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Case Report

Clinical and radiographic follow-up after the Wilkie procedure at 28 years: A case report ☆

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ABSTRACT

Rerouting of the parotid ducts posteriorly to drain into the tonsillar fossae (Wilkie procedure) was initially designed to address drooling in patients with cerebral palsy. This procedure was subsequently modified to include bilateral submandibular gland excision and extended to apply to other etiologies of sialorrhea. Our literature review failed to identify report of long-term follow-up beyond 2 decades following this procedure. We describe a 33-year-old female with spastic cerebral palsy who underwent the Wilkie procedure to treat sialorrhea and, 25 years later, developed right-sided facial swelling and dental infections in association with xerostomia. CT imaging showed symmetric atrophy of the parotid glands with fat replacement interspersed with fibrosis. Ultrasound analysis with shear wave elastography offered the additional interpretation of a greater degree of gland stiffness involving the tail of the recently inflamed right parotid gland compared to the left. This case report identifies long-term complications associated with xerostomia following initial success employing the Wilkie procedure to address sialorrhea. The radiographic evaluation supports the contention that rerouting of Stensen's duct may be associated with chronic obstructive changes to the parotid gland. Ultrasound shear wave elastography supplemented CT imaging by identifying asymmetric stiffness of the parotid glands as was consistent with the more recent right parotid inflammation.

Introduction

The Wilkie procedure as described by Dr Theodore F. Wilkie in 1967 involves rerouting Stensen's duct to redirect saliva to drain into the oropharynx to address sialorrhea in patients with spastic cerebral palsy [1]. Wilkie reported that the laryngopharyngeal phase of swallowing in this group of patients is commonly intact and proposed that by bypassing the impaired movement of the voluntary muscles of the anterior two-thirds of the tongue, the goal of decreased saliva buildup in the oral cavity could be achieved. Ten years later, Wilkie

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and Brody (3 months after Wilkie's death) published an update which expanded the indications for the procedure to include "persistent, severe drooling in any patient in whom non-operative methods have failed [2]." This updated Wilkie procedure was also modified to include excision of both submandibular glands.

The technical features of parotid-duct-rerouting vary by investigator but share the similar goal of positioning the parotid duct orifice into the tonsillar fossa [3,4]. Tonsillectomy is part of the procedure in selected patients with either enlarged tonsils or a history of recurrent tonsillitis.

Wilkie and Brody described results of 123 patients managed by this technique between 1964 and 1973 with at least 1.5-year follow-up for all patients. A standard postoperative course was characterized by discomfort for 3-4 days with facial edema peaking around 48 hours after surgery. Healing was reported as "complete" with drooling controlled by 2 weeks with reported results graded as either "good" or "excellent" in 86% of patients [2]. Forty-three complications were reported including parotid ductal cyst or fistula formation, wound dehiscence, difficulties in oral hygiene, increased dental or gingival problems, transient parotid swelling, and septic parotitis [2].

Other studies with short-term follow-up have subjectively rated similar success [3,5]. The longest follow-up we identified was reported within a series of 23 patients whose caregivers were contacted through telephone survey with follow-up ranging from 6 months to 15 years. Among the 21 with "adequate" follow-up, 18 reported significant improvement in drooling with 2 of these 18 reporting excessive dryness or dental caries [6].

Radiomics is a term describing methods to extract information from medical imaging and includes fat content analysis as determined by tissue density assessment within CT imaging reported in Hounsfield units (HU) [7]. The developing field of ultrasound radiomics has expanded to include shear wave elastography to assess tissue stiffness derived from analysis of the speed of shear waves (m/sec) produced from a secondary "push pulse" delivered by the ultrasound probe. This ultrasound technique is widely accepted as important in liver assessments to diagnose cirrhosis and has markedly diminished the need for biopsy procedures [8]. Emerging support for shear wave analysis in salivary gland analysis has been reported in its application to tumors as well as parenchymal disorders that include chronic parotitis, sialolithiasis, and Sjogren's syndrome [9–11].

This case report provides insight regarding long-term clinical outcomes and radiographic findings comparing CT and ultrasound with shear wave elastography in a patient 28 years following a Wilkie procedure. Informed consent was obtained from the patient. Solicited IRB approval identified exemption as non-human subjects research.

Case report

A 33-year-old female with a history of cerebral palsy with right-sided spasticity was treated in 1995 for drooling sufficient to require 4 shirt changes per day. Chart review identified that she underwent a Wilkie procedure to include rerouting the parotid ducts to the tonsillar fossae after tonsillectomy along with bilateral submandibular gland excisions. She was reported to have tolerated the procedure without complication with follow-up 5 days after the procedure identifying her mouth as feeling "strange" and dry. Record review identified the patient to be doing well 3 months after the procedure. Approximately 16 years later, a dentist advised complete tooth extraction to prevent odontogenic infection given her degree of xerostomia, but she and her family opted against this procedure.

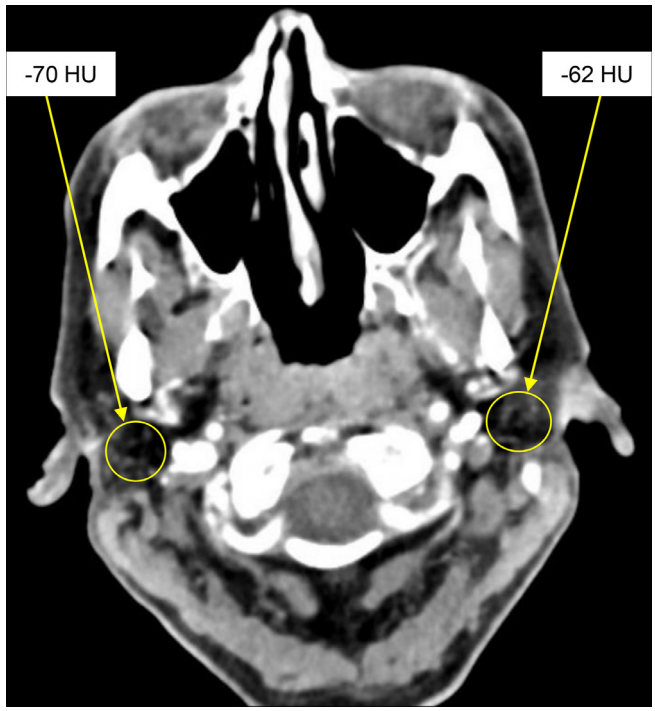

Fig. 1 – CT (March 2023), 28 years after Wilkie procedure. Hounsfield unit measurement in the expanded regions of interest (150 mm^2) is consistent with fat replacement (-70 and -62 HU). HU = Hounsfield units.

Twenty-five years after the Wilkie procedure, the patient began experiencing right-sided facial swelling without pain. Her absence of pain was explained by her sister as due to a loss of sensation with inability to identify pain sensation on that side accompanied by a longstanding right-sided paraplegia. CT imaging with contrast showed an abscess in the gingivolabial sulcus extending through the retromolar trigone in the expected region of the right molar tooth. She subsequently underwent extraction of 2 teeth (#3 and #5) with purulent drainage delivered from the right maxillary canine space to resolve the swelling.

A second episode of right-sided facial swelling occurred 2 years later (August 2022) addressed with another dental extraction and was followed by another facial swelling episode the following month. At that time, her dentist treated her again with antibiotics and referred her to our Otolaryngology service due to concerns about sialadenitis.

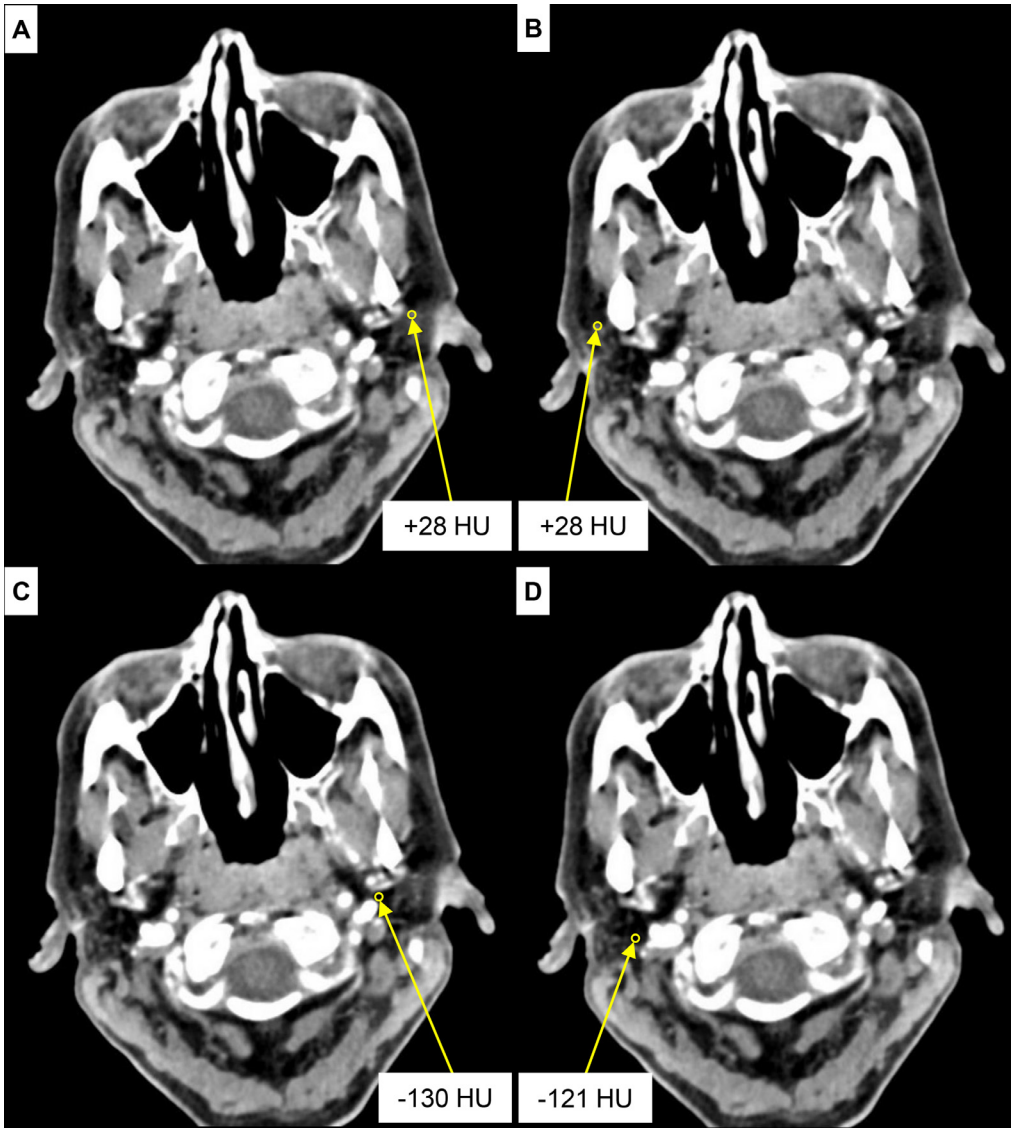

Fig. 2 – CT (March 2023), 28 years after Wilkie procedure. (A, B, C, D) Focused analysis (ROI = 1 mm^2) showing parenchymal heterogeneity. HU = Hounsfield units.

Her evaluation in our clinic (January 2023) revealed a dry oral cavity and scarring in the buccal mucosa consistent with the previous Wilkie procedure. No parotid duct orifice was identifiable in the tonsillar regions. No active infection was identified.

Review of the CT scan previously performed in November of 2020 and then repeated in March of 2023 identified similar symmetric atrophy of the parotid glands with heterogeneous attenuation assessed through measurement of HU with both a diffuse and a more focused analysis consistent with fibrosis and fat replacement. The most recent CT analyzed a larger region of interest (ROI = 150 mm [2]) to identify density measurements of -70 HU for the right and -62 HU for the left parotid glands (Fig. 1). A focused analysis (ROI = 1 mm [2]) identified the least dense regions to measure -130 HU (left) and -121 HU (right) in the parotid glands, correlating with fat density as per Pop et al. [7] (Fig. 2). The most dense areas of the left and right parotid glands measured +28 HU, considered to be consistent with fibrosis.

One month prior to the most recent CT, an ultrasound evaluation was done of the upper neck and salivary glands employing the Canon Aplio 500 system with 5 mm of gel and light application of the linear 5-14 MHz probe overlying the regions studied (Fig. 3). The mylohyoid muscles were followed bilaterally to identify changes to level I consistent with previous submandibular gland resection. B-mode analysis of the parotid glands identified both to be hypo-echoic and heterogeneous with prominent fibrous stranding.

Shear wave elastography (SWE) identified both the left tail and the superficial lobe of both parotid glands posterior-superior to the masseter muscles to be in the range from 1.56 to 1.97 meters/second (m/s) as was interpreted to represent fat replacement in the context of CT interpretation and B-mode imaging (Table 1). In contrast, elevated velocity of the shear

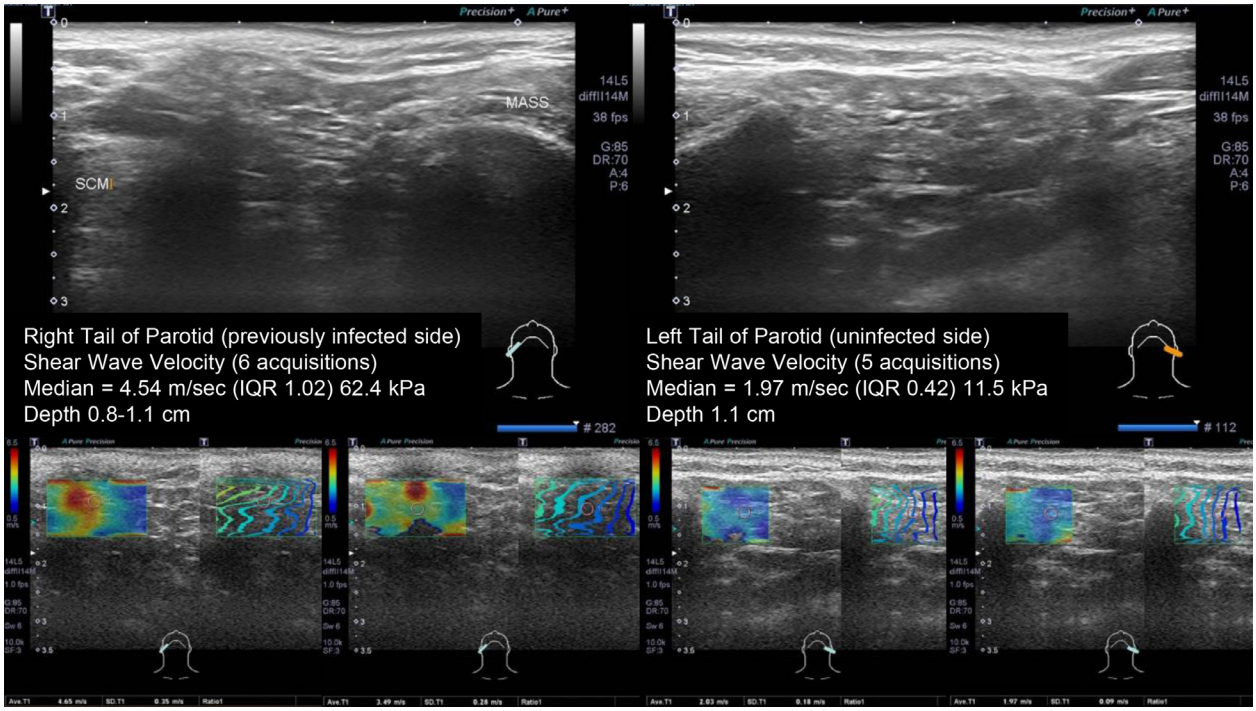

Fig. 3 – Ultrasound of the tails of both parotid glands, between the sternocleidomastoid (SCM) and masseter (MASS) muscles. The lower panel of images identifies representative samples from the multiple acquisitions employing shear wave analysis.

Table 1 – Shear wave analysis employing 5-14 MHz linear probe placed with minimal pressure to skin overlying region studied (ROI = 2) with 5-6 acquisitions each.

	Left parotid		Right parotid	
	Tail	Posterior-superior to masseter muscle	Tail	Posterior-superior to masseter muscle
Velocity (m/s) (median/IQR)	1.97/0.42	2.04/0.14	4.54/1.02	1.56/0.59
kPa (median/IQR)	11.6/5.0	12.4/1.7	62.4/25.7	7.2/5.1
Depth from skin (cm)	1.1 cm	0.9-1.0 cm	0.8-1.1 cm	0.8-0.9 cm

wave (4.54 m/s) was identified in the tail of the previously infected right parotid gland, interpreted to represent a greater degree of fibrosis with a less dominant presence of fat.

Final assessment suggested that the previous episodes of the right-sided facial swelling and infection were likely associated with xerostomia and an oral cavity/dental nidus with potential propagation to the right parotid. Treatment focused on oral hygiene care with management of xerostomia and consultation with infectious disease experts to consider intermittent versus chronic antimicrobial treatment.

Discussion

Wilkie and Brody affirmed in 1977 that "the role of saliva in dental and gingival health has never been clearly defined" and that their described procedure did not affect dental health [2]. Subsequent studies have refuted that contention in confirming the important role of saliva for speech, swallowing, lubrication, and protection against dehydration, infection, and dental caries [12]. Our report highlights the long-term impact of surgically directed xerostomia with associated complications.

Other approaches to address "significant" sialorrhea in the 1970s included tympanic neurectomy, chorda tympani sectioning, radiation therapy, parotid duct ligation, and physiotherapy, with all identified by Wilkie and Brody as inferior treatments for severe sialorrhea when compared to their surgical approach [2]. Other approaches that were not available at that time have been subsequently developed to provide a less aggressive approach to reducing saliva production. Medical therapy with drying agents such as glycopyrrolate, transdermal scopolamine, atropine drops, and amitriptyline, is now an established approach but suffers from the need for ongoing administration of drugs with recognized side effects. Botulinum neurotoxin injection to the salivary glands offers an option with a lower risk of side effects but is disadvantaged by

the need for repeated injections that in children may require general anesthesia.

The optimal management for sialorrhea remains difficult to determine and warrants an individualized approach with advocates for contemporary treatment to consider the additional surgical options of submandibular duct rerouting or ligation as well as neurectomy of the submandibular ganglion [13,14]. Although rerouting of the parotid duct has fallen from favor, parotid duct ligation is still advocated by some investigators as a less involved surgery with similar ductal obstructive outcomes accompanied by gland atrophy – often supplemented by treatment to the submandibular gland [15,16].

In his 10-year review, Wilkie denied others' claims that his procedure is "nothing more than a complicated way of doing a parotid duct ligation, and that the salivary control results from consequent parotid atrophy [2]." He defended the procedure by claiming the ability to demonstrate continued parotid function with radioactive technetium scans and ability to express saliva from the ducts. Whereas the reported short-term benefit of the Wilkie procedure may identify success in maintaining parotid salivary flow in selected cases, our study raises the specter of long-term complication associated with severe xerostomia.

Ida and Honda reported analysis of the radiation attenuation within the CT scans of normal parotid glands of 182 patients (age range 1-81 years). A mean parotid density of -10.0 HU was identified with a standard deviation of -24.1 HU and a range of -60 to +40 HU [17]. Comparison of these measurements to those of the parotid glands in our case report (-70 and -62 HU) indicates that this low-density measurement following the long-term duct rerouting is likely associated with fat replacement due to ductal obstruction.

The radiographic and clinical outcomes of our reported case are consistent with findings from Pogrel et al. of reduced parotid function in their study of 8 patients following the Wilkie procedure. Each patient underwent technetium scanning at 2 months and then again at 1 year postoperatively, with results demonstrating a mean parotid activity level of 35% of expected and 50% of expected, respectively [18]. In contrast to Wilkie and Brody's assertion, our results support the contention that the effort to redirect salivary flow by rerouting Stensen's duct may result in stenosis leading to complete obstruction of the duct associated with atrophy and fat infiltration [2].

Previous studies evaluating ultrasound with shear wave elastography have reported the mean velocity within the parotid gland to be 1.854 m/s (corresponding 10.38 kPa) [19,20]. Muntean et al. [21] through literature review, reported normal parotid gland assessment to result in a range of elasticity values from 5.39 to 26 kPa.

The elevated shear wave velocity of 4.54 m/s (elasticity 62.4 kPa) we identified in the tail of the right parotid supports the concept of a greater stiffness in this portion of the gland that is potentially related to inflammation associated with oral infections responding to antibiotics. Alternative explanations include the potential for artifacts from the study as can be induced in the process of gland compression during placement of the ultrasound probe [20]. The impact of this potential artifact was mitigated by performing 5 separate acquisitions to record the median result resulting from these independent measurements - each done with attention to applying minimal pressure from the probe placement.

Atrophy of the gland with fibro-fatty tissue replacement in association with increasing age has been reported as a cause for a decrease in the density of the parotid gland as reported by HU [12,17]. Although this normal aging process may be contributory in this patient's case, it does not likely completely account for the changes that occurred in the context of the additional CT and ultrasound findings supportive of atrophy with additional fibrosis.

To our knowledge, this long-term follow-up is the first of its type to identify the negative impacts of xerostomia 28 years following the Wilkie procedure. CT density assessment and ultrasound shear wave elastography are compared in a novel manner that emphasize the heterogeneity of the gland with fibrosis and fat replacement – with additional evidence for increased right parotid tail fibrosis in the context of recurrent infections isolated to this region. This presentation identifies potential long-term complications from surgical treatment of sialorrhea and offers support for considering less aggressive approaches for management that include medical management or botulinum toxin injection to the salivary glands.

Patient consent

Written consent was obtained from patient and patient's sister who was the durable power of attorney (DPOA) in the reported case study. The DPOA indicated that it is okay to use photos, videos, and case history for publication or presentation purposes in any media.

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