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Case Report A SCITECHNOL JOURNAL

Cerebrospinal Fluid Leak from Lateral Sphenoid Sinus after Nasopharyngeal Swab Test for COVID-19: A Case Report and Review of Literature

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Abstract

Background: During the Coronavirus Disease 2019 (COVID-19) pandemic, nasopharyngeal swab testing has been performed approximately 41 million times in Canada thus far. Extremely rare cases of nasopharyngeal swabs breaching the skull base and resulting in a Cerebrospinal Fluid (CSF) leak have been reported in the literature. Although rare, a traumatic CSF leak is a potentially serious complication that needs to be kept in mind to implement preventative measures and to ensure adherence of correct swab technique.

Methods: Case report and literature review.

Results: Our case report describes a previously unreported case of a CSF leak from a COVID-19 swab originating from the sphenoid sinus. The patient initially presented with CSF rhinorrhea and a new-onset headache following a COVID-19 swab. Upon recognition and diagnosis, the patient was successfully treated with surgical repair. This is the fifth reported case of iatrogenic CSF rhinorrhea following a nasopharyngeal swab. The second reported case where the patient had no suspected pre-existing condition with no previous physical or radiographic evidence of an encephalocele prior to the nasopharyngeal swab. This is the first case to report a traumatic breach of the skull base located within the sphenoid sinus following performance of a nasopharyngeal swab.

Conclusion: Early identification of a CSF leak following a COVID swab is crucial for patient care and management. Moreover, proper swabbing technique is essential for prevention and stricter technique guidelines should be implemented.

Keywords: COVID-19; Coronavirus; CSF leak; Encephalocele; Nasopharyngeal swab

Introduction

In March 2020, the Coronavirus Disease 2019 (COVID-19) emerged as a global pandemic. To date, there have been over 220 million confirmed positive cases of COVID-19 [1]. Nasopharyngeal swabs for Reverse Transcription Polymerase Chain Reaction (RT-PCR) testing have thus far remained the gold standard for confirming the presence of Severe Acute Respiratory Syndrome Coronavirus 2 (SARS-CoV-2) and are considered generally safe [2]. With hundreds of millions of swabs performed globally, extremely rare cases of nasopharyngeal swabs breaching the skull base and resulting in a Cerebrospinal Fluid (CSF) leak have been reported [3-6]. This report describes a previously unreported case of a CSF leak from a COVID-19 swab originating from the sphenoid sinus and a review of the literature regarding this rare iatrogenic complication.

Case Study

A case report describing a previously unreported case of a CSF leak from a COVID-19 swab originating from the sphenoid sinus is presented. Clinico-demographics, presenting symptoms, diagnosis and management are described.

A literature review regarding this rare complication was subsequently performed and discussed in detail.

The rarity and significance of this case was discussed with the patient and informed consent was obtained to include the patient's clinical information, photo and diagnostic imaging in this publication. Institutional ethics review from our institution, Hamilton Integrated Research Ethics Board (HIREB) is not required for such case reports.

Results

A 45-year-old female was required to undergo multiple COVID-19 nasopharyngeal swab tests for work purposes. During her first nasopharyngeal swab in December 2020, the patient reported the left-sided swab to be extraordinarily painful compared to her subsequent experiences. In the following two days, the patient started experiencing increasing left-sided clear metallic-tasting nasal discharge. By the following week, this became quite profuse and persistent. The patient eventually presented to her family doctor and was referred to an otolaryngologist. Four months after the swab, a CT scan was performed, and a beta-2-transferrin test was performed which came back positive, confirming the presence of CSF. She was then referred to our tertiary centre where an MRI of the skull base was performed.

The patient reported a persistent daily new-onset headache following the COVID-19 swab. The patient denied any previous history of nasal discharge, rhinosinusitis, meningitis, nasal surgery, head trauma, headache and any other visual or neurological symptoms. The patient's past medical history is significant for

depression, gastroesophageal reflux disease, hypertension, Crohn's disease necessitating two bowel resections requiring keloid scar repair and repair of a fistula and abscess which resulted in a mild chronic pain syndrome. Her medications include venlafaxine, mirtazapine, pantoprazole, vitamin B12 injections, morphine, oxycodone, irbesartan and vedolizumab. The patient has a remote history of smoking, very rare alcohol consumption and no recreational drug use.

A physical exam was performed which demonstrated a positional provocation of rhinorrhea with downward position and a positive teapot sign [7] with a clear liquid dripping from her nostril every 30 to 45 seconds as she bent her head forward (Figure 1). Flexible nasal endoscopy demonstrated some clear thin discharge throughout the left nasal cavity without any apparent anatomical abnormalities. A full neurological exam was performed which was unremarkable. An ophthalmological exam was also unremarkable with no evidence of papilledema or signs of increased intracranial hypertension. The patient's body mass index was 31 and vital signs were within normal limits.

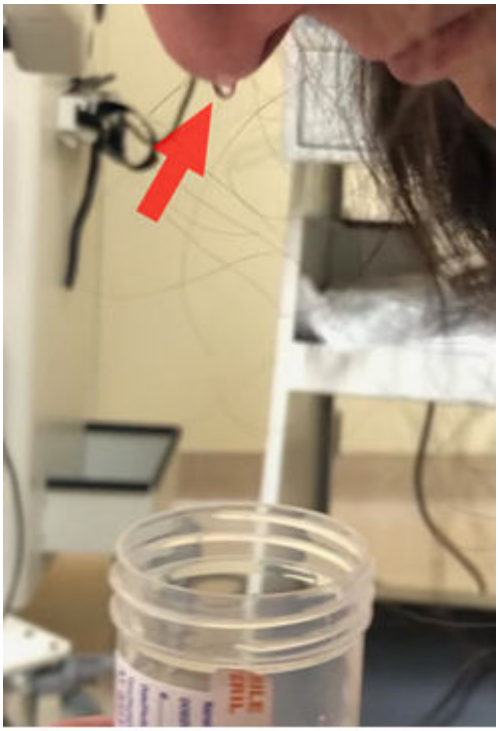

Figure 1: Image demonstrating positional provocation of rhinorrhea with downward face position and a short period of observation (positive teapot sign). A drop of CSF may be seen at the tip of the patient's nose (red arrow).

A Computed Tomography scan (CT) scan was performed which demonstrated a bony defect within the anterolateral portion of the sphenoid sinus approximately 8.2cm from the left nostril (Figures 2 and 3). Magnetic Resonance Imaging (MRI) was subsequently performed 1week later which demonstrated dehiscence of the lateral sphenoid wall with a small encephalocele containing mesial temporal lobe (Figure 4). Although this was not seen on the CT scan, it is hard to know if this was pre-existing (Figure 5). An empty sella was also noted on the MRI examination.

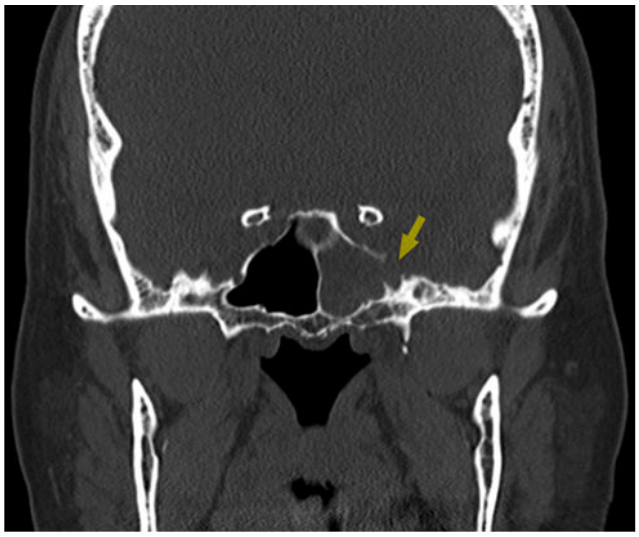

Figure 2: Coronal CT demonstrates the bony defect (yellow arrow) within the lateral sphenoid wall.

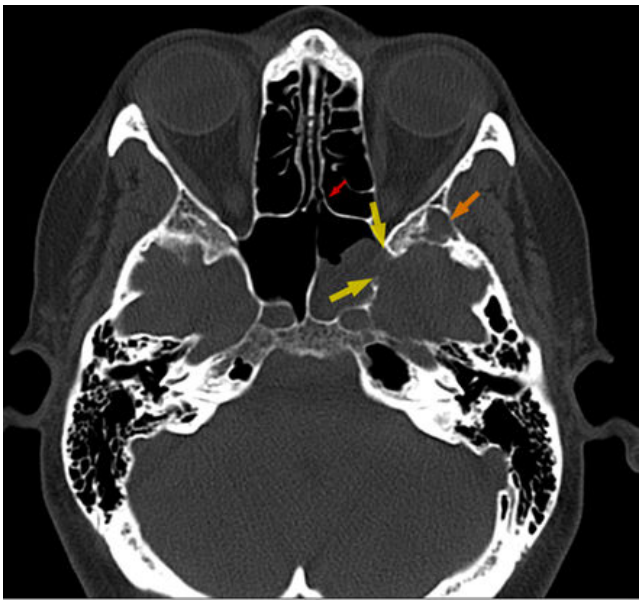

Figure 3: Axial CT demonstrates a bony defect (yellow arrows) in the lateral wall of the left sphenoid sinus. There is associated soft tissue opacification of the sinus. Red arrow indicates the sphenoid os. Orange arrow denotes an incidental arachnoid pit in the greater wing of sphenoid.

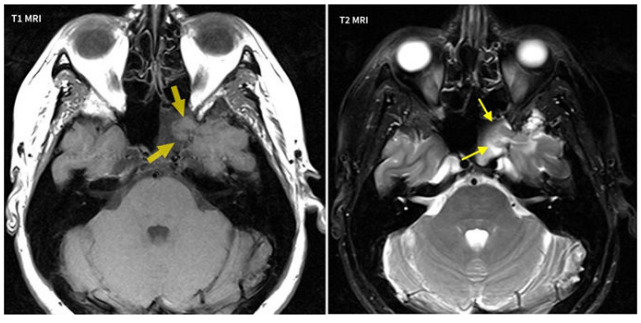

Figure 4: Axial T1 and T2 MRI sequence demonstrates brain parenchyma herniating through the bony defect of lateral wall of sphenoid sinus (yellow arrows).

Figure 5: Coronal T1 MRI demonstrating brain parenchyma (yellow arrow) herniating through the bony defect of lateral wall of the sphenoid sinus.

The patient was brought to the operating room for repair and underwent an endoscopic endonasal skull base approach. Prior to any surgical intervention, an identical COVID swab was used to replicate the suspected trajectory (Figure 6), which was easily achieved.

Intraoperative measurement of the COVID swab from the anterior naris to the encephalocele was 9.5 cm. A unilateral left-sided surgical approach was used to identify the sphenoid ostium with subsequent drilling and opening of the floor, anterior and lateral wall of the sphenoid exposing the encephalocele (Figure 7).

The encephalocele was then successfully reduced using bipolar cautery (Figure 8). An ipsilateral sphenopalatine artery based nasoseptal flap was harvested and the defect was repaired with a free fat graft sealed with fibrin glue.

The nasoseptal flap was then rotated and placed overlying the defect with placement of fibrin glue, additional free fat graft and subsequently HemoPore (Stryker, Kalamazoo, MI, USA) to help secure the repair (Figure 9).

At a two-week follow-up visit, the patient was doing well with no evidence of CSF leak recurrence.

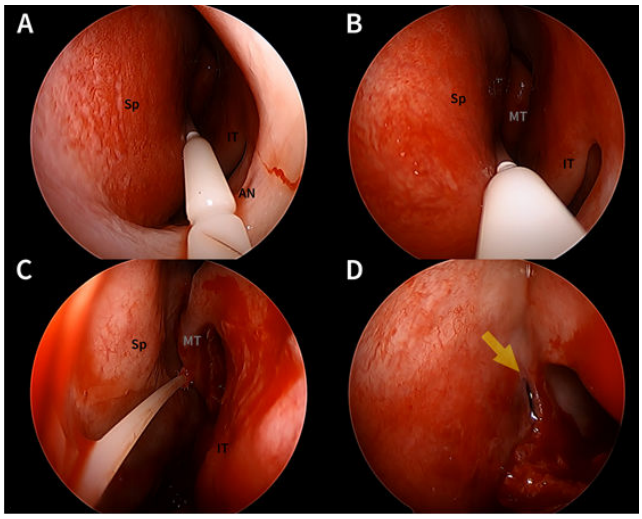

Figure 6: Intraoperative image demonstrating the suspected trajectory of the nasopharyngeal swab causing a discontinuity in the skull base. Figure D is after partial superior turbinectomy at natural sphenoid ostium (yellow arrow), demonstrating CSF. Sp: Septum; MT: Middle turbinate; IT: Inferior turbinate.

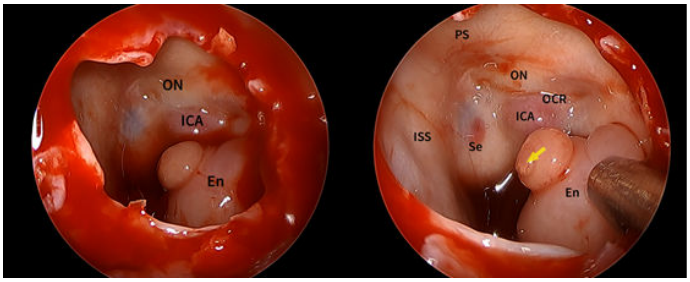

Figure 7: Intraoperative image through enlarged sphenoid ostium demonstrating the left sphenoid sinus and encephalocele. A drop of clear CSF may be visualized as well (yellow arrow). PS: Planum Sphenoidale; ON: Optic Nerve; OCR: Optico Carotid Recess; ICA: Internal Carotid Artery; Se: Sella turcica; ISS: Inter Sphenoid Septum; En: Encephalocele.

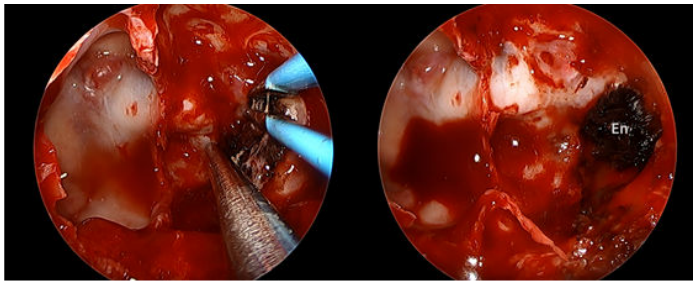

Figure 8: Intraoperative image demonstrating bilateral sphenoidotomy, intersphenoid septum removed and the reduced encephalocele. En: Encephalocele.

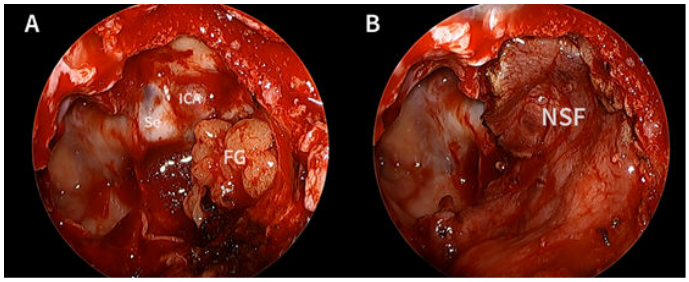

Figure 9: Intraoperative image demonstrating the surgical repair with free fat grafts, fibrin glue and the ipsilateral nasoseptal flap. FG: Fat Graft; NSF: Nasoseptal Flap; ICA: Internal Carotid Artery; Se: Sella.

Discussion

Following a review of the literature, this is the fifth reported case of iatrogenic CSF rhinorrhea following a nasopharyngeal swab. This is the second reported case where the patient had no suspected pre-existing condition with no previous physical or radiographic evidence of an encephalocele prior to the nasopharyngeal swab.

This is however the first case to report a traumatic breach of the skull base located within the sphenoid sinus following performance of a nasopharyngeal swab. Other reports describe a breach of the ethmoid roof or cribriform plate region.

Figures 6 and 10 are intraoperative and radiographic images indicating the suspected trajectory of the nasopharyngeal swab. It is likely that the flexible nasopharyngeal swab managed to enter the sphenoid os as this was easily replicated at prior to beginning surgical exposure. Given the patients' structural anatomy (lateral trajectory of anterior inter-sphenoid septum), the swab was subsequently deflected laterally through the lateral wall of the sphenoid sinus (Figure 10).

As previously described spontaneous lateral sphenoid encephaloceles are often associated with pneumatization of the lateral sphenoid recess (not significant in our patient) as well as presence of arachnoid pits. Although an incidental arachnoid pit was noted in the greater wing of the sphenoid it is not at the site of the bony defect (Figure 8). As such this case does not fulfill the criteria for type 1 or type 2 spontaneous sphenoid cephaloceles as described by Settlecase et al [8]. Given the provided history and temporal evolution the defect may have occurred secondary to a traumatic covid swab.

However, with no prior imaging, the possibility of a pre-existent lateral sphenoid skull base bony defect or encephalocele exists and the nasopharyngeal swab may have traumatically breached the sinus mucosa and dura and lead to the leak. The swab would have still been required to penetrate the sphenoid os, which is an unlikely event.

It is also possible that our patient had undiagnosed Idiopathic Intracranial Hypertension (IIH) given her age, gender, body habitus, and radiographic evidence of empty sella syndrome. An ophthalmological exam was, however, unremarkable with no characteristic findings of IIH. This exam, however, was after the CSF leak had ensued and the pressure may have consequently been mitigated by the leak.

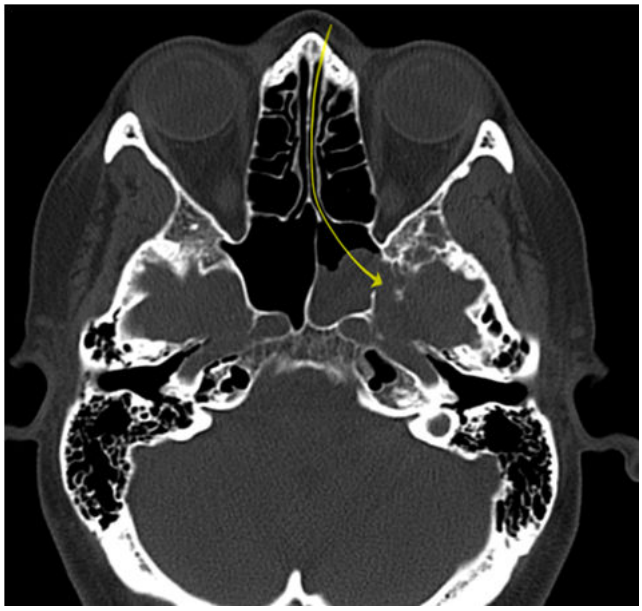

Figure 10: Radiographic image demonstrating the suspected trajectory of the nasopharyngeal swab causing a breach of the skull base within the sphenoid sinus (yellow arrow). Note the lateral direction of the sphenoid septum anteriorly likely guided the swab laterally.

This is the first reported case of a COVID swab causing a nasal CSF leak in Canada. As of September 9th, 2021, this would result in a national incidence of 1 in 41,151,117 [9]. Around 270 million COVID swabs have been performed in the United Kingdom [10], around 588 million in the United States [10] and globally, over 2 billion COVID swabs have been performed in the most impacted countries worldwide as September 9th, 2021 [10]. As previously mentioned, this is only the fifth reported case of a COVID nasopharyngeal swab associated with an iatrogenic CSF leak and the first such described in the sphenoid sinus. Considering the global total number of nasopharyngeal swabs performed, this case report demonstrates an extremely rare yet severe complication.

The importance of proper nasopharyngeal swab technique needs to be discussed and disseminated. Regarding this reported case, the patient had numerous subsequent COVID swabs without any complications. However, during the swab associated with the CSF leak, she reported immediate severe pain. There is significant variability and heterogeneity in performance of nasopharyngeal swabs [11]. The current physical length of our nasopharyngeal swabs is 15 cm (Figure 11) and the Center for Disease Control and prevention (CDC) have published guidelines regarding proper technique [12]. There is however no mention about maximum insertion length of the nasopharyngeal swab. An emphasis regarding proper head position is also important to highlight as patients may have a tendency to extend their neck back and tilt their head up. This potentially allows for the swab to travel in a more cephalic trajectory towards the skull base. Interestingly, this head position is a common image displayed across multiple sites including the CDC [13]. The distance to the posterior nasopharynx is 9-10 cm [14] however there is still no large population data on this measurement, so it is hard to recommend a maximum depth. The key is to stop if any significant resistance is met which is likely what occurred with our reported case. A nasal swab task trainer

has recently been developed to expedite training new swab administrators and is currently pending publication [15].

Figure 11: Image of a standard nasopharyngeal swab used at our center.

Currently, the gold standard for diagnosing COVID-19 is with the use of nasopharyngeal swabs. Although rare, we suspect that the overall incidence of traumatic CSF leaks associated with nasopharyngeal swabs is underreported given the millions of swabs being done daily across the world. Other techniques for diagnosing COVID-19 also being used and further studied. While this is a rare complication of nasopharyngeal swabbing, alternative approaches may need to be explored especially in patients with previous sinus/skull base surgery or known skull base abnormalities. Skull base injury should be considered when teaching nasopharyngeal swab technique and deciding which technique to use in general.

Conclusion

The use of nasopharyngeal swabs has significantly increased during the COVID-19 pandemic. Although rare, a traumatic CSF leak is a potentially serious complication that needs to be kept in mind in order to implement preventative measures and ensure correct swab technique are adhered to. Other patients in similar case reports had underlying pre-existing conditions that placed them at a higher risk for a CSF leak. However, our reported patient had no clear or known underlying risk factors. This case highlights the need for proper technique when performing a nasopharyngeal swab. The presence of significant pain and resistance during the swab should prompt a redirection and/or attempt at contralateral access using a more favourable trajectory.

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Author contributions

Concept and design: DD Sommer, T McHugh.

Acquisition, analysis, or interpretation of data: All authors.

Drafting of the manuscript: DD Sommer, T McHugh, K Reddy, LJ Sowerby.

Critical revision of the manuscript for important intellectual content: All authors.

Administrative, technical, or material support: DD Sommer, T McHugh, K Reddy.

Supervision: DD Sommer, T McHugh.

Disclosures

DD Sommer: Reported research support, advisory board fees from Glaxo Smith Kline and Sanofi and speaker fees from Medtronic. LJ Sowerby: Research and development of a nasopharyngeal swab trainer.

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