SPONTANEOUS SUBMANDIBULAR CUTANEOUS FISTULA DUE TO SIALOLITH EXTRUSION: A RARE PRESENTATION

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ABSTRACT

Cutaneous submandibular gland fistulae due to chronic sialadenitis are rare. We present one such case in which the patient presented with a spontaneous cutaneous fistula of the submandibular gland secondary to recurrent episodes of sialadenitis and expulsion of a calculus. We discuss the importance of early treatment of chronic sialadenitis with sialolithiasis.

Key words: *Salivary glands; fistula; sialolithiasis; sialadenitis; submandibular.*

INTRODUCTION

Fistulae of the submandibular gland are uncommon entities[1]. These may communicate with the oral cavity, oropharynx or externally through the skin[1,2,3]. Etiologically, most of these fistulae arise as a result of direct trauma[4]. Other causes for these fistulae include foreign bodies[5], salivary calculi[2,3] and congenital malformations[1]. Most of the eliminations of submandibular sialoliths occur through the opening of the Wharton's duct in the oral cavity and transcutaneous expulsion of submandibular calculi is extremely rare with a very few cases reported in the literature in the past [2,3]. The extreme rarity of such presentations prompted us to present this case.

CASE REPORT

A 46-year-old female presented to us with an opening in the left submandibular region of 4 weeks duration. The patient complained of clear discharge from the opening that was occasionally mixed with pus. The patient gave history of recurrent episodes (roughly once every 5 to 6 months) of abscess formation in that region since 5 to 6 years prior to the formation of the fistula. These abscesses used to burst spontaneously with pus discharge except on one occasion when she had an incision and drainage done from a private practitioner. The patient also gave history of expulsion of a hard, brownish, blood-stained 'stone' about 0.5cm.X 0.5cm. in size from the abscess just prior to formation of the fistula.

Examination revealed a firm and enlarged submandibular gland with a fistulous opening just below the gland (figure 1). There was no active discharge at the time of presentation. Per orally, no abnormality was

detected apart from a mild hyperemia of the openings of bilateral Wharton's ducts.

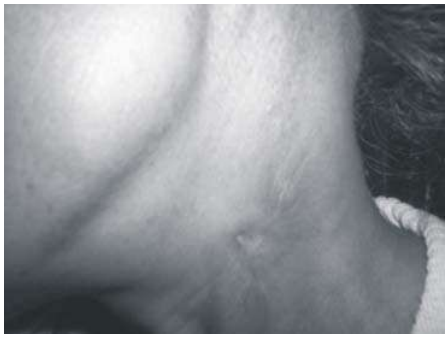

Fig. 1: Showing fistulous opening in left submandibular region.

The routine blood and urine investigations were within normal limits. A fistulogram was performed that revealed a fistulous tract communicating with the submandibular gland with normal dye flow up to the opening of the submandibular duct (figure 2). No filling defect suggestive of the presence of calculi in the gland or duct was identified. An FNAC of the submandibular gland was suggestive of chronic sialadenitis. The patient underwent excision of the submandibular gland along with the fistula under general anaesthesia. The histopathological evaluation of the excised gland was consistent with chronic sialadenitis. The patient was totally symptom-free when last seen one year postoperatively.

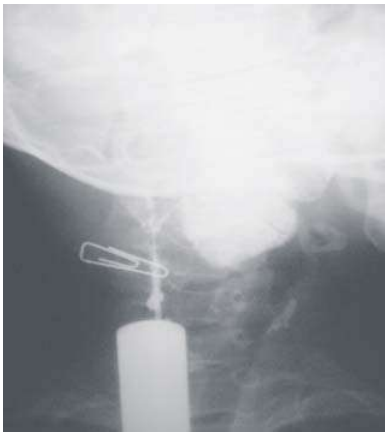

Fig. 2: Fistulogram showing cannulation of fistulous tract with the dye entering the left submandibular gland and duct.

DISCUSSION

Salivary calculi are most commonly seen in middle-aged men[2]. These calculi are more frequently seen in the submandibular glands as compared to the other salivary glands due to the difference in composition of secretions of these glands[3]. The main complication resulting from the presence of such a calculus is the chronic inflammation of the affected gland[6]. The patient may present with episodes of painful swelling in the region of affected submandibular gland. Long-standing infection may cause severe damage to the gland parenchyma producing a suppurative process. It may lead to fistula and pus drainage via the skin or mucosa[3,7]. Our patient presented in a similar manner with recurrent abscesses and formation of a cutaneous fistula.

Most of the symptomatic calculi are surgically removed. Other modalities in the form of shock wave lithotripsy may be used in selected cases[8]. The skin fistulae in relation to jaws are most commonly due to odontogenic infections[9]. Fistulae, as a result of sialolithiasis, are thus, extremely uncommon, yet very important from the management point of view. A fistulogram seems to be an extremely reliable investigation for their diagnosis. An early diagnosis of sialolithiasis and chronic sialadenitis may prevent a fistula formation. Unfortunately, this was not the case in our patient.

In summary, we would like to conclude that submandibular cutaneous fistula as a result of a sialolith expulsion is a rare occurrence and must be kept in the differential diagnosis of fistulae in this region. An early diagnosis of sialolithiasis and a prompt intervention may prevent the formation of such fistulae.

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