Review Article

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

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Received: 23 December 2019
Accepted: 26 February 2020

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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 puctum, 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 article 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. 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.

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 | Silver of fingernail nail    | 5         | FB           | Duct     |
| 1967 | Grass blade                  | 10        | FB           | Duct     |
| 1968 | Pin needle, piece of broom    | NA        | -            | Duct     |
| 1969 | Fish bone                    | NA        | -            |          |
| 1970 | Hair brush bristle           | NA        | FB           | Gland    |
| 1972 | Dried grass                  | 20        | FB           | Duct     |
| 1975 | Vegetable fibre              | NA        | FB           | Gland,   |
| 1982 | Grass blade                  | 20        | FB           | Duct     |
| 1988 | Fibres of sunflower seeds    | NA        | FB           | Duct     |
| 1990 | Fish bone                    | 10        | FB           | Duct     |
| 2001 | Vegetate fibres              | NA        | Nidus        | Duct     |
| 2002 | Feather of pet bird          | NA        | FB           | Duct     |
| 2003 | Shrapnel stuck 57 years ago   | 1         | Nidus        | Duct     |
| 2005 | Wooden splinter              | 35        | FB           | Duct     |
| 2005 | Thorn                        | 7         | Nidus        | Duct     |
| 2009 | Hair                         | NA        | FB           | Duct     |

Continued.
| Year | Type of foreign body              | Size (mm) | Retrieved as | Location   |
|------|----------------------------------|-----------|--------------|------------|
| 2009 | Fish bone                        | NA        | FB           | Duct       |
| 2011 | Silver of fingernail             | 5         | FB           | Duct       |
| 2012 | Fishbone                         | 13        | FB           | Duct       |
| 2013 | Linear seed                      | 5         | FB           | Duct       |
| 2014 | Facial hair strand               | 5         | Nidus        | Duct       |
| 2015 | Fish bone                        | 5         | Nidus        | Duct       |
| 2016 | Wood splinter for 3 years        | 20        | FB           | Duct       |
| 2017 | Fish bone 3 months               | 20        | FB           | Gland      |
| 2018 | Metallic wire                    | 20        | FB           | Duct       |
| 2019 | 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 | 6 D              | XR            | DE           | History of loss of nail in mouth was ignored.                                      |
| 1967 | 8 D              | PR, XR        | SA           | Retrospectively, history of habitual grass chewing                                  |
| 1968 | 0                |               | EX           |                                                                                   |
| 1969 | 0                |               | 0            |                                                                                   |
| 1970 | 14 D             | PR, SG        | SA           | Retrospectively, history of cleaning teeth with brush bristles.                    |
| 1972 | 4 M              | SG            | SA           | Diet contains dried grassy plant                                                  |
| 1972 | 1 M              | SG            | SA           | Diet contained grassy plant                                                       |
| 1975 | 10 D             | XR            | EX           |                                                                                   |
| 1982 | 1 M              | XR            | SA           | History of loss of grass below tongue                                              |
| 1988 | 8 M              | XR, FG        | SA           | Retrospective history of pain in submandibular region while eating sunflower seeds.|
| 1990 | 36 M             | XR, SG        | SA           | No acute presentation noted                                                        |
| 2001 | Acute            | SG, DS        | SA           |                                                                                   |
| 2002 | Acute            | CE            | EX           | Patient had a habit of nuzzling her cockatoo and sucking its feathers.             |
| 2003 | 2 M              | XR            | PR           | Patient was hit by a shrapnel 57 years ago                                        |
| 2005 | 18 M             | XR            | SA           | Retrospectively, history of ingestion of same wooden splinter                      |
| 2005 | 24 M             | XR            | DE           | Retrograde, history of thorn ingestion                                            |
| 2009 | 12 M             | SG, USG       | IS           | First case done with interventional Sialendoscopy                                |
| 2009 | 0                | XR            | SA           |                                                                                   |
| 2011 | Acute            | XR            | DE           | Reasoned why FB don’t decompose                                                   |
| 2012 | 6 M              | XR, USG       | IS           | Proposed sialendoscopy as a good alternative                                       |
| 2013 | 2 M              | USG, MRI      | SA           |                                                                                   |
| 2014 | 6 D              | CE            | DE           | Retrospectively, history of facial hair getting stuck in mouth                     |
| 2015 | 6 M              | USG           | DE           | Fish diet, no history of prick                                                     |
| 2016 | 12 M             | USG, CT, MRI  | SA           | 3 year back patient had maxillofacial trauma                                       |
| 2017 | 2 M              | XR, USG       | SA           | History of fish bone got stuck in floor of mouth and disappeared following drinking water |
| 2018 | 10 M             | XR, CT        | SA           | Retrospectively, remembered having broken a wire in mouth                          |
| 2019 | 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=diaagnostic 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.

 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 1: High resolution USG floor of mouth showing linear foreign body.](image1)

![Figure 2: Foreign body being milked out.](image2)

![Figure 3: Thorn foreign body.](image3)
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.5,7 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, Riccirco 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.7 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 asymptotic for years before obstruction appears. Marchal et al in one 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.6 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 intra-ductal 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.5,37

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 axial USG with interventional sialendoscopy is the hallmark of a future approach to Submandibular duct foreign bodies.

**Funding:** No funding sources

**Conflict of interest:** None declared

**Ethical approval:** Not required

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Cite this article as: Dar WQ, Nisar J, Batool QUA, Quzi SM. Foreign bodies of submandibular gland and Wharton’s duct: a review of literature. Int J Otorhinolaryngol Head Neck Surg 2020;6:803-9.