibular gland and Wharton's duct: a review of literature

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ABSTRACT

Foreign bodies of submandibular duct are rarely but consistently reported in literature. They usually present as obstructive submandibular sialadenitis. The diagnosis can be challenging because of rarity of such an event, and inability of traditional diagnostic methods to detect the foreign body. We sought to assemble the previous published literature to delineate the presentation, investigation and management of submandibular gland foreign bodies. We performed a comprehensive systematic literature review of PubMed, and Google Scholar, databases from 1960 to 2019, and analyzed the case reports and research articles proclaiming detection of foreign body in the submandibular gland or its duct. We also included a case found in our hospital in the review. A total of 28 articles full filled our criteria. The earliest article found was from year 1962 and latest in 2019, amounting to 28 research articles on this topic in last 58 years. The age ranged from 9 years to 78 years, with average age of 38.7 year. The left Submandibular gland was involved in 18 cases (75%), and right in 6 cases (25%). There was a varied array of foreign bodies retrieved. Patients underwent sialadenectomy, duct excision, intervention by milking, probing or with a forceps or more recently interventional sialendoscopy. To approach an early diagnosis, a patient history needs to be believed. High resolution oral ultrasonography with interventional sialendoscopy can become the hallmark of a future approach to submandibular duct foreign bodies.

Keywords: Wharton's duct, Foreign body, Salivary gland, Submandibular gland, Submandibular gland duct

INTRODUCTION

Obstructive submandibular sialadenitis (OSS) is a common disease characterized by obstruction of saliva secretion in the submandibular gland. It may be due to endogenous causes like calculi, fibro-mucinous plugs, stenosis or malformations of the duct system, leading to mechanical obstruction with associated stasis and subsequent infection. The disease can manifest as a sudden onset of painful swelling mostly related to a meal, often unilateral and can become chronic.^{1,2} Exogenous causes, such as foreign bodies, are relatively uncommon. A number of anatomic and physiological barriers like a small calibrated punctum, a rather mobile distal end, and constant flow of saliva through a near horizontal course

act as a defense against retrograde intrusion of a foreign body into the duct.³ Despite these protective features, patients with retrograde passage of foreign bodies into the submandibular duct have been consistently seen. There are two mechanisms for such an entry: penetrating trauma and a retrograde migration.⁴⁻⁷ An early diagnosis of a foreign body is particularly challenging and more often the diagnosis is retrospective following organ resection. Traditional diagnostic tools like plain radiographs (occlusal film), sialography, ultrasonography (USG), are often not clearly able to identify the internal duct features and site of obstruction.⁸ To date, a more definitive analysis of presentation and management of submandibular duct foreign bodies has been limited by the small size of published series. Due to the relative

rarity of these foreign bodies, the published literature has been limited to case reports and small case reviews, precluding larger analysis. The current study seeks to systematically assemble previously published evidence to more clearly delineate the presentation, investigation and management of submandibular duct foreign bodies. We anticipate that the conclusions of this report will help identify the suspected cases early and perhaps allow for a guided management.

METHODS

A comprehensive systematic literature review of PubMed, and Google Scholar, databases was performed with the help of a library specialist to identify all published data that proclaimed the finding of foreign body in the floor of mouth, Wharton's duct, or submandibular gland or sialadenitis caused by a Sialolith with a suspicious foreign body nidus, with a limit to the English language literature. The references of the found articles and all the related articles were also crosschecked to ensure that no printed manuscript was missed. We reviewed the literature that included case reports, case series and research articles from 1960 to 2019 using a dual-prong search designed to capitalize on MeSH terms, subheadings, and keywords. The titles and abstracts of retrieved articles were reviewed and appropriate studies were selected based on inclusion and exclusion criteria. To be included in the systematic review, studies had to contain documented evidence of a foreign body within submandibular gland or its duct, information on the characteristics of the foreign body, details of the investigations received and details of the procedure undergone to retrieve the foreign body. If an article found lesser information but a confirmed foreign body retrieval, it was included rather than ignored. The articles which included foreign body retrieval from parotid duct, or floor of mouth were excluded. Data was extracted from

individual articles and entered into standardized Excel worksheet. Clinical data utilized for subsequent analysis included demographic information, presenting signs and symptoms, any underlying medical illnesses, duration of symptoms, investigations ordered, intervention done, nature of foreign body retrieved, whether found as an identifiable foreign body or a nidus of a sialolith, and dimensions of the foreign body. We included in the list, the details of a submandibular duct foreign body seen in our institution, SMHS Hospital, a tertiary care hospital in Srinagar, Kashmir.

SYSTEMATIC REVIEW

The review included a total of 50 articles of which 28 research articles fulfilled the inclusion criterion and were included for further analysis. Twenty-three research articles were pure case reports, three were case reports with an additional review and two research papers.^{7,9,10} The earliest article found was from year 1962 and latest in 2019, amounting to just 28 research articles on this topic in last 58 years. The two research papers included 13 cases and 5 cases each, in which, all the patients had undergone sialendoscopies for retrieval of Stones, when a fish bone foreign body was found as a nidus.^{5,11}

PATIENT DEMOGRAPHICS AND CLINICAL PROFILE

Among the 26 case reports analyzed, the age ranged from 9 years to 78 years, with average age of 38.7 year. The male: female ratio was 1:1. Among 24 cases of available data, the left submandibular gland was involved in 18 cases (75%), and right in 6 cases (25%). There was a varied array of foreign bodies retrieved as detailed in Table 1. The size of foreign body retrieved ranged from 1 mm to 35 mm. The clinical details and interventions done is described in Table 2.

Table 1: Details of foreign body.

Year	Type of foreign body	Size (mm)	Retrieved as	Location
1962 ¹²	Sliver of fingernail nail	5	FB	Duct
1967 ²⁵	Grass blade	10	FB	Duct
1968 ¹⁰	Pin needle, piece of broom, piece of thorn	NA	-	Duct
1969 ⁴⁵	Fish bone	NA	-	
1970 ¹⁵	Hair brush bristle	NA	FB	Gland
1972 ²⁶	Dried grass	20	FB	Duct
1972 ²⁶	Vegetable fibre	NA		Gland,
1975 ⁴³	Fingernail	NA	FB	Duct
1982 ¹⁶	Grass blade	20	FB	Duct
1988 ²⁷	Fibres of sunflower seeds	NA	FB	Duct
1990 ²⁸	Fish bone	10	FB	Duct
2001 ²⁹	Vegetate fibres	NA	Nidus	Duct
2002 ³⁰	Feather of pet bird	NA	FB	Duct
2003 ³¹	Shrapnel stuck 57 years ago	1	Nidus	Duct
2005 ³³	Wooden splinter	35	FB	Duct
2005 ³²	Thorn	7	Nidus	Duct
2009 ⁸	Hair	NA	FB	Duct

Continued.

Year	Type of foreign body	Size (mm)	Retrieved as	Location
2009 ⁴⁴	Fish bone	NA	FB	
2011 ³⁶	Silver of fingernail	5	FB	Duct
2012 ¹⁷	Fishbone	13	FB	Duct
2013 ³⁷	Linear seed	5	FB	Duct
2014 ⁵	Facial hair strand	5	Nidus	Duct
2015 ⁶	Fish bone	5	Nidus	Duct
2016 ⁴⁰	Wood splinter for 3 years	20	FB	Duct
2017 ⁴¹	Fish bone 3 months	20	FB	Gland
2018 ⁴²	Metallic wire	20	FB	Duct
2019 ⁹	Fish bone	12	FB	Duct
2019	Thorn	15	FB	Duct

FB=Foreign body.

Table 2: Clinical profile.

Year	Symptom duration	Investigation	Intervention	Special comments
1962 ¹²	6 D	XR	DE	History of loss of nail in mouth was ignored.
1967 ²⁵	8 D	PR, XR	SA	Retrospectively, history of habitual grass chewing
1968 ¹⁰		EX		
1969 ³⁵	0	0	0	
1970 ¹⁵	14 D	PR, SG	SA	Retrospectively, history of cleaning teeth with brush bristles.
1972 ²⁶	4 M	SG	SA	Diet contains dried grassy plant
1972 ²⁶	1 M	SG	SA	Diet contained grassy plant
1975 ⁴³	10 D	XR	EX	
1982 ¹⁶	1 M	XR	SA	History of loss of grass below tongue
1988 ²⁷	8 M	XR, FG	SA	Retrospective history of pain in submandibular region while eating sunflower seeds.
1990 ²⁸	36 M	XR, SG	SA	No acute presentation noted
2001 ²⁹	Acute	SG, DS	SA	
2002 ³⁰	Acute	CE	EX	Patient had a habit of nuzzling her cockatoo and sucking its feathers.
2003 ³¹	2 M	XR	PR	Patient was hit by a shrapnel 57 years ago
2005 ³³	18 M	XR	SA	Retrospectively, history of in ingestion of same wooden splinter
2005 ³²	24 M	XR	DE	Retrograde, history of thorn ingestion
2009 ⁸	12 M	SG, USG	IS	First case done with interventional Sialendoscopy
2009 ⁴⁴	0	XR	SA	
2011 ³⁶	Acute	XR	DE	Reasoned why FB don't decompose
2012 ¹⁷	6 M	XR, USG	IS	Proposed sialendoscopy as a good alternative
2013 ³⁷	2 M	USG, MRI	SA	
2014 ⁵	6 D	CE	DE	Retrospectively, history of facial hair getting stuck in mouth
2015 ⁶	6 M	USG	DE	Fish diet, no history of prick
2016 ⁴⁰	12 M	USG, CT, MRI	SA	3 year back patient had maxillofacial trauma
2017 ⁴¹	2 M	XR, USG	SA	History of fish bone got stuck in floor of mouth and disappeared following drinking water
2018 ⁴²	10 M	XR, CT	SA	Retrospectively, remembered having broken a wire in mouth
2019 ⁹	14 D	XR, MK		
2019	5 D	HR-USG	MK	History of loss of thorn under tongue

D=days, M=months, XR=X ray, USG=ultrasonography, CT=computed tomography, MRI=magnetic resonance imaging, CE=clinical examination, HR USG=high resolution ultrasonography, PR=probing, SA=sialadenectomy, DE=duct excision MK=milking, SG=sialography, DS=diagnostic sialendoscopy, FG=fistulogram, IS=interventional sialendoscopy, EX=spontaneous extrusion

The duration of symptoms ranged from 5 days to 36 months. The duration of foreign body could not be found in most cases, with only 5 cases remembering the duration of an exact event. A confirmed history of loss of a foreign body in mouth or under tongue was noted in 4 cases but was conveniently ignored in all cases, owing to rarity of such a proposition. A preoperative confirmation of a foreign body by clinical examination and investigations was obtained in 6 cases (23%). In 5 cases the foreign body was present as a nidus inside a sialolith, while in 21 cases a foreign body could be identified by naked eye or confirmed by microscopy. Of the total reported 26 cases, the locations of the foreign bodies were 22 in the main duct (Wharton duct), 02 in the intraglandular duct, 01 penetrating the gland, and the remainder not clearly mentioned. A varied range of investigations were performed which included X-ray, probing, sialogram, USG, sialendoscopy, CT scan, and magnetic resonance imaging (MRI). In most cases the pre-op investigations did not prove helpful, and retrieval of a foreign body instead of a suspected stone, was a surprise plainly accepted. Preoperatively foreign body was confirmed by X-ray in 2 cases, sialendoscopy 1, and USG of gland in 1 case and high-resolution USG of floor of mouth in 1 patient. Fourteen (48%) of the patients underwent Sialadenectomy, 6 (20.6%) duct excision under LA, 7 (24.13%) foreign bodies were removed without surgical intervention by milking, probing or with a forceps, and 2 (6.8%) foreign bodies were removed by utilizing a sialendoscope.

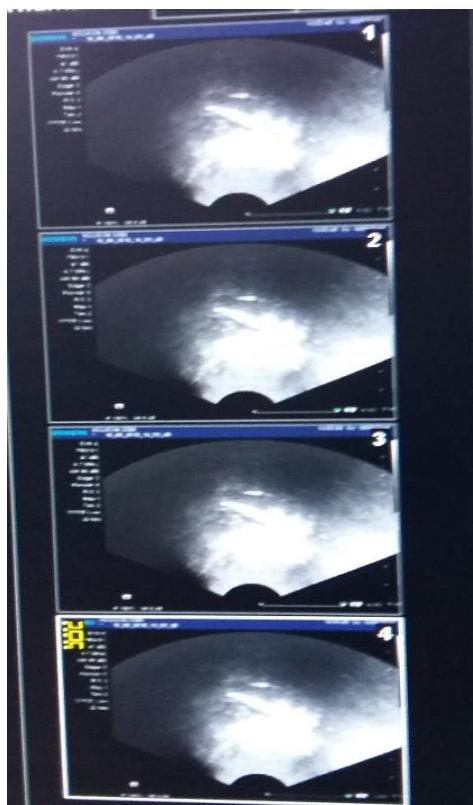


Figure 1: High resolution USG floor of mouth showing linear foreign body.

DETAILS OF CASE FOUND

A 20 year old lady presented with 5 days history of loss of a thorn under tongue followed by pain and swelling in left submandibular region. Clinical examination, X-ray, and USG gland proved inconclusive and patient was managed as acute sialadenitis. Next day due to insistence of rather enthusiastic resident, a high-resolution USG of floor of mouth was done, which confirmed a linear foreign body in Wharton's duct near to gland (Figure 1). Within 2 days of a dilemma of how to approach, the lady noted sensation under tongue following which a 1.5 cm long thorn was retrieved by milking the duct (Figure 2 and 3). Patient improved dramatically and was successfully followed.



Figure 2: Foreign body being milked out.



Figure 3: Thorn foreign body.

DISCUSSION

Foreign body of the submandibular gland has always been labelled as rare. However, overtime, a slow trickle of evidence has gathered in literature. An earliest available review of medical literature was done by Walker who reported 5 cases with earliest report found was in 1923 by Baggio.^{12,13} Many surgeons may find it difficult to believe that a foreign body could enter the submandibular duct accidentally. As challenging as a probing of Wharton's duct may sound, foreign bodies do find their place into the small orifice as is proven by these publications. Two reasons and hypotheses are considered for entering foreign bodies into the salivary gland: one of them is penetrating trauma and the other reason is retrograde migration.⁴⁻⁷ Mead as early as 1940 stated that it is possible for foreign objects to enter the orifice of Wharton's duct and partially occlude it.¹⁴ Prett et al hypothesized that due to location on floor of mouth, foreign bodies can be carried down into the duct by force of gravity.¹⁰ However, Ricciro et al tried to disprove the notion explaining the improbability of such an event by scientific reasoning like a small punctum of Wharton's duct, the relative mobility of its distal end and presence of continuous flow of saliva from the ducts.³ Smith et al supported this notion and proposed that a mucosal rather than a retrograde entry might have been a cause in previous reported cases also.¹⁵ Twelve years later, Watkins observed that even a positive history is more probable to be ignored rather than believed adding that both his previous similar reports included detailed and accurate histories that a piece of grass had been lost below the tongue just before the symptoms started.¹⁶ He added that each time there was considerable delay in diagnosis and appropriate treatment, in one patient it was 12 years. He opined that the delay was probably because of difficulty in accepting the patient's history.

A foreign body within the submandibular gland duct mostly presents as obstructive sialadenitis with features masquerading as acute or chronic sialadenitis. Obstructive sialadenitis is the most frequent non-neoplastic salivary disorder. It can be due to calculi, strictures and kinks of the ductal system, mucous plugs and rarely foreign bodies.¹⁷ Such a foreign body can be accidental with awareness of event or totally uneventful like eating fish or sea food.¹¹ If a foreign body does enter through the ductal orifice, it might either cause acute obstruction or infection or be asymptomatic for years before obstruction appears. Marchal et al in one of their theories of lithogenesis have proposed a retrograde migration of foods, bacteria, or foreign bodies from the oral cavity to the duct system can act as a nidus for a sialolith.¹ There are many reports of foreign body as a nidus for a sialolith.^{5,11}

It is very difficult to make the correct diagnosis with the traditional diagnostic strategies. Over a period, various modalities have been tried. X ray is usually ineffective unless the foreign body is radiopaque or has calcified into

a radiopaque stone. Sialogram and probing of duct have been employed and can be suggestive of an obstruction or filling defect at best. In recent years ultrasound of submandibular gland, or CT scan or MRI have been utilized.^{18,19} Chuangqi et al mentioned that magnetic resonance sialography is a new method of diagnosing anomalies in the duct; it provides 2 or 3-dimensional images of the salivary gland without contrast medium or excessive exposure to radiation.¹¹ It also provides evidence of the shape of the sialolith in the hilus in its natural state. Contemporary, innovative, small high resolution ultrasound probes allow access to the ducts in both the submandibular and parotid glands via an intraoral approach, and together with extra-oral sonography can become alternatives to conventional and magnetic resonance (MR) sialography.^{21,22} Brown et al presented a case showing the first reported use of a small, digitally guided, intra-oral ultrasound probe to image a calculus in a submandibular salivary duct.²³ The use of high resolution ultrasound was not found in any study in our current review except utilized on the patient in our hospital. Such ultrasound apparatus with thin probe are rather routinely used for performing trans-vaginal ultrasound and can be equally utilized for floor of mouth as proved by our case. Such a modality may be used more often to note ductal stones or a foreign body if transcutaneous USG is negative.

However, these imaging techniques are only an indirect visualization, can be expensive and time consuming and expose patients to unnecessary radiation. In 5%-10% of recurrent swelling of the salivary glands, the etiology cannot be identified clearly.⁸ In our review, only five cases had been diagnosed preoperatively. The treatment has traditionally been a sialadenectomy or a duct excision but in recent years, sialendoscopy has offered a promising approach in diagnosing and managing intraductal pathologies avoiding much discomfort and unnecessary sialadenectomies.¹⁷ Sialendoscopy was introduced in the 1990s as an endoscopic technique and since then has been greatly refined, to be utilized to completely explore the ductal system and provide an insight into what is happening within the duct.^{11,24} Sialendoscopy is also effective in removing sialoliths, mucous plugs as well as dealing with strictures and kinks of the ductal system and can very well be utilized for removing foreign bodies as was done in 2 cases identified in our review.^{5,17}

Nonetheless, due to expertise requirement and cost of instrument not all centers can afford such a facility and conventional techniques like meticulous examination, retrograde milking, or duct excision and at times, sialadenectomy may be the only available alternative.

CONCLUSION

We conclude that submandibular gland foreign bodies are rarely but consistently reported over the years. To approach an early diagnosis, a patient history, if

suggestive needs to be believed, followed by meticulous examination. One has to maintain a degree of suspicion of an intra-ductal pathology including a foreign body, in case of a stone negative acute or recurrent sialadenitis. High resolution oral USG with interventional sialendoscopy is the hallmark of a future approach to Submandibular duct foreign bodies.

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