



Sinonasal Lobular Capillary Hemangioma After Continuous Self-tests for COVID-19: A Case Report and Review of the Literature

COVID-19 için Sürekli Yapılan Kendi Kendine Testlerden Sonra Sinonazal Lobüler Kapiller Hemanjiyom: Bir Olgu Sunumu ve Literatür Taraması

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ABSTRACT

Sinonasal lobular capillary hemangiomas (LCH) are rare benign vascular lesions commonly arising from the nasal septum. Nasopharyngeal, nasal mid-turbinate, and anterior nasal swabbing is the preferred method of screening for coronavirus disease-2019 (COVID-19). Herein, we present a case of a sinonasal LCH in a child after continuous self-tests for COVID-19, with an anterior nasal swab. The child presented with a well-defined red mass in the anterior part of the nasal septum, which was removed endoscopically. Histopathology revealed a LCH. This is the first report of a complication other than epistaxis with the use of an anterior nasal swab. Our literature review identified 32 studies reporting complications of COVID-19 screening. Cerebrospinal fluid leaks and foreign body retention are the most common ones. A proper specimen collection technique and a quick patient history with an emphasis on risk factors are the best practices to prevent complications from COVID-19 screening.

Keywords: Lobular capillary hemangioma, swabs, COVID-19, screening, complication

ÖZ

Sinonazal lobüler kapiller hemanjiyomlar (LCH), genellikle nazal septumdan kaynaklanan, nadir görülen iyi huylu vasküler lezyonlardır. Nazofaringeal, nazal orta konka ve anterior nazal sürüntü, koronairüs hastalığı-2019 (COVID-19) için tercih edilen tarama yöntemidir. Burada, anterior nazal sürüntü ile COVID-19 için sürekli kendi kendini test eden bir çocukta sinonazal LCH olgusunu sunuyoruz. Çocuk, nazal septumun ön kısmında endoskopik olarak çıkarılan, iyi tanımlanmış kırmızı bir kitle ile başvurdu. Histopatoloji sonucu LCH olarak raporlandı. Bu çalışma, anterior nazal swabın kullanımıyla epistaksi dışındaki bir komplikasyona ilişkin ilk rapordur. Literatür taraması sonucunda, COVID-19 taramasının komplikasyonlarını bildiren 32 çalışma belirlendi. Beyin omurilik sıvısı sızıntıları ve yabancı cisim retansiyonu bunlardan en yaygın olanlarıdır. Uygun bir örnek toplama tekniği ve risk faktörlerini vurgulayarak hızlı bir hasta öyküsü, COVID-19 taramasından kaynaklanan komplikasyonları önlemek için en iyi uygulamalardır.

Anahtar kelimeler: Lobüler kapiller hemanjiyom, sürüntüler, COVID-19, tarama, komplikasyon

INTRODUCTION

Lobular capillary hemangiomas (LCH) are benign vascular lesions, commonly affecting the skin and oral cavity mucosa¹. Hemangiomas are common lesions of the head and neck area, although they rarely derive

from the nasal cavity². Sinonasal LCH most commonly present with nasal obstruction, epistaxis, nasal discharge, and postnasal drip³. Their etiology is not completely understood, but trauma, hormonal stimulation, and microscopic arteriovenous malformations are thought to play a role in the pathogenesis^{1,2}. Since March

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Received: 18 October 2022

Accepted: 08 December 2022

Online First: 16 December 2022

Cite as: Fountarlis AL, Lachanas V, Zacharouli K, Hajioannou J, Kalogritsas N, Skoulakis C. Sinonasal Lobular Capillary Hemangioma After Continuous Self-tests for COVID-19: A Case Report and Review of the Literature. Medeni Med J 2022;37:339-345

2020, coronavirus disease-2019 (COVID-19) has been recognized as a pandemic, and every effort has been put to identify patients early in the disease course and stop its spread⁴. Nasopharyngeal, nasal mid-turbinate, and anterior nasal specimen collection by swabbing is the preferred method of initial diagnostic testing for COVID-19⁵. Herein, we present a case of a sinonasal LCH in a child after continuous self-tests for COVID-19 with a nasal swab.

CASE REPORT

A 9-year-old boy presented to our department with a 4-month history of recurrent epistaxis and nasal obstruction. The episodes of epistaxis were self-limited and increased in frequency. Anterior rhinoscopy revealed a well-defined red mass in the anterior part of the nasal septum of the left nostril (Figure 1). Nasal endoscopy did not reveal any other lesions. The child's caregiver reported that before the onset of epistaxis, the child was attending a series of school lessons on how to properly perform a self-test for COVID-19 using a nasal swab in the left nostril.

The child had a normal birth, no other health problems were reported, so he was scheduled for surgical excision under general anesthesia. The mass was removed endoscopically, and the patient was discharged the next day. The histopathological report was consistent with a diagnosis of LCH (Figure 2a, b). No recurrence was noted on a 1-year follow-up.

Written informed consent was obtained from the patient's caregiver for the publication of this case report and the accompanying images.

DISCUSSION

Sinonasal LCH are rare lesions, and a few case series and case reports have been published in the literature. They arise most frequently from the nasal septum in Kiesselbach's area between the 3rd and 5th decade of life⁶. Differential diagnosis includes hematomas or abscesses, nasal polyps, benign tumors of the nasal cavity such as juvenile angiofibromas, hemangiopericytomas,

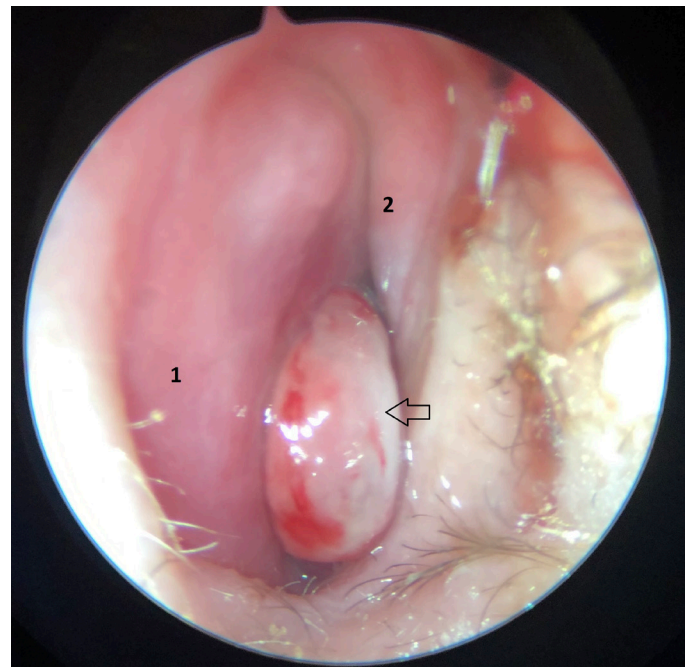


Figure 1. Endoscopic image of the lobular capillary hemangioma. (1) nasal septum, (2) inferior nasal turbinate, arrow: lobular capillary hemangioma.

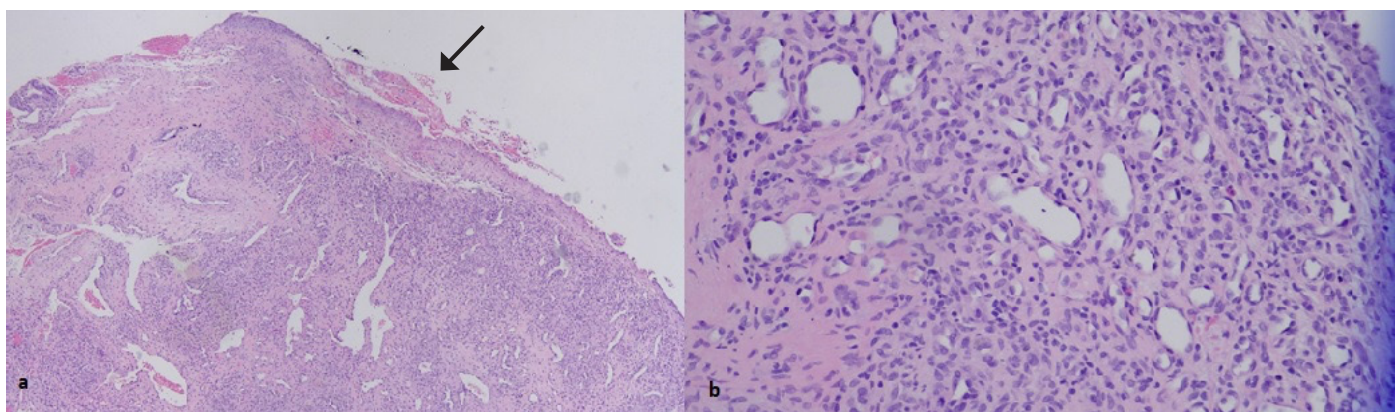


Figure 2. Histopathology. Hematoxylin-eosin staining. **a.** X4 magnification, **b.** X20 magnification. Histopathology revealed a nodule with surface ulceration (arrow) (**a**) and lobular vascular proliferation consisting of vessels of capillary caliber (**b**). There is no atypia or mitotic activity.

paragangliomas, and papillomas, as well as malignant tumors of the nose. In this case, the mass was red, well-circumscribed, and located in Kiesselbach's area, and a hematoma was ruled out, making hemangioma the most likely diagnosis.

Trauma, as mentioned, is one of the postulated etiopathogenic factors of LCH^{2,3,6,7}. Specifically, in this case, we consider that continuous trauma in the nasal septum by a nasal swab led to the formation of a hemangioma. This is the first case of LCH formation secondary to nasal swab collection for COVID-19 screening. However, whether the hemangioma was present, the nasal swab triggered the episodes of epistaxis, or the nasal swab itself led to its formation cannot be proven.

Several other complications of COVID-19 screening have been reported in the literature, with the most common being epistaxis⁸. Some rarer complications are presented in Table 1, and foreign body retention in the nasal cavity and gastrointestinal tract (GIT) and cerebrospinal fluid (CSF) leaks are the most frequent. To our knowledge, 13 reports of CSF leaks have been published, and four of them have been complicated with meningitis or meningoencephalitis⁹⁻¹². Of note, in 7 of these 13 cases, several predisposing factors have been recognized, with encephalocele as the most common. Most of these cases have been managed surgically, and just in one case, minimal neurologic sequelae were reported⁹. The most frequent areas of the leak were the sphenoid sinus^{4,11,13,14}, cribriform plate^{5,10,15,16}, and fovea ethmoidalis^{17,18}. This means that a correct specimen collection technique, with the direction of the swab just above and parallel to the floor of the nasal cavity, would have prevented the leak. Foreign body retention in the nasal cavity and GIT was reported in 15 and 8 patients, respectively. In all of them, the foreign bodies were successfully removed, apart from one patient, with a history of multiple surgeries in the GIT, in whom swallowing of the swab led to intestinal perforation¹⁹. Specimen collection swabs have a breakpoint mechanism to prevent contamination when transferring the shaft into the container, which makes them susceptible to accidental rupture²⁰. In the presence of predisposing factors, such as nasal septal deviation^{20,21}, or in an uncooperative patient^{20,22,23}, the risk of accidental breakage of the swab increases. Dislocation of the temporomandibular joint²⁴, nasal septal⁸ and pharyngeal abscesses²⁵, orbital cellulitis^{26,27}, ethmoidal silent sinus syndrome²⁸ and neurally mediated syncope²⁹ have also been reported as complications of screening for severe

acute respiratory syndrome coronavirus-2. Interestingly, the vast majority of the reported adverse events were caused by nasopharyngeal swabs, suggesting that this specimen collection technique bears the highest risk. The LCH formation, presented in this paper, is the first reported complication following the use of an anterior nasal swab.

These cases underline that despite nasal and nasopharyngeal swab sampling being safe and well-tolerated procedures⁸, serious adverse events can occur. During the current COVID-19 pandemic, thousands of rapid and self-tests are performed daily as a public health strategy to detect suspicious cases early. These tests are performed by physicians, nurses, pharmacists, and individuals, who, in many cases, are not familiar with the anatomy of the nasal cavity, increasing the risk of adverse events. To minimize complications, proper knowledge of the sampling technique and the anatomy of the nose is important. The three main swab collection methods are the anterior nasal, mid-turbinate, and nasopharyngeal methods⁵. Nasopharyngeal swabs are considered more reliable than the other methods but are more invasive¹⁰. Nasopharyngeal and mid-turbinate swabs should be inserted in a plane parallel to the floor of the nasal cavity, pointing to the level of the external auditory canal. The inclination of the swab should not exceed 30° from the floor of the nasal cavity. Moreover, maneuvers should be gentle, and scratching violently the nasal septum should be avoided. The distance between the entrance of the nose and the posterior wall of the nasopharynx is 8-10 cm in adults and 6-7 cm in children³⁰. A quick medical history, with a focus on previous surgeries in the nose, presence of nasal septal deviation, encephaloceles, meningoceles, idiopathic intracranial hypertension, or coagulopathies could also prevent adverse events. If there is a high risk of complication, alternative specimen collection techniques, such as oropharyngeal or anterior nasal swabbing, could be chosen.

Sinonasal LCH are rare benign vascular lesions appearing as red well-circumscribed masses in the nasal cavity. This is the first presentation of an LCH after continuous self-tests for COVID-19 with an anterior nasal swab. Healthcare professionals should be aware that screening tests with nasal or nasopharyngeal swabs are not free from complications. When testing, the swab should be advanced in a plane parallel to the floor of the nasal cavity and its angle from that plane should not exceed 30°.

Table 1. Literature review: study characteristics.							
Author (year)	Type of study	Age/sex	Type of testing	Complications	Possible predisposing factors	Management	Outcomes
Föh et al. ²⁴ (2021)	Case series	1. 53/M, 2. 55/M, 3. 29/F	1. 2. nasal, 3. oropharyngeal	1. 2. Foreign body in the nose, 3. Dislocation of the TMJ	None	External jaw repositioning	1. 2. Successfully removed 3. resolution of symptoms
Fabbris et al. ⁸ (2021)	Case series	1. 72/F, 2. 79/M, 3. 53/M, 4. 58/F	NA	1. Nasal septum abscess, 2. 3. Foreign body in the nose, 4. Foreign body in the GIT	NA	1. Incision and drainage, 2. 3. endoscopic removal, 4. conservative	NA
Koskinen et al. ²¹ (2021)	Case series	NA	Nasopharyngeal	Three cases of foreign body in the nasal cavity, 1 in the GIT	Nasal septal deviation in 1 case	endoscopic removal in 3, conservative in 1	Successfully removed
Azar et al. ³¹ (2020)	Case report	99/M	Nasopharyngeal	Foreign body in the nasal cavity	None	Endoscopic removal	Successfully removed
Wyman et al. ³² (2021)	Case report	52/F	Nasopharyngeal	Foreign body in the nasal cavity	None	Endoscopic removal	Successfully removed
Mughal et al. ²² (2020)	Case report	70s/M	Nasopharyngeal	Foreign body in the nasal cavity	Uncooperative (dementia)	Endoscopic removal	Successfully removed
Clark et al. ³³ (2021)	Case report	30s/NA	Nasopharyngeal	Foreign body in the nasal cavity	None	Endoscopic removal under GA	Successfully removed
Suresh ³⁴ (2021)	Case report	12/F	Nasal	Foreign body in the nasal cavity	None	Endoscopic removal under GA	Successfully removed
Rigante et al. ³⁵ (2021)	Case report	32/M	Nasopharyngeal	Foreign body in the nasal cavity	Important nasal septal deviation	septoplasty and removal	Successfully removed
Gaffuri et al. ²⁰ (2021)	Case report	37/M	Nasopharyngeal	Foreign body in the nasal cavity	Uncooperative (Down's syndrome)	Endoscopic removal under GA	Successfully removed
Hussain et al. ³⁶ (2020)	Case report	51/F	Through tracheostomy tube	Foreign body in the trachea	None	Endoscopic removal	Successfully removed
Medas et al. ³⁷ (2020)	Case report	64/M	Nasopharyngeal	Foreign body in the stomach	None	Endoscopic removal	Successfully removed
de Luca and Maltoni ³⁸ (2021)	Case report	47/M	Nasopharyngeal	Foreign body in the stomach	None	Endoscopic removal under GA	Successfully removed
Molinár et al. ³⁹ (2021)	Case report	45/M	Oropharyngeal	Foreign body in the stomach	None	Endoscopic removal	Successfully removed
Farina and Nelson ⁴⁰ (2021)	Case report	35/F	Nasopharyngeal	Foreign body in the duodenum	None	Endoscopic removal	Successfully removed

Table 1. continued							
Author (year)	Type of study	Age/sex	Type of testing	Complications	Possible predisposing factors	Management	Outcomes
Tümer and Ardiçlı ²³ (2021)	Case report	22m/F	Nasopharyngeal	Foreign body in the duodenum	Uncooperative (age)	Endoscopic removal under GA	Successfully removed
Verstuijls et al. ⁹ (2022)	Case report	56/M	Oropharyngeal	Intestinal perforation	Multiple previous surgeries (obesity & hernias)	Surgery	Wound dehiscence
Lapeyre et al. ²⁵ (2021)	Case report	73/NA	Nasopharyngeal	Pharyngeal abscess	History of craniopharyngioma surgery	Conservative	Death
Sullivan et al. ¹⁸ (2020)	Case report	40s/F	Nasal	CSF leak	Preexisting encephalocele	Surgery	Na
Holmes and Allen ⁹ (2021)	Case report	54/F	Nasopharyngeal	CSF leak and meningitis	Congenital meningocele	Surgery	Minimal neurologic sequelae
Ovenden et al. ⁴¹ (2021)	Case report	34/F	Nasopharyngeal	CSF leak	None	Conservative	Resolution of symptoms
Mistry et al. ¹⁰ (2021)	Case report	67/F	Nasopharyngeal	CSF leak and meningitis	None	Surgery	Resolution of symptoms
Asiri ¹¹ (2021)	Case report	36/F	Nasopharyngeal	CSF leak and meningoencephalitis	IIH	Conservative	Resolution of symptoms
Alberola-Amores et al. ² (2021)	Case report	41/F	Nasopharyngeal	CSF leak and meningitis	None	Conservative	Improvement of symptoms
Yilmaz et al. ¹⁷ (2021)	Case report	47/M	Nasopharyngeal	CSF leak	Prior head trauma	Surgery	Resolution of symptoms
Rajah and Lee ⁴ (2021)	Case report	59/M	Nasopharyngeal	CSF leak and encephalocele	Possible preexisting encephalocele	Surgery	Resolution of symptoms
Agamawi et al. ¹³ (2021)	Case report	40/M	Nasopharyngeal	CSF leak	None	Surgery	Resolution of symptoms
Samadian et al. ⁵ (2021)	Case report	47/F	Nasopharyngeal	CSF leak	None	Surgery	Resolution of symptoms
Knížek et al. ¹⁶ (2021)	Case report	40s/M	Nasopharyngeal	CSF leak	None	Surgery	Resolution of symptoms
Paquin et al. ¹⁵ (2021)	Case report	38/F	Nasopharyngeal	CSF leak and encephalocele	Possible preexisting encephalocele	Surgery	Resolution of symptoms
Dünder et al. ¹⁴ (2022)	Case report	61/M	Nasopharyngeal	CSF leak and encephalocele	Possible preexisting encephalocele	Surgery	Resolution of symptoms

Table 1. continued

Author (year)	Type of study	Age/sex	Type of testing	Complications	Possible predisposing factors	Management	Outcomes
Fazekas et al. ²⁷ (2021)	Case report	35/F	Nasopharyngeal	Preseptal cellulitis and infraorbital abscess	None	Conservative	Resolution of symptoms
Cantarella et al. ²⁶ (2022)	Case report	79/M	Nasopharyngeal	Orbital cellulitis	Previous ESS	Conservative	Resolution of symptoms
Ribeiro et al. ²⁸ (2022)	Case report	40s/M	Nasopharyngeal	Ethmoid silent sinus syndrome	None	Surgery	Resolution of symptoms
Bloom and Bloom ²⁹ (2022)	Case report	73/F	Nasopharyngeal	Neurally mediated syncope	None	Conservative	Resolution of symptoms
Fountarlis et al. (2022)	Case report	9/M	Anterior nasal	Lobular capillary hemangioma	None	Surgery	Resolution of symptoms

NA: Not available, M: Male, F: Female, IIH: Idiopathic intracranial hypertension, ESS: Endoscopic sinus surgery, GIT: Gastrointestinal tract, GA: General anesthesia, CSF: Cerebrospinal fluid, m: Months

Ethics

Informed Consent: Written informed consent was obtained from the patient’s caregiver for the publication of this case report and the accompanying images.

Peer-review: Externally and internally peer-reviewed.

Author Contributions

Surgical and Medical Practices: A.L.F., K.Z., C.S., Concept: A.L.F., V.L., C.S., Design: A.L.F., V.L., N.K., C.S., Data Collection and/or Processing: A.L.F., J.H., Analysis and/or Interpretation: A.L.F., V.L., K.Z., J.H., Literature Search: A.L.F., V.L., N.K., Writing: A.L.F.

Conflict of Interest: The authors have no conflict of interest to declare.

Financial Disclosure: The authors declared that this study has received no financial support.

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