The Journal of Laryngology & Otology, E11, 1 of 4.

doi:10.1017/S0022215107001417

Online Only Clinical Record

Congenital salivary fistula of accessory parotid gland: imaging findings

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Abstract

We report the imaging findings in a rare case of an accessory parotid gland fistula.

Material and methods: An eight-year-old boy was presented with complaints of serous discharge from his left cheek since birth. As part of the radiological investigation, magnetic resonance imaging, computed tomography sialography with fistulography, and digital sialography with fistulography were performed.

Results: Magnetic resonance imaging demonstrated the exact location of an accessory parotid gland but failed to demonstrate the accessory duct. The presence of an accessory gland was well delineated on computed tomography fistulography and computed tomography sialography. Fistulography revealed a small accessory parotid duct and gland. No communication between the ductal systems of both glands was demonstrated.

Conclusions: In such cases, pre-operative imaging (with sialography, magnetic resonance sialography and computed tomography sialography with fistulography) is helpful for exact delineation of the ductal anatomy. To the best of our knowledge, only four previous cases of congenital accessory parotid gland fistula have been reported in the English literature.

Key words: Parotid Gland; Sialography; Computed Tomography; Fistula

Introduction

Congenital salivary fistulas are unusual entities which can arise from the parotid gland, submandibular gland, ectopic salivary glands or, even more infrequently, from accessory parotid glands.[1] To the best of our knowledge, only four cases of congenital fistula arising from an accessory parotid gland have previously been reported in the English literature.[1–4] In this article, we present the imaging findings in a case of congenital accessory parotid gland fistula.

Case report

An eight-year-old boy was presented with complaints of clear, serous discharge from his left cheek since birth, which increased during food intake. There was no history of trauma or surgery.

On physical examination, there was a punctate opening approximately 1.5 cm lateral to the left angle of the mouth, with no signs of cellulitis (Figure 1). A left periaural appendix was also present. Intraoral examination revealed bilateral, normal openings of the parotid and submandibular ducts.

Magnetic resonance imaging (MRI) and MR sialography of the parotid region were performed. The parotid and submandibular glands were normal bilaterally in location and signal intensity (Figure 2a). A small area of soft tissue with a signal intensity similar to that of the main parotid gland on T1- and T2-weighted images was seen along the lateral aspect of the left masseter muscle, suggestive of an accessory parotid gland (Figure 2b). On MR sialography, the main parotid duct was well visualised. However, the duct draining the accessory gland could not be visualised.

Computed tomography (CT) sialography and CT fistulography were performed to enable better delineation of the anatomy and presurgical planning. Noncontrast CT confirmed the location of an accessory parotid gland lateral to the left masseter muscle. Computed tomography sialography revealed a normal parotid duct and opacification of the main left parotid gland (Figure 3a). The accessory gland was not opacified. Computed tomography fistulography, performed after injecting contrast through the external opening, showed opacification of the accessory gland (Figure 3b). A duct draining the accessory parotid was not well visualised. No communication was demonstrated between the ductal systems of the main and accessory parotid glands.

Digital fistulography and sialography were performed in order to better demonstrate the ductal systems of both glands. Fistulography demonstrated a narrow tract passing posterosuperiorly and draining

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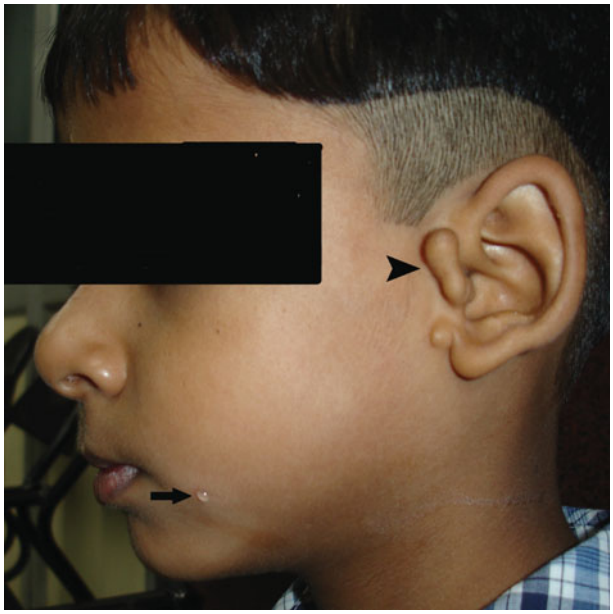

FIG. 1

Clinical photograph of eight-year-old boy, showing serous fluid discharge from a small, punctate opening located 1.5 cm lateral to the angle of the mouth (arrow). A periaural appendix is also present (arrowhead).

the accessory gland overlying the left hemimandible, anterior to the parotid fossa (Figure 4a). Sialography was performed after cannulating the left parotid duct, demonstrating a normal main parotid duct with a normal branching pattern. No communication with the fistulous tract was seen (Figure 4b).

A diagnosis of congenital accessory parotid duct fistula was made. At the time of writing, the patient was awaiting surgery.

Discussion

A salivary fistula is an abnormal communication between a surface and a salivary duct or gland, through which saliva is discharged. Salivary fistulas may be classified according to presumed aetiology into two general categories: congenital and acquired.[5] Acquired fistulas are more frequently seen and mostly result from surgery and trauma. Congenital salivary fistulas are thought to arise from aberrant development of the salivary gland and are extremely uncommon.[1,5]

Congenital salivary fistulas can be further classified according to the gland of origin (parotid gland, submandibular gland, ectopic salivary gland or accessory parotid gland). Most salivary fistulas are of parotid origin.[1] The reported sites of external opening include the retroauricular region, skin of the cheek, oral mucosa and skin of the cervical region.[1–8] Congenital fistulas arising from accessory parotid glands are still rarer, with only four reported cases.[1–4] The external opening of a parotid or accessory parotid fistula is classically located lateral to the angle of the mouth, which corresponds to the site of fusion of the maxillary and mandibular

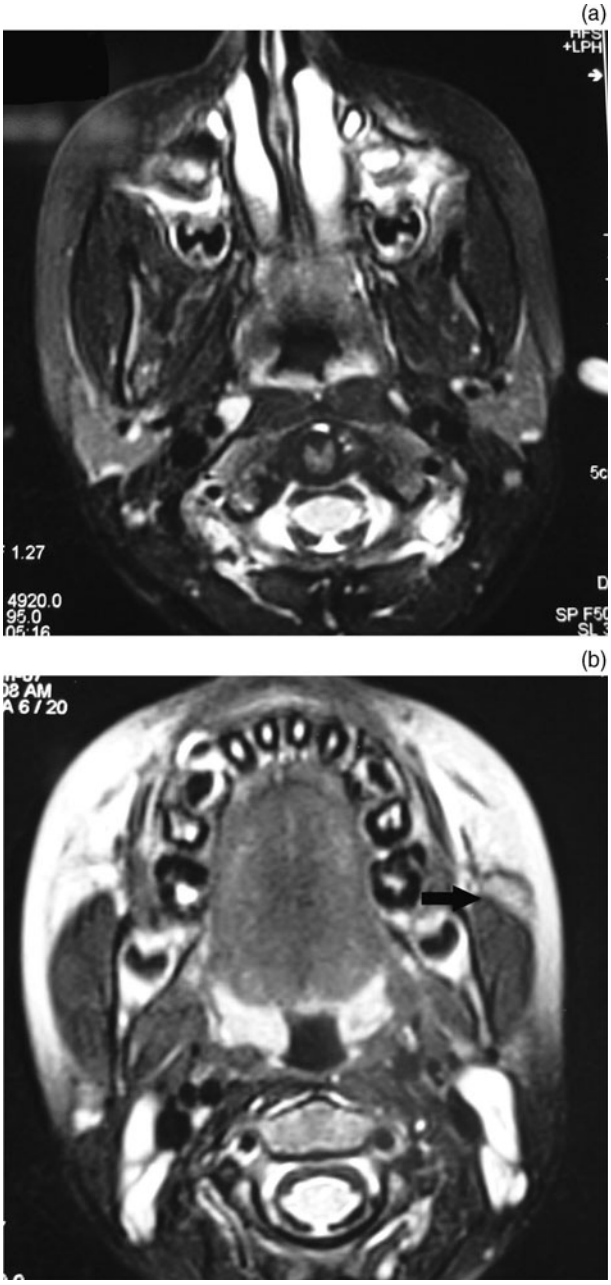

FIG. 2

(a) Turbo spin echo T2-weighted axial magnetic resonance (MR) image, demonstrating normal signal intensity and location of bilateral parotid glands. (b) T2-weighted MR image taken at a more caudal level, showing a small, left-sided accessory parotid gland overlying the left masseter muscle (arrow).

protuberances.[3] An important and consistent association is the presence of periaural appendages.[4,9] In our case, the external opening was located 1.5 cm lateral to the left angle of the mouth, and a left periaural appendix was present.

In a post-mortem study, the prevalence of accessory parotid glands was reported to be 21 per cent. The accessory gland typically lies anterior to the parotid gland along the masseter muscle, with an average diameter of 3 cm. The average distance between the main parotid gland and the accessory parotid gland is

https://doi.org/10.1017/S0022215107001417 Published online by Cambridge University Press

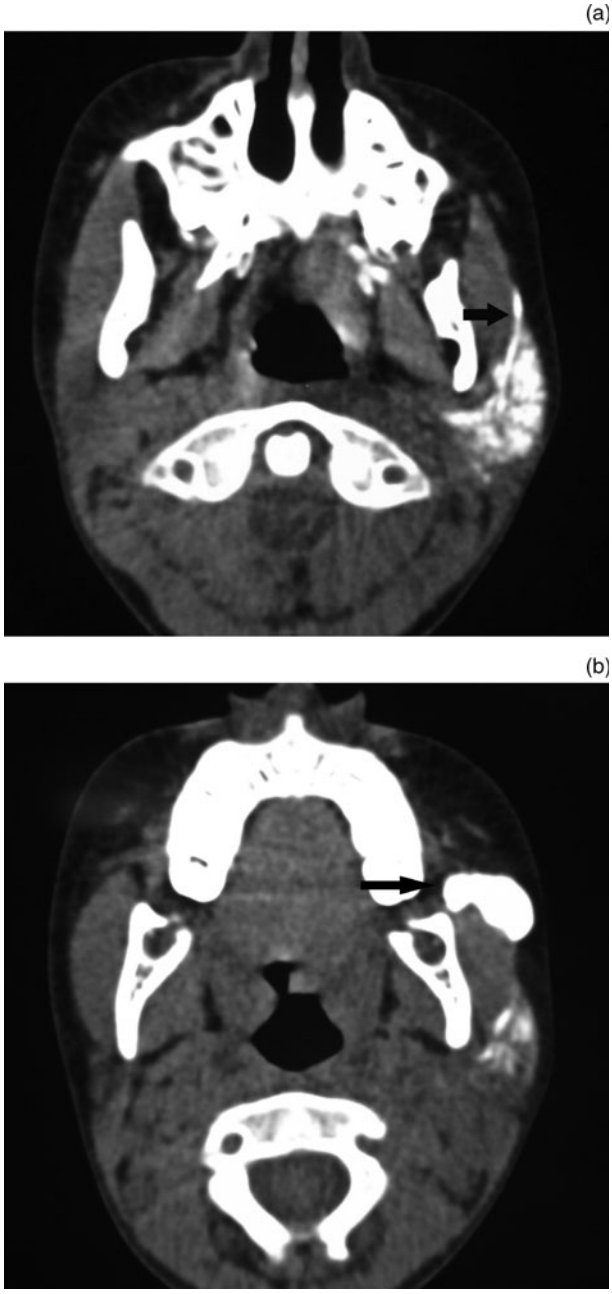

FIG. 3

(a) Computed tomography sialogram demonstrating opacification of the left parotid gland and main duct (arrow). (b) Computed tomography fistulogram demonstrating opacification of the left accessory parotid gland overlying the left masseter muscle (arrow). No communication between main and accessory parotid ducts was demonstrated.

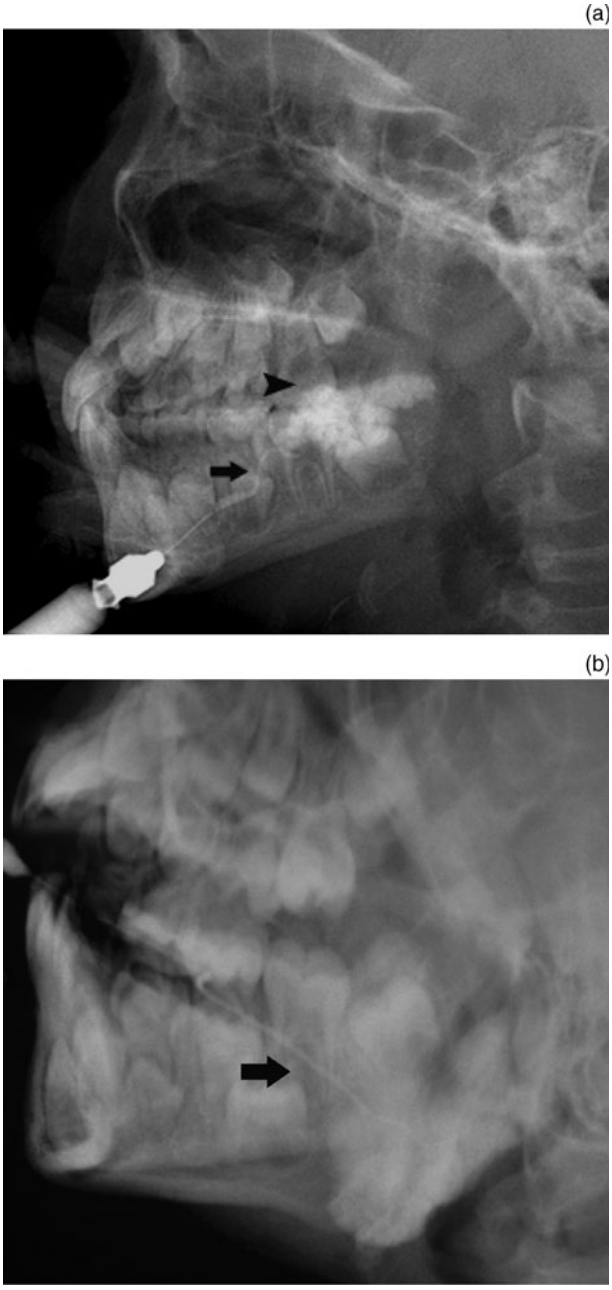

FIG. 4

(a) Fistulogram performed after cannulating the external opening, revealing a narrow duct (arrow) draining glandular tissue overlying the left hemimandible, anterior to the parotid fossa (arrowhead). (b) Digital sialogram of the left parotid gland, demonstrating a normal main duct (arrow). No communication with the accessory duct was demonstrated.

6.0 mm.[10] The accessory gland usually lies cephalad to Stensen's duct, and its major tributary usually drains alongside the main parotid gland into Stensen's duct.[11] In our case, the accessory parotid gland was overlying the masseter muscle.

In our case, MRI and MR sialography were performed to assess the anatomy of the accessory gland and duct. Magnetic resonance sialography is a non-invasive technique of evaluating the ductal system of major salivary glands, and produces sialographic images similar to those of conventional sialography, without the use of contrast media or radiation.[12] In our case, MRI demonstrated an accessory parotid gland overlying the left masseter muscle. The signal intensity of this accessory gland was similar to that of the main parotid gland. Image formation in MR sialography is dependent on duct calibre, adequate duct distention and the T2 relaxation time of saliva. Magnetic resonance sialography demonstrated the normal main parotid duct but failed to delineate the accessory parotid duct, probably because of the duct's narrow calibre.

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Following the advent of MR sialography, CT sialography is now no longer used for evaluation of salivary glands and ducts.[13] However, in our case CT sialography and fistulography were performed, as MR sialography failed to demonstrate the accessory parotid duct. Computed tomography sialography and fistulography confirmed the MRI findings. No communication between the accessory duct and the main duct was present, and the glands were opacified after administration of contrast through their respective openings.

Conventional or digital sialography is considered the 'gold standard' for delineation of the ductal anatomy of the major salivary glands. In our case, digital fistulography revealed a narrow accessory parotid duct, and sialography revealed a normal main parotid duct and intraglandular branches. No communication between the normal and accessory parotid ducts was visualised.

Our literature search revealed only four previously published case reports of congenital accessory parotid fistula.[1–4] In all previously reported cases[1–3] except one,[4] conventional sialography or clinical examination was used to diagnose the congenital parotid fistula. The accessory duct is usually narrower and sometimes runs parallel to the main duct. Moon *et al.*[4] diagnosed a congenital fistula from an accessory parotid gland, using CT sialography and CT fistulography. In their case, the main parotid gland was located anterior to the masseter, in addition to the fistula from the accessory parotid gland. The accessory parotid gland was seen in the buccal space. In our case, however, the accessory parotid gland was present in the masticator space. We attempted to demonstrate the accessory parotid gland and duct using MRI and MR sialography. Magnetic resonance imaging demonstrated the accessory parotid tissue; however, the accessory duct could not be visualised. Conventional and CT fistulography delineated the accessory parotid duct well.

References

1 Yamasaki H, Tashiro H, Watanabe T. Congenital parotid gland fistula. *Int J Oral Maxillofac Surg* 1986;**15**:492–4
2 Zhao K, Wang LM, Qi DY. Congenital extraoral fistula from an auxiliary parotid gland. *Oral Maxillofac Surg* 1992;**50**:752–3
3 Gilbert PM. Congenital external fistulae of the parotid duct. *Br J Plast Surg* 1987;**40**:99–101
4 Moon WK, Han MH, Kim IO, Sung MW, Chang KH, Choo SW *et al*. Congenital fistula from ectopic accessory parotid gland: diagnosis with CT sialography and CT fistulography. *Am J Neuroradiol* 1995;**16**(suppl):997–9
5 Hemenway WG, Bergstrom L. Parotid duct fistula: a review. *South Med J* 1971;**64**:912–18
6 Rothner AD. Aberrant salivary fistulas. *J Pediatr Surg* 1973;**8**:931–3
7 Uekusa Y, Matsumoto K, Ohmae M, Sakuda M. Congenital submental salivary fistula. *Int J Oral Maxillofac Surg* 1999;**28**:400
8 Tasar M, Yetiser S. Congenital salivary fistula in the external auditory canal associated with chronic sialoadenitis and parotid cyst. *J Oral Maxillofac Surg* 2003;**61**:1101–4
9 Grundfast KM, Barber CS, Kubicki SP. Congenital aberrantly located cutaneous Stensen's duct orifice – a newly described anomaly. *Int J Pediatr Otorhinolaryngol* 1987; **14**:197–202
10 Frommer J. The human accessory parotid gland: its incidence, nature, and significance. *Oral Surg Oral Med Oral Pathol* 1977;**43**:671–6
11 Lewkowicz A, Levy Y, Zeltser R, Zagury A, Nahlieli O. Accessory parotid gland masses. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2000;**89**:610–12
12 Kalinowski M, Heverhagen JT, Rehberg E, Klose KJ, Wagner HJ. Comparative study of MR sialography and digital subtraction: sialography for benign salivary gland disorders. *Am J Neuroradiol* 2002;**23**:1485–92
13 Madani G, Beale T. Inflammatory conditions of the salivary glands. *Semin Ultrasound CT MR* 2006;**27**:440–51

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Dr A Seith takes responsibility for the integrity of the content of the paper.
Competing interests: None declared