UNUSUAL SPONTANEOUS SALIVARY FISTULA: A RETROSPECTIVE REVIEW OF THE LAST 12 YEARS

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ABSTRACT

***Aims**: To analyse retrospectively all cases of spontaneous salivary fistulae studied by radiological imaging from January 1999 to December 2011. Salivary fistulae are uncommon; they could be congenital or acquired caused by injury, neoplasm, surgery or may represent a rare long-term complication of sialolithiasis and/or recurrent sialoadenitis.*

***Materials and methods**: We retrospectively report our 12 years experience in patients with proved unusual intraoral spontaneous salivary fistula. All ours patients were referred for classical symptoms as recurrent salivary infection and purulent oral discharge.*

***Results**: 6 cases of intraoral inflammatory salivary fistulae were studied (5 submandibular, 1 parotid), 3 cases were related to sialoliths and other 3 to sialoadenitis; among them 2 have both sialoliths and active adenitis. All patients underwent to sialography and fistulography and all fistulous tracts were detected. Different diagnostic options were applied according to the localization of fistulae.*

***Conclusion**: Looking to our experience we suggest studying patients with sialography and/or fistulography and Magnetic Resonance (MR) especially for patients with a high probability of being subjected to major surgery. MR allows to see fistulae, relation between fistulae and near tissue, and to distinguish inflammation from other pathology. Computed Tomography (CT) is ideal to study shape and dimension of sialolith and to evaluate eventual bone involvement.*

***Key words**: salivary gland, fistula, sialography, fistulography.*

***Received** February 18, 2014; **Accepted** June 19, 2014*

Introduction

Fistulae are usually due to: traumatic injury, malignant neoplasm, secondary to surgery and rare complication of recurrent inflammations involving the gland regions, or congenital. Non-traumatic salivary fistula is an uncommon complication of salivary stones, or infection and strictures.

Trauma is the most common cause of fistula in parotid gland, for its superficial location, followed by malignancy, operative complication and infection[1].

Traumatic fistulae are rare also in the submandibular salivary glands[2].

Long-term sialolithias with recurrent sialodochitis and/or sialoadenitis may infrequently cause spontaneous elimination of the sialoliths or opening of "new ductal course" or fistula.

Lithiasis affects (90% of cases) the submandibular gland and less frequently (10% of cases) the parotid gland, so fistula may shows the same prevalence and localization looking also to the anatomical thinness of the Wharton duct's wall[3,4].

Even if stones are more frequent in the proximal part of main salivary duct, perforation of the floor of the mouth often happens in the anterior part, in this region the mucosa is thinner, so it could ulcerate more easily, than posterior region and allow fistulous communication to the oral floor[3].

On the one hand fistula may temporarily resolve the colic and eventually the infection but on the other hand may frequently cause recurrence of some infections for the loss of the anatomical function of the ostial orifice and necessitate surgical revision. Due to the anatomical complexity of the

salivary gland regions a detailed imaging assessment of the fistula, the communications with the salivary ducts and the tissue involved are mandatory in order to avoid recurrence of vascular and nerve injury.

Fistulous communication between the posterior portion of the duct and the floor of the mouth is a very rare condition.

There are few cases reported in literature; most of them are reports of single clinical presentations.

The aim of this work is to present the different imaging characteristics of unusual non-traumatic spontaneous salivary fistulae met in 12 years in our hospital.

Materials and methods

Between January 1999 and December 2011, 6 patients (5 women, 1 man; average 52,4 years old) were referred to the Radiology Department from the Maxillofacial Unit and Ear Noose and Throat (ENT) Department for history of recurrent salivary gland infections and purulent discharge in the oral cavity, suspected for salivary fistula. 4/6 patients referred a long history of salivary colic and multiple infections treated with antibiotics but they had refused imaging investigation or surgical removal of calculi (patient with parotid fistula and 3/5 patients with submandibular fistula). 2/6 referred a single episode of salivary colic, lasted in one case, more than 3 days and resolved after a plentiful secretion of saliva and a salivary stone; the other one referred self-made manipulation of the stone that was pulled out through the fistula with the aid of a scissor.

To detect the cause of the recurrent infections the patients were submitted to sialography (2/6 patients), fistulography (4/6 patient), and sialo-Computed Tomography (CT) (2/6 patients) and magnetic resonance (MR) (2/6 patients).

Looking retrospectively to a long period of time, the imaging study differs for methodology and performed exams and it unfortunately cannot be standardized.

Sialography was performed after cannulation of the Stensen's duct or the Wharton duct, when recognizable, with Rabinov catheter or a 24G cannula or olive-tipped cannula (William Cook Europe, Bjaeverskov, Denmark) and retrograde injection of iodinate non ionic, dimeric, hydrosolubile contrast medium (Iodixanolo, Visipaque 270, Nycomed, Amersham Health, Norway). Images were acquired using a C arch apparatus (Eurocolumbus TR3D, Milano, Italy digitalized from 2000). The injection was made manually with a 2.5 ml syringe during non subtractive or subtractive acquisition, in the recent period of 2 images per second, and stopped when complete opacification of the ductal system and eventually the fistula, was obtained. When the duct orifice was not recognizable or when it was of abnormal calibre, it was explored after gland stimulation of the oral cavity by using optical magnification devices along the course of the duct and all salivary secreting holes were probed and eventually cannulated by using different size of Rabinov catheter, vascular dilator, paediatric Foley balloon catheter and olive tipped cannula. All sialograms and fistulograms, were performed by two experienced radiologists (S.S and A.L.C.).

Sialo-CT was performed with a GE HI Speed dual (GE healthcare Wisconsin USA since 2004) and GE Bright speed 16 (GE healthcare Wisconsin USA from 2004) after cannulation either of the duct of the fistula and retrograde injection of contrast media using 1 mm thickness.

MR was performed using a 1.5 T (Picker Eclipse MR Cleveland USA) with Multiplanar SE T1 before and after paramagnetic i.v. contrast medium- FSE T2 and FIR sequences.

A 5 antibiotics therapy (1 gr. Spyramicin/day) was prescribed after the radiological procedure in all patients[5].

Obviously in this retrospective review collecting cases from over two decades it is unreasonable to assume that this diagnostic paradigm would be followed in every case.

Results

Our study included 5 fistulae of submandibular gland and a parotid gland one.

All submandibular glands fistulae were detected in the posterior part of floor of the mouth, while the parotid one was detected in the anterior part.

5/6 fistulae were associated with salivary glands infection (Fig. 1); 3/5 submandibular fistulae were associated to single and multiple sialolithiasis (Fig. 2); 1 submandibular fistula was discovered few months from surgical excision of the salivary gland and was misdiagnosed at the time of the surgical approach (Fig. 3). The patient made a lateral trans cervical excision, the skin incision was made 2 centimetres from the mandible lower border to

avoid lesions of the mandibular branch of the facial nerve, this classical access allows to minimizing postoperative complication such as hematoma and infection, even if there is a risk of xerostomia[6].

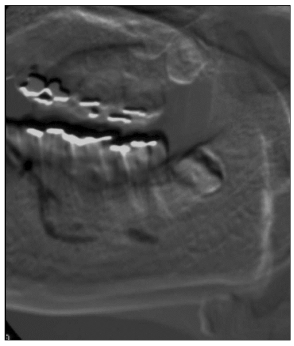

Fig. 1: Anterior submandibular fistula. Sialography, lateral image during a cannulation of the Wharton's duct a second contrast opacification of the fistulous course is detected (arrow).

The 5 inflammatory fistulae were related to sialoduct-adenitis, two of these, the parotid fistula were related to a premasseter space abscess (fig. 4-5).

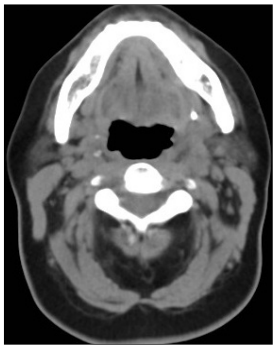

Fig. 2: Anterior submandibular fistula with stone demonstration on CT, axial image.

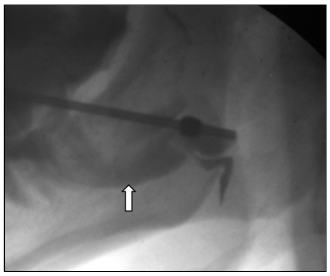

Fig. 3: Posterior submandibular fistula. Fistulography, lateral view of the retrograde opacification of Wharton duct through the fistula (white arrow).

Fig. 4: Parotid fistula with a premasseter space collection opening in the buccal space. Sialography with image subtraction, oblique view.

No anatomical anomalies in the main salivary duct were found in any patients after sialography.

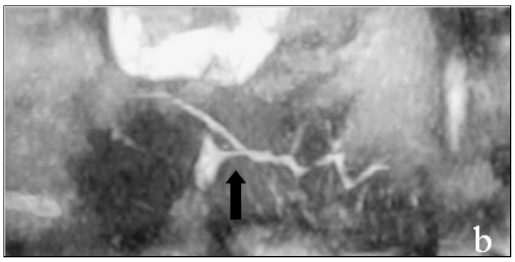

Fig. 5: Parotid fistula with a premasseter space collection opening in the buccal space. MR sialogram.

Discussion

Salivary calculi are common and are found in 1.2 % of population at necroscopy[7]. Salivary fistulas are uncommon entities and the great majority are due to traumatic injury; more rarely are congenital, caused by calculi or foreign bodies[8,9,10].

Perforation of the floor of the mouth often occurs in the anterior ductal calculi, while it is very infrequent in the posterior ductal and intraglandular lithiasis[7]. So preferential localization of the submandibular fistula is the oral floor, while parotid fistulas are often cutaneous[11].

Most of the fistulas are case reports of single clinical presentations, a retrospective analysis of these case reports both intraoral and cutaneous salivary fistula shows non-unique management of these patients[1-2, 10-17].

When a cutaneous salivary discharge is discovered by biochemical verification of saliva-amylase in the secretion, first step of its management (more often cervical discharge) is sialography if this imaging does not show fistula or when it is not possible to identify and to cannulate the ductal orifice fistulography is the next step[11-15].

In literature three cases of inflammatory salivary cutaneous fistula MR were performed by T2 weighted sequences allowing discovery and study of the fistula in two patients[12,13] and in the other patient the chronic inflammatory glandular changes and fibrosis[14].

Acute inflammatory processes of submandibular glands with stones may lead to posterior perforation of the floor of mouth, but they usually don't result in fistula formation[10].

Management of inflammatory intraoral salivary fistula is similar, but most frequently it stops at the detection of sialolithis, because these patients are often submitted to local surgery for stone retrieval and associated medical therapy, not require

often complete gland excision. So they often don't require a regional study with CT and/or MR that are usually required for surgical planning[10,16-17].

History of chronic sialoadenitis was present in 4/6 patients. Sialography and fistulography managed to recognize all the fistulous tracts.

CT fistulography may support as described the sialography and fistulography in cases of accessory parotid gland with ectopic fistulous duct[18].

Sialo-CT or sialo-MR was performed to detect the anatomic relation between fistula and the nearest structures for surgical management.

Our management varies due to the long time of the retrospective review. In fact we submitted patients to different imaging technique, this heterogeneity is due to the long term of our study and the consequent technological evolution, so the imaging data and techniques used cannot be standardized.

Concluding, to value the complexity of the fistula and its course different type of imaging modalities may be required, this is crucial to direct patient to elective surgery and reduction of complication and recurrence[19].

Analysing our past cases and literature our current approach to any study of patients with inflammatory salivary fistulas is the sialography and/or fistulography; sialography allows to recognize duct inflammation and sialoliths too[20]. Due to availability we suggest for intraoral and cutaneous fistula sialography and or fistulography to detect the fistula and MRI with sequence FAT SAT T2 and STIR sequences to emphasise the fistula visualization and define the relationship between the fistula and the nearest structures especially when major surgery is required. CT can be performed for direct visualization of the stone shape and diameter and eventual bone involvement (mandibular or ear structures) because calcium is not well detected on MR or for study eventual complications (abscesses) especially in emergency.

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