Congenital duplication of submandibular ducts

Soliman Ahmed, MSc, MD, *Hussain Al-Jawad,* FRCP, *Abdurrahman Al-Sayyari,* MSc, *Ali N. Khan,* FRCR, FRCS.

Bilateral duplication of submandibular ducts ending in a ranula was revealed by sialography and confirmed by T2-weighted SSh sequence using magnetic resonance cholangio-pancreatography (MRCP) parameters. A literature search revealed 2 previous reports of submandibular duct duplication, but a ranula associated with submandibular duct duplication has not been previously reported. A review of congenital anomalies of the submandibular ducts is presented and the value of non-invasive MR imaging is discussed.

A 19-year-old woman is presented with one centimeter, translucent, fluctuant, sublingual cystic swelling. The swelling appeared to increase in size following a meal. There was no previous history to note; and a full blood count and blood biochemistry appeared normal. No salivary duct calculus was identified on conventional radiographs. At sialography, bilateral submandibular duct duplication showed the ducts terminating in a ranula. The anatomy was elegantly and non-invasively depicted by T2-weighted SSh rad, the same parameters used for MRCP, TR8000 and TE1100 (**Figure 1**). Congenital anomalies of the submandibular ducts are rare. Pownell et al[1] reported 5 newborns that presented with cystic lesions of the floor of mouth. Four of these patients proved to have congenitally imperforate submandibular salivary gland ducts, and the other newborn proved to have a duplication anomaly of the submandibular gland duct and gland. The diagnosis of congenital anomalies of the submandibular gland and duct can be made on physical examination. Magnetic resonance imaging can be helpful in differentiating congenital imperforate submandibular duct and duplication anomalies of the ductal system. A failure in diagnosis and treatment may result in ranula formation or sialoadenitis requiring more extensive therapy. Amin and Bailey[2] presented 2 infants with unilateral cystic swellings in the floor of the mouth as a result of imperforate submandibular ducts. This is thought to result from a congenital failure of canalization of the terminal end of the duct. Both cases responded to simple incision and decompression of the fluid-filled duct. Early treatment is important to avoid feeding difficulties and to prevent later complications such as ranula or sialadenitis. Other anomalies of the submandibular salivary gland and duct include bilateral Wharton duct, bilateral submandibular duct atresia, bilateral congenital absence of the duct orifice, and congenital dilatation of the submandibular ducts and imperforate salivary ducts with a result of sublingual cystic swelling.

Differential diagnosis of cyst-like swelling of the mouth floor. The differential diagnosis of cyst-like swelling in the floor of the mouth includes sialolithiasis, mucous retention phenomenon (mucocele, ranula), dermoid, epidermal inclusion cyst, thyroglossal duct cyst, branchial cleft cyst, hemangioma, lymphangioma, cystic hygroma, lipoma, and occasionally pleomorphic adenoma. Ranula is a raised mucocele on the oral floor. When the mucocele extends and passes the sublingual space and invades the

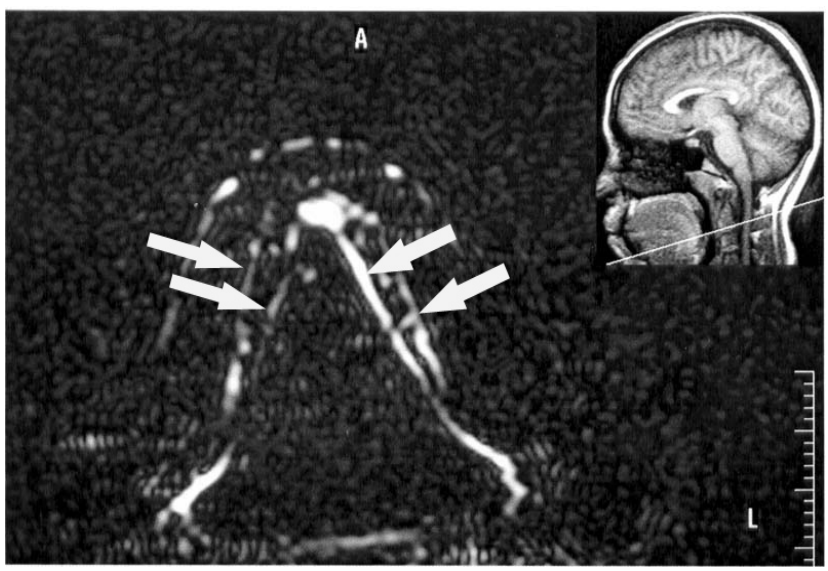

Figure 1 - An MR sialogram, T2-weighted SSh axial cuts through the floor of the mouth showing bilateral submandibular duct duplication (arrow) terminating in a ranula.

submandibular space it may be called "plunging ranula". Its exact etiology is not completely known. A plunging ranula can be readily identified preoperatively with CT as a cystic mass in the suprahyoid anterior neck. Surkin et al[3] encountered an unusual case of a mucocele arising from the submandibular gland. The unique characteristics of this mass permitted definitive diagnosis by CT scanning. Mucoceles originating from the submandibular gland are extremely rare. The diagnosis of these lesions is complicated because of the lack of specific clinical diagnostic criteria and the similarity between submandibular mucoceles and plunging or cervical ranulas. A CT, and specifically the presence of a so-called "tail" sign are pathognomonic for plunging ranula. This sign is absent in mucoceles originating in the submandibular glands.[4] Hydatid cysts can also be encountered in the differential diagnosis, particularly, in endemic areas. Giant cystic dilatation in the floor of the mouth can be due to multiple sialolithiasis of the submandibular gland and, therefore, should be included in the differential diagnosis. An HIV-positive patient, with lymphoepithelial cysts of the parotid glands can have lymphoepithelial cysts in the submandibular glands.

Imaging. The MR imaging has proven to be effective in depicting a wide variety of pathologic changes of the salivary gland. The MR appearance of normal and pathologic states of the submandibular gland has been described by Kaneda et al.[5] The MRI can show the presence, extent, margins, and signal intensity changes of pathologic conditions of the submandibular gland. All normal submandibular glands show higher signal intensity than surrounding muscle but a lower intensity than fat on T1-weighted and T2-weighted images. Post contrast images show moderate enhancement of the gland. Tumors have lower signal intensity than the normal submandibular gland on T1-weighted images, and an intermediate to high signal intensity relative to the normal submandibular gland on T2-weighted images. Benign lesions are well defined, while malignant tumors tend to be poorly defined. In sialadenitis, the submandibular gland shows diffusely different signal intensities from the normal gland on both T1-weighted and T2-weighted images. The MR sialography has become an alternative imaging technique for ductal salivary gland disorders. The MR sialography with a heavily T2-weighted sequence is highly successful in the non-invasive visualization of the ductal system of major salivary glands. It is useful for diagnosing sialolithiasis and sialadenitis. Digital subtraction sialography, an invasive technique, has a substantial procedural failure rate, particularly for the submandibular duct. However, because of its higher spatial resolution, successfully completed digital subtraction sialography achieved superior diagnostic information compared with that of MR sialography. Salerno et al[6] performed conventional fistulography, MRI, and MRI fistulography on a 30-year-old woman that developed a fistula of the Wharton's duct following excision of the submandibular gland, and it proved that the MRI and MRI fistulography is invaluable in detecting the exact extent of the fistula. Morimoto et al[7] performed MR sialography of the parotid gland, the submandibular gland ducts, or both, using 3D fast asymmetric spin-echo sequencing. Their aim was to perform 3D reconstruction images and virtual endoscopic views of the parotid gland ducts using MR sialography data sets of 3D-FASE sequences. Their initial experience showed that virtual MR endoscopy could be performed to observe the endoluminal tracts of parotid and submandibular glands, although the clinical use of the virtual MR endoscopy for salivary gland ducts has yet to be established. Nevertheless, the future applications of the 3D-reconstruction images and virtual endoscopic views using MR sialography data sets of 3D-FASE sequences are very attractive, and further expansion of this field is expected.

Congenital anomalies of the salivary ductal system are rare but their early recognition is important if effective treatment is to be instituted. Cystic lesions of the floor of the mouth are also a rare entity but when found clinically, a wide differential and accurate diagnosis is important to be undertaken for effective treatment. The appropriate role of MRI in achieving the diagnosis is discussed.

Received 31st January 2007. Accepted 1st May 2007.

From the Department of Medical Imaging, King Fahad National Guard Hospital, Riyadh, Kingdom of Saudi Arabia. Address correspondence and print request to: Dr. Soliman Ahmed, Department of Medical Imaging, King Fahad National Guard Hospital, Kingdom of Saudi Arabia. Tel. +966 (1) 2520088 Ext. 11383. Email: soliman002@yahoo.com

References

1. Pownell PH, Brown OE, Pransky SM, Manning SC. Congenital abnormalities of the submandibular duct. ***Int J Pediatr Otorhinolaryngol*** 1992; 24: 161-169
2. Amin MA, Bailey BM. Congenital atresia of the orifice of the submandibular duct: a report of 2 cases and review. ***Br J Oral Maxillofac Surg*** 2001; 39: 480-482. Comment on: ***Br J Oral Maxillofac Surg*** 2002; 40: 455.
3. Surkin M, Remsen K, Lawson W, Som P, Biller HF. A mucocele of the submandibular gland. ***Arch Otolaryngol*** 1985; 111: 623-625.
4. Anastassov GE, Haiavy J, Solodnik P, Lee H, Lumerman H. Submandibular gland mucocele: diagnosis and management. ***Oral Surg Oral Med Oral Pathol Oral Radiol Endod*** 2000; 89: 159-163. Comment on: ***Oral Surg Oral Med Oral Pathol Oral Radiol Endod*** 2000; 90: 404-405.
5. Kaneda T, Minami M, Ozawa K, Akimoto Y, Kawana T, Okada H, et al. MR of the submandibular gland: normal and pathologic states. ***AJNR Am J Neuroradiol*** 1996; 17: 1575-1581.
6. Salerno S, Cannizzaro F, Lo Casto A, Barresi B, Speciale R. The value of magnetic resonance imaging in a fistula of Wharton's duct. ***Dentomaxillofac Radiol*** 2000; 29: 125-127.
7. Morimoto Y, Tanaka T, Yoshioka I, Masumi S, Yamashita M, Ohba T. Virtual endoscopic view of salivary gland ducts using MR sialography data from three dimension fast asymmetric spin-echo (3D-FASE) sequences: a preliminary study. ***Oral Dis*** 2002; 8: 268-274.