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## Case Report

# Conventional and MR-sialography of accessory submandibular glands: A case report <sup>☆</sup>

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## ABSTRACT

Our review of the literature shows anatomical variation of the submandibular gland is a rare entity, unlike the variation that can be seen in parotid glands. Specifically, bilateral submandibular abnormality has only been reported on one occasion with limited imaging in our review. We report a 78-year-old female with a history of sialadenitis and sialolithiasis who presents with swelling and pain in the right submandibular gland. Sialography of the right submandibular gland disclosed a second primary duct branching off the main duct to a second submandibular gland. Cannulation of the left submandibular duct was unsuccessful due to scarring of the duct orifice; however, subsequent MR sialography identified marked submandibular duct dilation and the incidental finding of a second anteriorly located left submandibular gland. The anatomic detail provided by conventional digital sialography is contrasted to the useful but less-defined imaging provided by MR sialography. To our knowledge, this is the first reported case of bilateral accessory submandibular glands that has been evaluated using both conventional and MR sialography.

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## Introduction

Anatomic variation of submandibular glands is a rare finding, unlike the more commonly variable parotid gland [1]. Abnormal findings have been characterized by unilateral duplications of submandibular ducts with or without an accessory submandibular gland [2]. These accessory (or duplicate) submandibular glands have most commonly been identified in the course of an evaluation for symptoms of sialadenitis prompting sialography that demonstrates the presence of a

second duct [2–4]. Reports have also described a pleomorphic adenoma as the initial cause of submandibular swelling leading to identification of submandibular accessory glands [5,6]. The majority of cases of duplicate submandibular glands we identified through literature review were evaluated with conventional sialography, with only 2 reported cases evaluated concurrently with MR sialography [3,7,8].

We present a case of a 78-year-old female with bilateral accessory submandibular glands, evaluated with ultrasound, conventional sialography, and MRI sialography. From our review, this patient is the first reported case of bilateral sub-

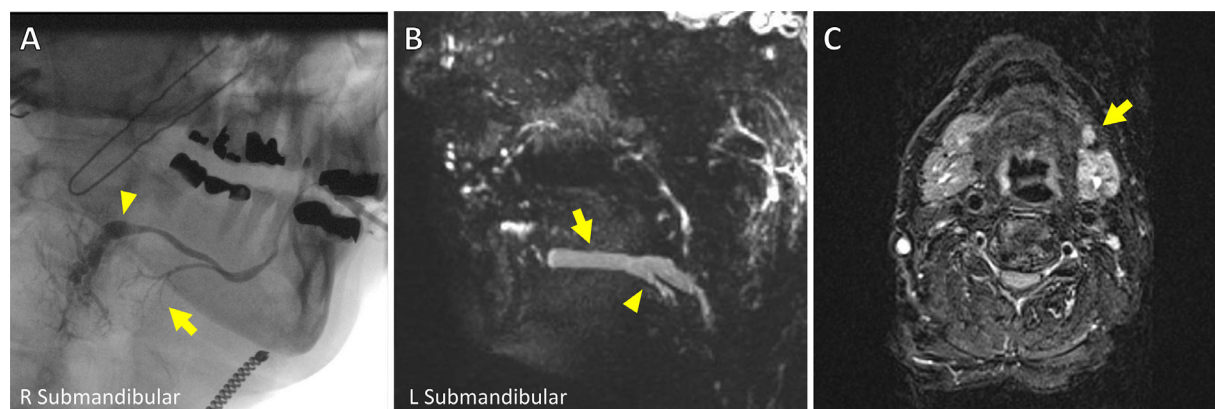
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**Fig. 1 – Fluoroscopic and MR sialogram. (A) Right submandibular digital sialogram with Isovue 370 contrast agent displays a second submandibular duct (yellow arrow) that can be observed with its opening near the papilla. There is marked irregularity and dilatation to the posterior ductal system (yellow arrowhead). (B) High-resolution Space 3D T2 MRI of the oropharynx/oral cavity with administration of Gadavist IV contrast. MR sialogram of left submandibular duct displays markedly dilated submandibular duct of the gland to the papilla, which appears bulky with surrounding enhancement (yellow arrow). The accessory duct can be visualized joining with the main submandibular duct (yellow arrowhead). (C) High-resolution T2 images show a small anterior accessory submandibular gland on the left side (yellow arrow). There is increased enhancement of the left submandibular gland.**

mandibular anatomical variation that is evaluated with all 3 imaging techniques.

### Case report

A 78-year-old female with a 15-year history of sialadenitis and sialolithiasis of the left submandibular gland previously addressed with multiple in-office transoral procedures to remove stones presented with new onset of pain and swelling of the contralateral right submandibular gland over the past year. Examination identified slight tenderness to both submandibular glands on bimanual palpation, with the left gland slightly larger than the right. Bilateral sialograms were attempted, with inability to cannulate the left duct due to complete scarring of the duct orifice.

The right submandibular duct was cannulated successfully with Isovue 370 insufflation, employing standard technique with the modified Seldinger technique using a 22 gauge angiocatheter as previously described [9]. Initial cannulation identified an unremarkable right submandibular gland. Continued infusion with slight withdrawal of the angiocatheter identified a distal take-off to a larger branch off the main duct through which a second abnormal, more proximal submandibular gland was observed (Fig. 1A).

MR sialography was accomplished several months later to further characterize the left submandibular anatomy in preparation for planned open ductoplasty. The MR sialogram identified a markedly dilated left submandibular duct (Fig. 1B). Both right submandibular glands were also imaged with the incidental finding of a fourth submandibular gland as a smaller anteriorly located accessory gland on the left (Fig. 1C). Re-review of the ultrasound done prior to the sialogram confirmed the initial observation of left submandibular sialadenitis with ductal dilation. Further re-interpretation

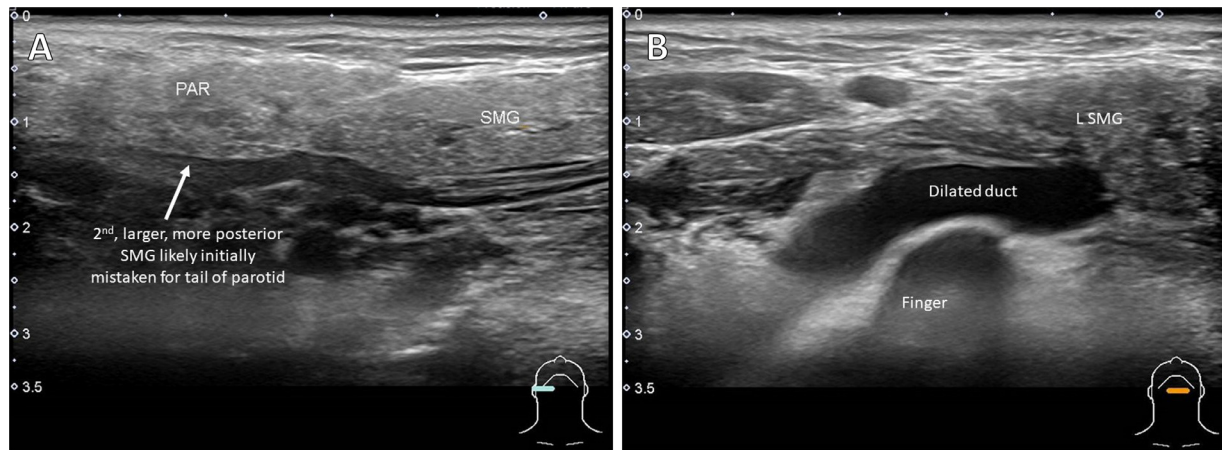
identified that which had been considered the tail of the right parotid gland to more likely represent the larger more proximal of the 2 right submandibular glands with ultrasound characteristics of the sialadenitis as identified on the sialogram (Fig. 2). Repeat ultrasound at 6-month follow-up demonstrated markedly heterogeneous and hypoechoic features of the left submandibular gland, correlating with increased tenderness and swelling of the left gland. Ultrasound-guided imaging again continued to show marked dilation of the left submandibular duct, suggestive of distal scarring. The decision pursue left mandibular sialendoscopy with open ductoplasty was therefore revisited and scheduled.

### Discussion

Our review of the literature identified reports of variation in anatomy of the submandibular gland to be rare. Fewer than 15 cases of accessory submandibular glands have been reported in our review of the literature, whereas accessory lobes to the parotid gland are common and have been in 32.1% of cases examined [1,2].

Two case reports have utilized MR sialography to identify accessory submandibular glands [2,3,8]. However, in both instances, there was unilateral duplication of the submandibular gland with an accessory duct joining the main submandibular duct distal to the papilla. Our review identified a single publication documenting case of bilateral duplication of submandibular glands and ducts identified through conventional sialography, but no case has been observed with MR sialogram [10].

Conventional digital sialography provides detailed imaging of ductal anatomy including tertiary ducts extending to the parenchyma as seen with the intraglandular duct dilation and tortuosity of the posterior submandibular gland present



**Fig. 2 – Submandibular ultrasound. (A) Right submandibular gland which measures at 33.6 x 12.2 x 34.8 mm was hyperechoic and homogenous. What was initially interpreted as the parotid tail showed hypoechoic irregularity, which after further re-interpretation, was determined to be a second submandibular gland. (B) The left submandibular gland was less hyperechoic with some degree of heterogeneity. Measured at 33.3 x 12.5 x 35.0 mm with markedly dilated hilum and duct emanating from the hilum.**

in this patient. MR sialography remains useful as an alternative imaging tool for cases in which ductal cannulation is not possible but, as identified in this comparison, suffers from lack of definition and inability to image tertiary ducts. Ultrasound is a useful adjunct to sialography but is not expected to provide detailed ductal definition.

Comparison between sialograms done for the reported patient identifies the conventional digital sialography with duct cannulation and contrast infusion to provide a clear representation of the ductal anatomy that disclosed the right-sided division into an accessory gland. MR sialography subsequently identified the presence of a small additional left submandibular accessory glands and, despite the lack of resolution compared to the conventional sialogram, was successful in disclosing gross left gland ductal pathology useful for surgical planning. To our knowledge, this is the first reported case of bilateral accessory submandibular glands that has been evaluated using both conventional and MR sialography.

### Patient consent

Written consent was obtained from patient in reported case study, indicating it is okay to use photos, videos, and case history for publication or presentation purposes in any media.

### REFERENCES

- [1] Rosa MA, Lazarz DP, Pekala JR, Skinningsrud B, Lauritzen SS, Solewski B, et al. The accessory parotid gland and its clinical significance. *J Craniofac Surg* 2020;31(3):856–60.
- [2] Torres-Gaya J, Marques-Mateo M, Dualde-Beltran D, Sada-Malumbres A, Garcia-San Segundo MDM, Pucho-Torres M. Accessory submaxillary gland: two new case reports and a literature review. *J Clin Exp Dent* 2020;12(9):e892–5.
- [3] Sanchez Barrueco A, Santillan Coello J, Sobrino Guijarro B, Villacampa Auba JM, Cenfor Espanol C. Sialolithiasis in an accessory submandibular gland identified by magnetic resonance sialography. *Ann Otol Rhinol Laryngol* 2016;125(7):603–6.
- [4] Alexander AD. Accessory submaxillary gland. *N Engl J Med* 1957;256(6):265–6.
- [5] Bryan S, Bodner L, Manor E, Brennan PA. Pleomorphic adenoma occurring outside the submandibular gland: a case report of an accessory submandibular gland. *J Oral Maxillofac Surg* 2013;71(10):1703–5.
- [6] Desai RS, Meshram D, Jangam SS, Singh JS. Pleomorphic adenoma of an accessory submandibular salivary gland: a rare entity. *Br J Oral Maxillofac Surg* 2015;53(8):e33–5.
- [7] Afzelius P, Nielsen MY, Ewertsen C, Bloch KP. Imaging of the major salivary glands. *Clin Physiol Funct Imaging* 2016;36(1):1–10.
- [8] Gadodia A, Seith A, Neyaz Z, Sharma R, Thakkar A. Magnetic resonance identification of an accessory submandibular duct and gland: an unusual variant. *J Laryngol Otol* 2007;121(9):e18.
- [9] Truong K, Hoffman HT, Policeni B, Maley J. Radiocontrast dye extravasation during sialography. *Ann Otol Rhinol Laryngol* 2018;127(3):192–9.
- [10] Codjambopoulo P, Ender-Griepkoven I, Broy H. Bilateral duplication of the submandibular gland and the submandibular duct. *Rofo* 1992;157(2):185–6.