

CAUCASIAN MEDICAL JOURNAL

www.caucasianmedj.com

Year: July 2023 Volume: 1 Issue: 2

- **Lipoma of the Quadrigeminal Cistern as a Rare Cause of Vertigo: Case Report with Literature Review**
Ziya Karimov, Sunay Mustafayeva, Aynur Aliyeva
- **Association of Chlamydia pneumoniae and Borrelia burgdorferi Infections with Multiple Sclerosis**
Ulvi Zahir Nasirov
- **Leptospirosis Case**
Ulvi Zahir Nasirov, Gulam Rustamzada
- **Breast Metastasis from Primary Lung Adenocarcinoma: A Rare Occurrence with Diagnostic Challenges and Treatment Implications**
Rizvan Rustamov, Masoud Mireskandari, Iver Petersen
- **A Case of Esophageal Leiomyoma Resected After Selective Circular Myotomy in Submucosal Tunneling Endoscopic Resection**
Abdullah Murat Buyruk
- **Carbon Dioxide Cholangiography in Diagnosis of Gallbladder Perforation: Deciphering Black, and White**
Ismail Caymaz, Anar Mammadov, Mahmut Bilal Doğan, Ayşenur Buz Yaşar





FOOD INTOLERANCE TEST



SUNLAB
LABORATORİYASI

📍 Centr: Mirali Gashgai 139

📍 Ahmadli branch: Khatai district, Sarajevo 113 A-113 C

📷 sunlablaboratoriya

📌 sunlablaboratoriya

🌐 sunlab.az

***7707**





Academic Jamil Aziz Aliyev

**Academic of Azerbaijan and Georgian National Academies of Sciences,
Academic of the Russian Academy of Sciences, Laureate of “Sharaf” and
“Shohrat” Orders of Azerbaijan**

The National Center of Oncology (NCO) of the Ministry of Health of the Republic of Azerbaijan is the main structure responsible for the coordination of scientific, methodological and preventive activities providing specialized cancer care for the population in our country. The major objective of the Center is to achieve the best survival rate for patients; this is carried out by early diagnosis and such diversified treatment methods of cancer as modern surgery, radiotherapy and chemotherapy.

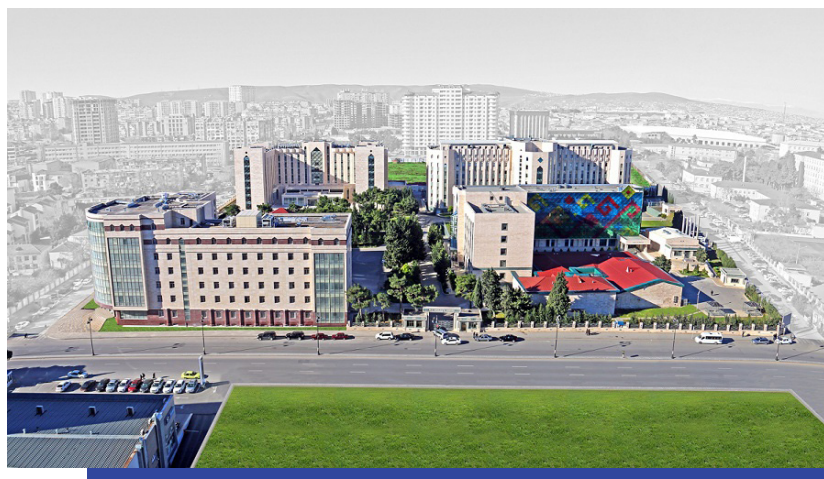
Having functioned as the Republican Oncological Scientific Center with a long historical development path, it received its contemporary title in 1995. Jamil Aziz Aliyev, an Academic of the Azerbaijan National Academy of Sciences, has been the General Director of the Center since March, 1990.

Both the NCO itself and the oncology service in the whole country have been tremendously developing last 20 years. In 2009, the Surgery Building equipped with the most modern medical facilities was opened at the NCO with the participation of the President of the Republic of Azerbaijan Ilham Aliyev. In 2012, the Pediatric Oncology Clinic was launched at the NCO with the participation of the First Vice-President of the Republic of Azerbaijan Mehriban Aliyeva. The Clinic has all departments necessary for the treatment of childhood cancers. Additionally, the Treatment Building and Molecular Oncology Laboratory also started operating at NCO in 2014.

In 2014, the National Center of Oncology and ‘Varian Medical Systems’, a US Company designing and manufacturing the most advanced technologies for radiation treatment, signed an agreement. According to it, the most advanced universal system “TrueBeam™ STx” was installed at the Center, and it was accepted as the reference center of ‘Varian Medical Systems’. At the present time, the NCO has seven linear accelerators. Furthermore, in 2016, the Nuclear Medical Center was opened at the NCO with the participation of officials of the International Atomic Energy Agency, and for the first time in Azerbaijan, PET/CT examination was launched for diseases diagnosis. The Center is included in the Research for Life – the EARL accreditation program established by the European Nuclear Medicine Association.

Performing as the country’s leading medical center, the National Center of Oncology has a clinical base of 1500 beds, twenty-one scientific and fifty-one clinical departments. Currently, the medical personnel of the NCO consist of 1452 people – among them one Academician, eleven Doctors of Science, six Professors, two Honored Scientists and forty-seven Doctors of Philosophy.

The National Center of Oncology effectively cooperates with the world’s leading cancer institutions and centers in the global fight against cancer. Many examples of this include cooperative relations with the University of Texas MD Anderson Cancer Center, the International Prevention Research Institute, Hacettepe University and more. These relations comprise mutual scientific and research programs, the exchange of scientists and medical specialists, the conduction of joint meetings, workshops and training programs aiming to improve the knowledge and experience of the NCO’s personnel. This all leads to better care for patients.




National Center of Oncology

YOU CAN TRUST



 Ahmad Rajabli 208, Baku, Azerbaijan. AZ1052

 (+994 12) 564 84 42

 (+994 77) 403 71 04; (+994 77) 403 71 05





CAUCASIAN MEDICAL JOURNAL



2023
Volume: 1
Issue: 2

EDITORIAL BOARD

Honorary Editor

Teymur Musayev

Ministry of Health of the Republic of Azerbaijan
t.musayev@health.gov.az

Editor-in-Chief

Fatih Aslan

Koç University Faculty of Medicine Hospital,
Department of Gastroenterology, Istanbul,
Turkiye

fatihaslandr@gmail.com

ORCID ID: 0000-0002-1002-7202

Nargiz Afandiyeva

National Oncology Center, Department of
Gastroenterology, Baku, Azerbaijan

dr.nargizafandi@gmail.com

ORCID ID: 0000-0002-8205-3805

Editorial Board

Gülüstan Babayeva

Azerbaijan State Advanced Training Institute for
Doctors named after A. Aliyev, Department of
Internal Medicine, Baku, Azerbaijan

ghbabayeva@gmail.com

ORCID ID: 0000-0002-5805-3741

Jamal Musayev

Baku Pathology Center, Department of
Pathology, Baku, Azerbaijan

patolog.jamalmusaev@gmail.com

ORCID ID: 0000-0002-9202-6990

Kanan Yusif-Zade

Leyla Medical Center, Department of Surgery,
Baku, Azerbaijan

yusifzadekr@yahoo.com

ORCID ID: 0000-0002-7382-4495

Ömer Faruk Özkan

University of Health Sciences Turkey, Istanbul
Ümraniye Training and Research Hospital, Clinic
of General Surgery, Istanbul, Turkey

ozkanfomer@gmail.com

ORCID ID: 0000-0002-6644-2413

Taryel Omerov

Azerbaijan Medical University, Department of
Surgery, Baku, Azerbaijan

Taryelomerov@gmail.com

ORCID ID: 0000-0002-7216-7980

Gunay Aliyeva

National Centre of Oncology, Department of
Nuclear Medicine, Baku, Azerbaijan

galiyeva@rocketmail.com

ORCID ID: 0000-0002-1751-3684

Serhat Bor

Ege University Faculty of Medicine, Department
of Gastroenterology, Izmir, Turkiye

serhat.bor@ege.edu.tr

ORCID ID: 0000-0001-5766-9598

Dmitry S. Bordin

Healthcare Institution of Moscow, A.S. Loginov
Moscow Clinical Scientific Center, Department
of Pancreatic, Biliary and Upper Digestive Tract
Disorders, Moscow, Russia

dmitrybordin@gmail.com

ORCID ID: 0000-0003-2815-3992

Andrii E. Dorofieiev

Shupyk National Healthcare University of
Ukraine, Department of Therapy and Geriatrics;
Department of Internal Medicine and Geriatric,
Kyiv, Ukraine

dorofeyevand@gmail.com

ORCID ID: 0000 0002 2631 8733

Gulay Mammadzada

Private Practice - Psychiatrist, Baku, Azerbaijan

gm20@ic.ac.uk

ORCID ID: 0000-0002-3736-9245

English Language Editor

Emin Mammadov

Mediland Hospital, Department of Internal
Medicine and Gastroenterology, Baku,
Azerbaijan

dr.emin.m8@gmail.com

ORCID ID: 0000-0003-4629-350X

Sevda Aghayeva

Azerbaijan State Medical University, Baku
Medical Plaza Hospital, Department of
Gastroenterology, Baku, Azerbaijan

seva_agayeva@yahoo.com

ORCID ID: 0000-0001-8959-3647

Redactor

Galenos Publishing House

Please refer to the journal's webpage (caucasianmedj.com) for "Editorial Policy", "Instructions to Authors" and "Ethical Policy".

Caucasian Medical Journal and its editors follow the recommendations of "International Committee of Medical Journal Editors (ICMJE)", "World Association of Medical Editors (WAME)", "Council of Science Editors (CSE)", "Committee on Publication Ethics (COPE)", "European Association of Science Editors (EASE)", and "National Information Standards Organization (NISO)."

Responsible Manager: Fatih Aslan



Publisher Contact

Address: Molla Gürani Mah. Kaçamak Sk. No: 21/1
34093 Istanbul, Turkey

Phone: +90 (530) 177 30 97 / +90 (539) 307 32 03

E-mail: info@galenos.com.tr/iyayin@galenos.com.tr

Web: www.galenos.com.tr

Publisher Certificate Number: 14521

Online Publishing Date: July 2023

E-ISSN: 2980-1818

International periodical journal published three times in a year.



CAUCASIAN MEDICAL JOURNAL




2023
Volume: 1
Issue: 2

CONTENTS

CASE REPORTS

- 18** **Lipoma of the Quadrigeminal Cistern as a Rare Cause of Vertigo: Case Report with Literature Review**
Ziya Karimov, Sunay Mustafayeva, Aynur Aliyeva; İzmir, Turkey, Baku, Azerbaijan, Ohio, USA
- 22** **Association of Chlamydia pneumoniae and Borrelia burgdorferi Infections with Multiple Sclerosis**
Ulvi Zahir Nasirov; Baku, Azerbaijan
- 24** **Leptospirosis Case**
Ulvi Zahir Nasirov, Gulam Rustamzada; Baku, Azerbaijan
- 26** **Breast Metastasis from Primary Lung Adenocarcinoma: A Rare Occurrence with Diagnostic Challenges and Treatment Implications**
Rizvan Rustamov, Masoud Mireskandari, Iver Petersen; Gera, Germany
- 30** **A Case of Esophageal Leiomyoma Resected After Selective Circular Myotomy in Submucosal Tunneling Endoscopic Resection**
Abdullah Murat Buyruk; İzmir, Turkey
- 33** **Carbon Dioxide Cholangiography in Diagnosis of Gallbladder Perforation: Deciphering Black, and White**
İsmail Caymaz, Anar Mammadov, Mahmut Bilal Doğan, Ayşenur Buz Yaşar; İstanbul, Muğla, Samsun, Turkey

Lipoma of the Quadrigeminal Cistern as a Rare Cause of Vertigo: Case Report with Literature Review

 Ziya Karimov¹,  Sunay Mustafayeva²,  Aynur Aliyeva³

¹Ege University Faculty of Medicine, Medicine Program, Izmir, Turkey

²Health Department of International Committee of Red Cross, Baku, Azerbaijan

³Cincinnati Children's Medical Hospital, Clinic of Pediatric Otorhinolaryngology-Head and Neck Surgery, Ohio, USA

ABSTRACT

Intracranial lipomas are very rare mesenchymal tumors, constituting 0.1% of all cases. These tumors may be located in the quadrigeminal cistern. Most quadrigeminal lipomas do not cause any clinical symptoms and are found incidentally. If the lesion is large enough, it can cause headaches, seizures, and diplopia. We present a 56-year-old male patient who presented with the complaint of vertigo for five years and was diagnosed with quadrigeminal plaque lipoma, accompanied by magnetic resonance imaging findings and literature review. According to our knowledge, this is the first case in the literature in which a quadrigeminal cistern lipoma caused vertigo.

Keywords: Lipoma, intracranial lipoma, quadrigeminal cistern, quadrigeminal plate, quadrigeminal lipoma, vertigo

INTRODUCTION

Lipoma is a common benign mesenchymal-induced tumor with variable distribution. Intracranial lipomas (ILs) are rare and account for 0.1% of all primary brain tumors [1,2]. They are usually diagnosed incidentally [3]. IL is located in the midline regions such as the corpus callosum and quadrigeminal plate [4]. Computed tomography (CT) and magnetic resonance imaging (MRI) are usually sufficient for diagnosing lipomas. Quadrigeminal lipomas (QL) generally present as asymptomatic. However, they can cause neurologic symptoms if they are large enough to cause a mass effect [5]. Symptomatic treatment and follow-up are the main management of these cases. Surgical removal of QL carries risks of postoperative complications and morbidity due to adjacent brainstem structures. Therefore, if the patient is asymptomatic, follow-up is recommended. We report a case of a 56-year-old male who presented with vertigo caused by quadrigeminal plate lipoma.

CASE PRESENTATION

A 56-year-old white male was admitted to the otolaryngology department with vertigo as his main complaint for the past five years. He also described experiencing multiple episodes of nausea and visual disturbances during this period. The patient

had a long history of seizures, treated by a neurologist. No other comorbidity was described. Family history was not remarkable. On physical examination, left-side hypesthesia was detected, but he had a normal gait and cerebellar and vestibular test results. Other findings were unremarkable. Routine laboratory tests were within the reference range. Blood tests were within the normal 60 ranges. In the MRI report, an approximately 4x3.3 10 mm lesion with a hyperintense signal on T1 and T2-weighted images was observed on the quadrigeminal cistern adjacent to the tectum (Figure 1A-E). Due to susceptibility artifacts, the lesion was hypointense on fat-saturated T1 and magnetic susceptibility weighted imaging sequences without post-contrast enhancement compatible with lipoma. No malformations or hydrocephalus were detected. The patient's condition was consulted with the neurosurgeons. Neurosurgery and neurology consultations resulted in symptomatic medical treatment and non-surgical follow-up. Betahistine 24 mg three times daily was planned and there was a slight improvement in the patient's vertigo complaint at 24-month follow-up.

DISCUSSION

ILs are very rare tumors whose development is poorly understood, with an incidence of about 0.1-0.5% of all brain tumors [2,4,6,7]. They are usually located in the midsagittal



Address for Correspondence: Aynur Aliyeva MD, PhD, Cincinnati Children's Medical Hospital, Clinic of Pediatric Otorhinolaryngology-Head and Neck Surgery, Ohio, USA

Phone: +1 513-636-4200 E-mail: dr.aynuraliyeva86@gmail.com ORCID ID: 0000-0001-9398-4261

Received: 20.04.2023 Accepted: 22.05.2023



©Copyright 2023 by the Azerbaijan Gastroenterology Invasive Endoscopy Society / Caucasian Medical Journal published by Galenos Publishing House. This journal is licensed under a Creative Commons Attribution-NonCommercial 3.0 International (CC BY-NC 3.0).

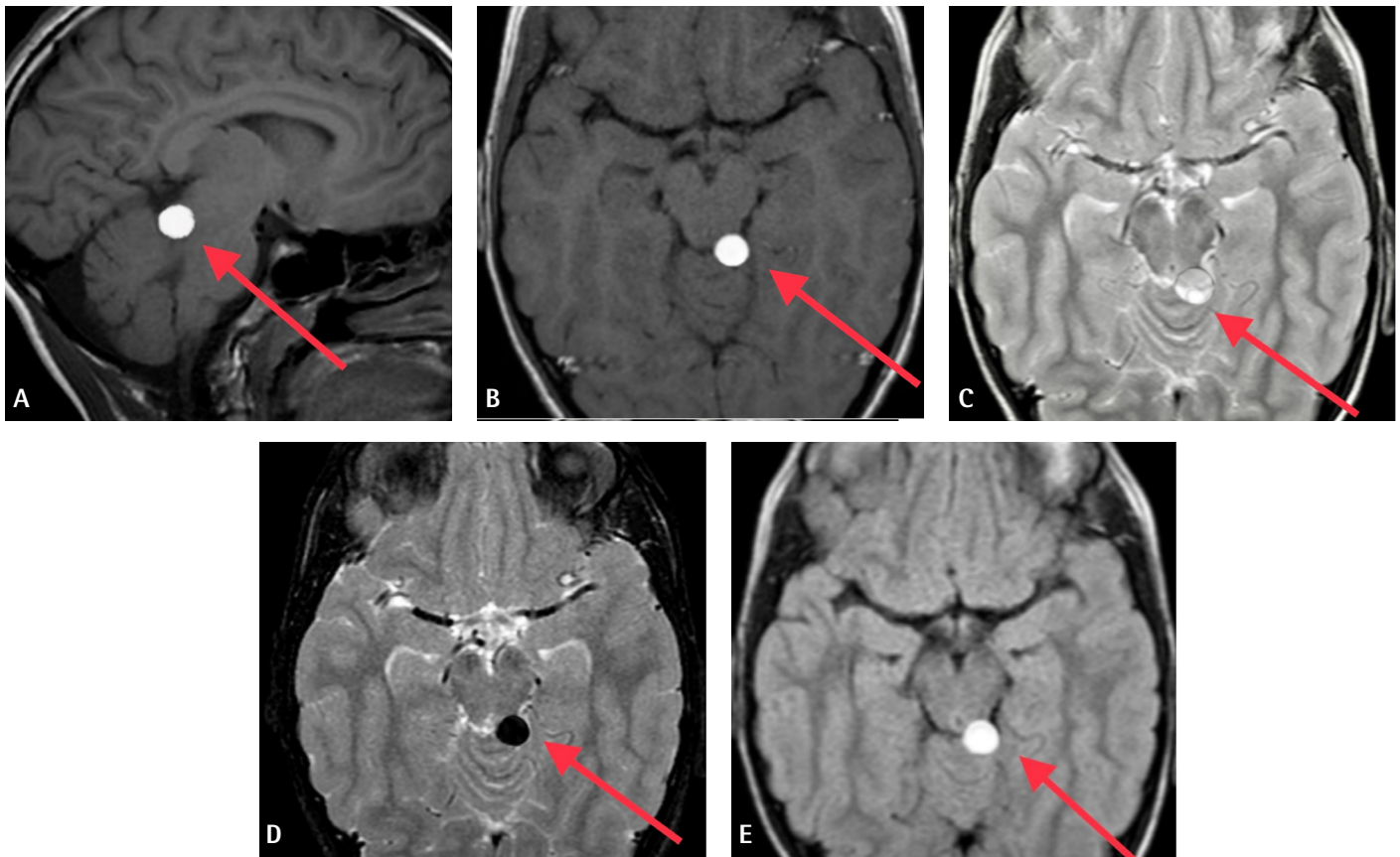


Figure 1. MRI images of the lipoma of the quadrigeminal cistern (the lesion is shown with a red arrow). (A) Sagittal T1, (B) axial T1, (C) axial T2, (D) axial T2-fat sat, (E) axial T2 FLAIR

MRI: Magnetic resonance imaging, FLAIR: Fluid-attenuated inversion recovery

region, mostly in the corpus callosum, quadrigeminal cistern, suprasellar cistern, cerebellopontine angle cistern, intercerebellar fissure, and Sylvian cistern regions [6-10].

The locations where lipomas frequently occur appear to correspond to a temporal sequence in which the primitive meninx, the embryonic tissue from which the meninges develop, dissolves during fetal development. This supports the idea that ILs form due to the meninx's abnormal persistence and differentiation. This developmental explanation for the formation of ILs also accounts for the frequent occurrence of brain hypoplasias [7]. CT and MRI can be used to diagnose IL, but MRI is an excellent imaging modality to accurately characterize these lesions and possible associated anomalies that can be considered more valuable than CT. The diagnosis is usually made based on imaging findings and does not require histological confirmation [2,4]. QL usually shows hyperintense on T1-weighted and mixed intensity on T2-weighted MRI [11,12]. Our case imaging findings are characteristic of a fat-containing lesion located in the quadrigeminal cistern. CT demonstrates a non-enhancing fat attenuating (HU: -50 to -100) mass in the quadrigeminal cistern without any peripheral calcification. MRI

reveals high signal intensity signal characteristics of fat in T1 and T2, no enhancement in T1 C+ (Gd), and low signal intensity fat-saturated sequences.

They are mostly asymptomatic, self-limited, slowly growing lesions, diagnosed incidentally, and require no therapy. However, they may sometimes present with headaches, seizures, dementia, tinnitus, diplopia, visual hallucinations, trochlear nerve palsy, or dropping of the eyelid that needs conservative or surgical treatment [11,13-18]. Symptomatic treatment with periodic follow-up is the main management of these cases. Surgical tumor excision is considered if the lesion causes several clinical symptoms.

Studies have reported that ILs, especially corpus callosum-located lipomas, are associated with central nervous system anomalies [2,4,19,20]. QLs are usually self-limited tumors, unlike this [11]. The mean age was 36-43 years [3,11,12,14,19]. Our patient was a 56-year-old male, older than reported in the literature. Most of the QLs were tubulolobular shapes [14]. In this study, the lesion was of the same shape. There is no significant difference between males and females regarding the incidence of IL [12,14,19].

The differential diagnosis of QL includes tectal plate cysts, arachnoid cysts, epidermoid cysts, gliomas, and pineal gland tumors within the differential diagnosis [21,22-24]. Subacute hemorrhage may show similar T1-weighted (W) MRI patterns. T2-W images and hypointense appearance in T1-W images with fat saturation are helpful in the differential diagnosis (4). QL may mimic intracranial air on CT; MRI is helpful in that situation [22].

Total excision of the lesion is the treatment option in symptomatic patients [5,25]. Ventriculoperitoneal shunt placement is an alternative in cases with hydrocephalus [24]. Surgical excision of tumors has risks for postoperative complications and morbidity because of proximity to the critical neural structures in the midbrain. Cerebellar ataxia, 6th cranial nerve paresis, and diplopia were reported as postoperative complications [23]. The infratentorial supracerebellar approach is preferred as the surgical approach to minimize the risk of complications [5,23,26].

Moreover, surgical tumor excision is considered only if the lesion causes several clinical symptoms. In the case of a patient with an asymptomatic QL, symptomatic treatment with periodic follow-up is the main management approaches. Alternative treatment options for symptomatic QL include ventriculoperitoneal shunt placement in cases with hydrocephalus. However, this option only relieves hydrocephalus symptoms, not the tumor itself. Additionally, it is important to note that conservative management is the best approach for asymptomatic lipomas. Diagnosing and managing them accurately is essential to prevent misdiagnosis and unnecessary procedures [27-29].

Our case presentation has some limitations that should be considered. First, we have no long-term patient outcomes. As it is an infrequent cause of vertigo, there is a lack of discussion on the possible causes or mechanisms of quadrigeminal cistern lipoma formation and their relation to vertigo. Despite these limitations, the case report provides valuable information on the rare condition and can serve as a starting point for future studies

Clinicians must know the characteristics of a quadrigeminal cistern lipoma to prevent misdiagnosis and unnecessary procedures. Based on current literature, this is the first reported case of a QL causing vertigo. We hope that this case report will make a significant contribution to the scientific community and increase awareness among clinicians in outpatient clinics.

Ethics

Informed Consent: The patient consented to the publication of this case report.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: A.A., Concept: Z.K., A.A., S.M., Design: Z.K., A.A., S.M., Data Collection or Processing: Z.K., A.A., S.M., Analysis or Interpretation: Z.K., A.A., S.M., Literature Search: Z.K., A.A., S.M., Writing: Z.K., A.A., S.M.

Conflict of Interest: No conflict of interest was declared by the authors.

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

References

1. Ichikawa T, Kumazaki T, Mizumura S, Kijima T, Motohashi S, et al. Intracranial lipomas: demonstration by computed tomography and magnetic resonance imaging. *J Nippon Med Sch.* 2000;67:388-91.
2. Thakkar DK, Patil A, Thakkar D, Jantre MN, Kulkarni VM, et al. Quadrigeminal cistern lipoma: a rare case report with review of literature. *Med J Dr DY Patil Univ.* 2015;8:267-70.
3. Sharma P, Maurya V, Ravikumar R, Bhatia M. Lipoma of the quadrigeminal plate cistern. *Med J Armed Forces India.* 2016;72(Suppl 1):S74-6.
4. Jabot G, Stoquart-Elsankari S, Saliou G, Toussaint P, Deramond H, et al. Intracranial lipomas: clinical appearances on neuroimaging and clinical significance. *J Neurol.* 2009;256:851-5.
5. Yilmazlar S, Kocaeli H, Aksoy K. Quadrigeminal cistern lipoma. *J Clin Neurosci.* 2005;12:596-9.
6. Yildiz H, Hakyemez B, Koroglu M, Yesildag A, Baykal B. Intracranial lipomas: importance of localization. *Neuroradiology.* 2006;48:1-7.
7. Truwit CL, Barkovich AJ. Pathogenesis of intracranial lipoma: an MR study in 42 patients. *AJR Am J Roentgenol.* 1990;155:855-64; discussion 865.
8. Ghazwani SM, Alzaki ES, Fadhel AM, Assiri KA, Alhabsi LW, et al. Sylvian fissure lipoma: an unusual etiology of seizures in adults. *Cureus.* 2022;14:e21407.
9. Kassimi M, Guerroum H, Amriss O, Habi J, Moutaouakil K, et al. Curvilinear pericallosal lipomas diagnosed incidentally during evaluation following trauma with corpus callosum abnormalities in two patients. *BJR Case Rep.* 2020;7:20200081.
10. Yilmaz N, Unal O, Kiyamaz N, Yilmaz C, Etlik O. Intracranial lipomas--a clinical study. *Clin Neurol Neurosurg.* 2006;108:363-8.
11. Uchino A, Hasuo K, Matsumoto S, Masuda K. MRI of dorsal mesencephalic lipomas. *Clin Imaging.* 1993;17:12-6.
12. Seidl Z, Vaneckova M, Vitak T. Intracranial lipomas: a retrospective study. *Neuroradiol J.* 2007;20:30-6.
13. Eftekharian K, Sharifi G, Eftekharian A, Bidari-Zerehpooch F. Contralateral tinnitus and hearing loss due to a tumor at the region of inferior colliculus: illustrative case. *J Neurosurg Case Lessons.* 2022;3:CASE21624.
14. Taydas O, Ogul H, Kantarci M. The clinical and radiological features of cisternal and pericallosal lipomas. *Acta Neurol Belg.* 2020;120:65-70.
15. Ammor R, Ajja A. Symptomatic quadrigeminal cistern lipoma. *Pan Afr Med J.* 2015;20:328.
16. Majumdar K, Saran RK, Tyagi I, Shankar R, Singh D. Role of intraoperative squash smear cytology as a diagnostic modality in lipoma of quadrigeminal cistern. *J Neurosci Rural Pract.* 2013;4:59-62.
17. Sehgal V, Patil PS, Mitra S. A rare presentation of quadrigeminal cistern (tectal plate) lipoma with visual hallucinations in a patient of schizophrenia: a case report. *Cureus.* 2022;14:e31132.
18. Choi NH, Kim WJ, Kim MM. Trochlear nerve palsy caused by quadrigeminal cistern lipoma. *J Korean Ophthalmol Soc.* 2018;59:1087-90.

19. Yilmaz MB, Egemen E, Tekiner A. Lipoma of the quadrigeminal cistern: report of 12 cases with clinical and radiological features. *Turk Neurosurg.* 2015;25:16-20.
20. Schell-Apacik CC, Cohen M, Vojta S, Ertl-Wagner B, Klopocki E, Heinrich U, et al. Gomez-Lopez-Hernandez syndrome (cerebello-trigeminal-dermal dysplasia): description of an additional case and review of the literature. *Eur J Pediatr.* 2008;167:123-6.
21. Ogbole G, Kazaure I, Anas I. Quadrigeminal plate cistern lipoma. *BMJ Case Rep.* 2009;2009:bcr07.2009.2110.
22. Mashiko R, Shibata Y. Quadrigeminal cistern lipoma mimicking intracranial air. *BMJ Case Rep.* 2014;2014:bcr2014203738.
23. Nikaido Y, Imanishi M, Monobe T. Lipoma in the quadrigeminal cistern--case report. *Neurol Med Chir (Tokyo).* 1995;35:175-8.
24. Kawamata T, Aoki N, Sakai T, Takakura K. Congenital triventricular hydrocephalus associated with a small lipoma in the quadrigeminal plate cistern. *Childs Nerv Syst.* 1995;11:121-3.
25. Sala F, Talacchi A, Scarpa P, Bricolo A. Surgical treatment of quadrigeminal plate lipoma presenting with seizures and behavioural disorders. *J Neurol Neurosurg Psychiatry.* 1998;64:818-9.
26. Le Feuvre DE, Semple PL, Peter JC. Intradural cervical lipomas with intracranial extension: a management strategy based on a case report and review of the literature. *Br J Neurosurg.* 2004;18:385-8.
27. Fandiño J, Bermúdez J, Arán E. Lipoma de la cisterna cuadrigémina y cisura calcarina: caso clínico y revisión de la literatura [Quadrigeminal cistern and calcarine fissure lipoma: case report and review of the literature]. *Neurocirugia (Astur).* 2005;16:173-6.
28. Ahmadi SA, van Landeghem FK, Blechschmidt C, Lieber K, Haberl EJ, et al. Intratentorial osteochondrolipoma in a 9-year-old boy. *J Neurosurg Pediatr.* 2009;3:386-91.
29. Elgassim MAM, Wafer A, Ahmed A, Elfaki A, Satti A, et al. Intracranial lipoma extending extracranially in a five-year-old patient. *Cureus.* 2022;14:e21816.

Association of *Chlamydia pneumoniae* and *Borrelia burgdorferi* Infections with Multiple Sclerosis

 Ulvi Zahir Nasirov

Mediland Hospital, Microbiology Laboratory, Baku, Azerbaijan

ABSTRACT

Many pathogens, including *Chlamydia pneumoniae* and *Borrelia burgdorferi*, have been associated with multiple sclerosis (MS). MS can be significant pathology in some patients who are infected with *Chlamydia pneumoniae* and *Borrelia burgdorferi*. In MS, the immune system attacks the myelin sheath that covers nerve fibers, resulting in the interruption of communication between the brain and body. This damage may be permanent.

Keywords: *Chlamydia pneumoniae*, *Borrelia burgdorferi*, multiple sclerosis

INTRODUCTION

In multiple sclerosis (MS), the immune system attacks the myelin sheath that covers nerve fibers, resulting in the interruption of communication between the brain and body. This damage may be permanent.

The symptoms of MS vary widely among patients and depend on the location of the damage of the nerve fibers in the central nervous system and severity of the damage. Some individuals with severe MS may lose the ability to walk or move independently, while others may experience long periods of remission without new symptoms, depending on the type of MS they have.

Common symptoms;

Numbness and weakness in one or more parts of the body at the same time, tingling sensation, electric shock sensations with certain neck movements, especially forward bending of the neck (Lhermitte's sign), the lack of coordination, unstable balance or inability to walk, the partial or complete loss of vision, double vision for a long time, blurred vision, vertigo, problems related to sexual, bowel, and bladder function fatigue, weak speech.

CASE PRESENTATION

Our patient is a 30-year-old individual who occasionally experiences severe headaches, muscle weakness, and severe muscle pain. Numbness is felt in both legs and arms. After magnetic resonance imaging examination, the patient was

diagnosed with MS. However, more interesting facts are revealed after taking a deeper medical history. A year ago, the patient had pneumonia and despite using certain antibiotics, recovery was delayed (not atypical pneumonia, but bacterial pneumonia was considered).

When taking a thorough medical history, it was revealed that the patient had rash on the soles of their feet for a long time and felt itching for a while. Our research is ongoing. After all, these clinical signs, laboratory tests were requested.

DISCUSSION

We initially requested laboratory tests for *Chlamydia pneumoniae* and *Borrelia burgdorferi*, and the results were very interesting [1]. Both *Chlamydia pneumoniae* and *Borrelia burgdorferi* immunoglobulin G tested positive [2]. Treatment was started immediately. The treatment included a combination of doxycycline, azithromycin, and metronidazole as advised by an infectious disease specialist.

Two months after starting the treatment, relapses continued. After 7 months, the patient's complaints were significantly reduced, and there were no new symptoms.

The treatment lasted 12 months and in the end there was complete remission, no complaints. Currently, 3 years after treatment, there is no recurrence [3].

Ethics

Informed Consent: Patient consent was obtained.

Peer-review: Externally peer-reviewed.



Address for Correspondence: Ulvi Zahir Nasirov MD, Mediland Hospital, Microbiology Laboratory, Baku, Azerbaijan

Phone: +994 515284179 E-mail: dr.nesirov@gmail.com ORCID ID: 0009-0008-1701-4552

Received: 14.04.2023 Accepted: 12.07.2023



©Copyright 2023 by the Azerbaijan Gastroenterology Invasive Endoscopy Society / Caucasian Medical Journal published by Galenos Publishing House. This journal is licensed under a Creative Commons Attribution-NonCommercial 3.0 International (CC BY-NC 3.0).

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

REFERENCES

1. Ivanova MV, Kolkova NI, Morgunova EY, Pashko YP, Ziganirova NA, et al. Role of Chlamydia in multiple sclerosis. *Bull Exp Biol Med.* 2015;159:646-8.
2. Fainardi E, Castellazzi M, Casetta I, Cultrera R, Vaghi L, et al. Intrathecal production of Chlamydia pneumoniae-specific high-affinity antibodies is significantly associated to a subset of multiple sclerosis patients with progressive forms. *J Neurol Sci.* 2004;217:181-8.
3. Fritzsche M. Chronic Lyme borreliosis at the root of multiple sclerosis--is a cure with antibiotics attainable? *Med Hypotheses.* 2005;64:438-48.

Leptospirosis Case

Ulvi Zahir Nasirov, Gulam Rustamzada

Mediland Hospital, Microbiology Laboratory, Baku, Azerbaijan

ABSTRACT

Leptospirosis is a zoonotic infection that is more commonly found in tropical regions. This spirochete is motile, aerobic, and spiral-shaped with two species: *Leptospira interrogans* and *Leptospira biflexa*. The type that causes infection in humans is *L. interrogans*. The main entry points for infection in humans are the skin and mucosa (cuts and abrasions on the skin and conjunctiva). Animals spread the infection through urine, which is why water pollution plays a significant role in transmission. These bacteria can survive in water for several weeks. Drinking water plays little role in transmission. The incubation period is 2-25 days. After the incubation period, the bacteria spread through the bloodstream and attach to parenchymatous organs, particularly kidneys and liver, causing a rise in temperature.

Keywords: Leptospirosis, Weil's disease, jaundice, renal failure

INTRODUCTION

Leptospirosis is a zoonotic infection that is more commonly found in tropical regions [1-5]. This spirochete is motile, aerobic, and spiral-shaped with two species: *Leptospira interrogans* and *Leptospira biflexa*. The type that causes infection in humans is *L. interrogans* [6]. The main entry points for infection in humans are skin and mucosa (cuts and abrasions on the skin and conjunctiva). Animals spread the infection through urine, which is why water pollution plays a significant role in transmission. These bacteria can survive in water for several weeks. Drinking water plays little role in transmission. The incubation period is 2-25 days. After the incubation period, the bacteria spread through the bloodstream and attach to parenchymatous organs, particularly kidneys and liver, causing a rise in temperature.

Non-icteric forms are observed in 90% of patients with leptospirosis, while Weil's disease, which is characterized by fever, jaundice, bleeding tendency, acute liver and renal failure, is observed in 5-10% of cases. The mortality rate for this disease, which is accompanied by severe jaundice and hepatorenal failure, is very high [2].

Routine laboratory tests are not specific for diagnosing leptospirosis. Diagnosis is made on the basis of the doctor's request and clinical examination, and blood and urine culture and serological tests. Early initiation of antibiotic therapy

plays an important role in controlling infection and reducing mortality [3].

CASE PRESENTATION

A 27-year-old patient with complaints of headache, nausea, fever and gradually increasing jaundice was admitted to the intensive care unit of our hospital. After admission, jaundice, encephalopathy, high fever, and acute renal failure were detected.

The temperature was 37.4 °C, pulse was 140-142 (sinus tachycardia), A/T 90-50, and complete blood count revealed high neutrophil count, and biochemical analysis showed high levels of jaundice and renal failure (Table 1).

DISCUSSION

Weil's disease is a severe clinical form of *Leptospira* infection. In these individuals, the disease can rapidly progress over 5-10 days and can lead to clinical conditions such as jaundice, kidney and liver failure, hypotension, and even coma.

This patient presented with jaundice, bleeding from the mouth and nose, fever, thrombocytopenia, and liver and kidney failure. The presence of hyperbilirubinemia and partially liver failure necessitated further investigation, such as ultrasound examination, to identify any pathology that could



Address for Correspondence: Ulvi Zahir Nasirov MD, Mediland Hospital, Microbiology Laboratory, Baku, Azerbaijan

Phone: +994 515284179 E-mail: dr.nesirov@gmail.com ORCID ID: 0009-0008-1701-4552

Received: 14.04.2023 Accepted: 13.07.2023



©Copyright 2023 by the Azerbaijan Gastroenterology Invasive Endoscopy Society / Caucasian Medical Journal published by Galenos Publishing House. This journal is licensed under a Creative Commons Attribution-NonCommercial 3.0 International (CC BY-NC 3.0).

Table 1. Levels of jaundice and renal failure

	1 day	3 day	7 day	7 day
Hemoglobin (g/dL)	8.8	10.9	8.9	9.5
Leukocyte (mm ³)	24.54	25.79	23.36	11.96
Platelet (mm ³)	104	122	188	450
SGPT (U/L)	25	62	27.0	25.1
SGOT (U/L)	71.7	78	26.1	24.3
Total bilirubin (mg/dL)	53.5	42.3	27.3	14.6
Direct bilirubin (mg/dL)	39.6	32.9	-	-
Na (mmol/L)	136	139	137	140
K (mmol/L)	3.0	3.5	3.8	4.1
Creatinine (mg/dL)	8.2	5.35	2.67	1.88
CRP (mg/dL)	9.4	3.0	1.9	0.9
Procalcitonin (ng/mL)	7.9	6.4	1.51	0.27

SGPT: Serum glutamic oxaloacetic transaminase, SGOT: Serum glutamate pyruvate transaminase, CRP: C-reactive protein

cause jaundice. However, no pathology that could lead to jaundice in the liver or urinary tract was detected during this examination. Although the clinical picture was similar to toxic and viral hepatitis, no virus or toxic substance was identified in the laboratory tests. Toxic and viral hepatitis were excluded. Until the diagnosis was confirmed by laboratory tests, the patient was given penicillin and tetracycline antibiotics, and treatment was continued to maintain electrolyte balance.

Considering the patient's lifestyle, sanitation conditions, and clinical condition, the physician investigated leptospirosis infection at the request of the clinical specialist. *Leptospira* IgM was detected as a positive in the initial examination [4].

One of the main reasons for the high mortality rate in Weil's disease is the development of kidney failure. Hypovolemia, vasoconstriction after endotoxin secretion, ischemia, acute tubular necrosis, and other processes lead to kidney failure.

Despite being a rare infection, the unique aspect of this case was the rapid change in the patient's clinical condition and laboratory parameters (note Table 1, the direct bilirubin increased to 39.6 mg/dL, which is a rare occurrence).

After an accurate diagnosis was made, the patient's clinical condition improved with proper treatment, and this was also reflected in the laboratory tests. The patient was discharged on the condition of following the recommendations.

Ethics

Informed Consent: Informed consent was obtained from the patient.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: U.Z.N., G.R., Concept: U.Z.N., G.R., Design: U.Z.N., G.R., Data Collection or Processing: U.Z.N., G.R., Analysis or Interpretation: U.Z.N., G.R., Literature Search: U.Z.N., G.R., Writing: U.Z.N., G.R.

Conflict of Interest: No conflict of interest was declared by the authors.

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

REFERENCES

- Cardoso J, Gaspar A, Esteves C. Severe leptospirosis: a case report. *Cureus*. 2022;14:e30712.
- Rozalena S, Handayani L, Arman A, Permata M, Hudari H. Weil's disease in a 36 years old female: a case report. *IOP Conference Series: Earth and Environmental Science*. 2018;125:012066.
- Valarezo-Sevilla D, Sarzosa-Terán V. Leptospirosis: serie de casos en un centro penitenciario de la costa de Ecuador [Leptospirosis: case-series report in a prison of the coast in Ecuador]. *Rev Esp Sanid Penit*. 2014;16:20-3. Spanish.
- Faggion Vinholo T, Ribeiro GS, Silva NF, Cruz J, Reis MG, et alF. Severe leptospirosis after rat bite: a case report. *PLoS Negl Trop Dis*. 2020;14:e0008257.
- Kularathna MDSV, Kularatne SAM, Pathirage M, Nanayakkara PTMA. Severe leptospirosis complicated with multiorgan dysfunction successfully managed with plasma exchange: a case report. *J Med Case Rep*. 2021;15:584.
- Çelik M, Gözübüyük AA, Ceylan MR, Cesur S, Esmer F. Leptospirosis: a case repor. *Troia Med J*. 2021;2:115-7.

Breast Metastasis from Primary Lung Adenocarcinoma: A Rare Occurrence with Diagnostic Challenges and Treatment Implications

 Rizvan Rustamov,  Masoud Mireskandari,  Iver Petersen

SRH Poliklinik Gera GmbH, SRH Wald-Klinikum Gera, Institute of Pathology, Gera, Germany

ABSTRACT

Breast metastasis from non-mammary sites is a rare occurrence with a prevalence ranging from 0.5 to 3%. Accurate diagnosis is crucial as it influences treatment planning and management. Distinguishing primary breast cancer from metastatic disease can be challenging, and an incorrect diagnosis can result in unnecessary surgical interventions. Immunohistochemistry plays a crucial role in identifying the primary tumor origin, especially when cytological patterns appear atypical. Here we present a case of a 53-year-old female patient with breast metastases from a primary lung adenocarcinoma. Pathology from the lung biopsy revealed a well to moderately differentiated adenocarcinoma with epidermal growth factor receptor mutation. Immunohistochemical analysis using specific markers such as TTF-1 and GATA-3 was instrumental in determining the malignant cell origin and formulating a treatment plan. This report highlights the importance of early diagnosis, thorough investigation and the role of immunohistochemical markers in managing this rare occurrence.

Keywords: Breast metastasis, lung cancer, core biopsy, EGFR mutation

INTRODUCTION

Breast metastasis from non-mammary sites is a rare occurrence, with an incidence of 0.5% to 3%. Accurate diagnosis is crucial as it influences treatment planning and management. Distinguishing between primary breast cancer and metastatic disease can present diagnostic challenges, potentially leading to unnecessary surgical interventions. The distinction becomes even more critical when cytological patterns appear atypical, necessitating the use of complementary diagnostic tools. Metastasis to the breast from extra mammary malignancies, such as lung adenocarcinoma, is an unexpected finding due to the relatively uncommon nature of such events. Lung neoplasms can metastasize through hematologic and lymphatic routes with distant metastases most commonly affecting organs such as the liver, adrenal glands, bones, and brain. However, the breast involvement remains rare, making its diagnosis particularly challenging. In this context, we present a case report of a 53-year-old female patient with breast metastases from a primary lung adenocarcinoma. Our aim is to emphasize the clinical significance of early and accurate diagnosis and the challenges associated with differentiating primary breast cancer

from metastatic disease. The role of immunohistochemistry (IHC) in assisting with the identification of malignant cell origin and the formulation of a tailored treatment plan will also be discussed.

CASE PRESENTATION

A 53-year-old female patient was referred to the ENT station by the maxillofacial surgeon because of confirmed adenocarcinoma manifestations in the right occipital area. The primary tumor has not yet been found. Complaints regarding the shortness of breath, cough, hemoptysis, or B-symptomatic were denied. She had no pre-existing oncological disease. Only the patient's mother had a malignancy of the abdomen (uterus, not further specified), otherwise, she had no family history of known risk factors and no history of exposure to noxious agents. Computed tomography-thorax imaging with contrast medium revealed some pulmonary changes but not a clear tumor mass. It showed an unclear structure in the caudomedial area of the left breast. The patient underwent a bronchoscopy with EBUS and a lung biopsy. The histopathologic examination of biopsies and cytology materials indicated a well to moderately



Address for Correspondence: Rizvan Rustamov MD, SRH Poliklinik Gera GmbH, SRH Wald-Klinikum Gera, Institute of Pathology, Gera, Germany

Phone: +4915129469205 E-mail: rizvan.rustamov1991@gmail.com ORCID ID: 0000-0002-9892-4142

Received: 11.06.2023 Accepted: 27.07.2023



©Copyright 2023 by the Azerbaijan Gastroenterology Invasive Endoscopy Society / Caucasian Medical Journal published by Galenos Publishing House. This journal is licensed under a Creative Commons Attribution-NonCommercial 3.0 International (CC BY-NC 3.0).

differentiated adenocarcinoma. In further molecular analyzes, an epidermal growth factor receptor mutation [c.2573T>G, p. Leu858Arg (27%, NM_005228.5)] was detected in tumor cells. The tumor infiltration was primarily in the lingula area with the involvement of the distal and medial main bronchus on the left side.

With suspicion of a second carcinoma in the breast tissue, a core biopsy of the breast mass was performed. Histopathological evaluation revealed adenocarcinoma with a predominantly tubulopapillary growth pattern. Primary immunohistochemical evaluations showed a triple negative adenocarcinoma (ER, PR, and Her2 negative). Considering the recently detected bronchial adenocarcinoma of the lung and the untypical histological image for a primary triple-negative breast carcinoma, a diagnosis of breast metastasis by tumor cells of bronchial carcinoma was considered. This assumption could be confirmed by additional immunohistochemical evaluations. The tumor cells in the breast biopsy were also negative for GATA-3 and mammaglobin. However, they were strongly positive for CK7 and TTF1 (Figure 1). The results of conventional histopathological and immunohistochemical evaluations confirmed breast metastasis by bronchial carcinoma.

Further imaging analyzes revealed meningiosis carcinomatosa with subarachnoid tumor nodes without any involvement of brain tissue. Magnetic resonance imaging of the spine also showed multifocal bone metastases with complete infiltration of thoracic vertebrae 11 and 4 with a suspected risk of fracture. In addition, larger metastases were detected near the baseplate in thoracic vertebra 12 and left in sacral vertebra 1 without evidence of a clear spread into the spinal canal.

DISCUSSION

Lung adenocarcinoma is the most common subtype of non-small-cell lung cancer, accounting for approximately 40% of all lung cancer cases [1]. Metastatic disease of the breast is an uncommon occurrence with reported clinical cases ranging from 0.2% to 2.7% [2,3]. Metastatic cancer presenting as a breast mass is an unexpected finding in female patients because it is relatively uncommon due to the large area of fibrous tissue and poor blood supply in the breast [4]. According to a study by Riihimäki et al. [5], metastases from lung adenocarcinoma most frequently occur in the bone, liver, and brain, while metastases to the breast are relatively rare.

Accordingly, the presence of synchronous tumor masses in the lung and breast of a patient is more consistent with two distinct

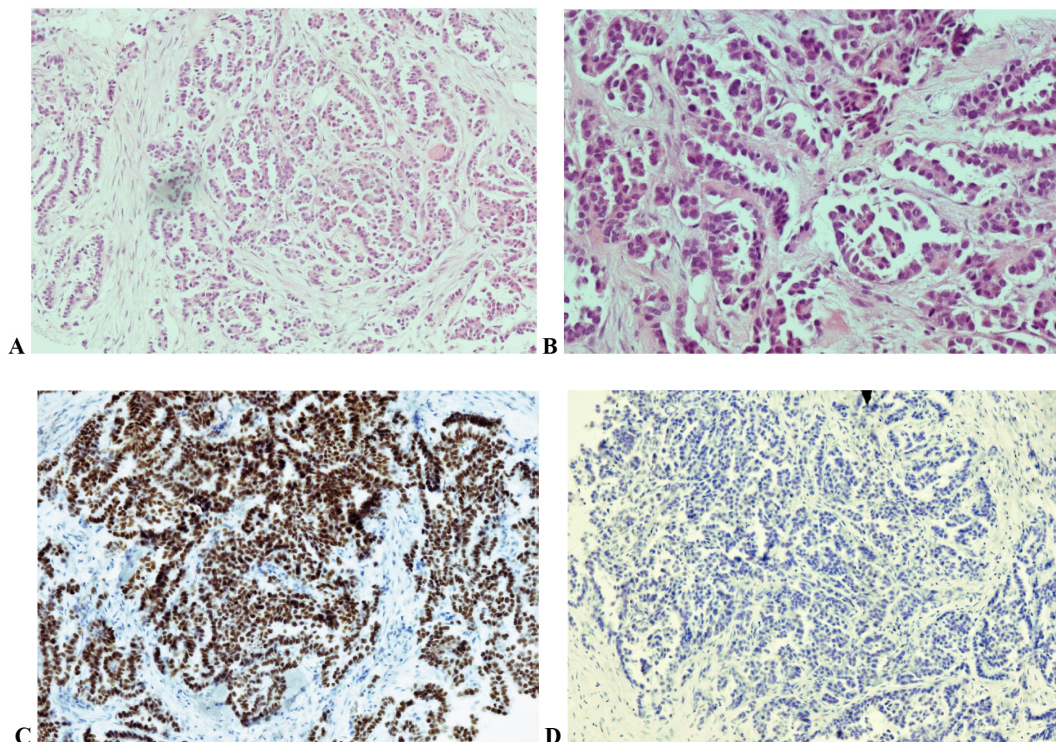


Figure 1. (A, B) The biopsy of the breast shows a tubulopapillary epithelial growth of invasive adenocarcinoma (H and E, $\times 100$; $\times 200$); (C, D) immunohistochemical staining of the tumor shows widespread nuclear positivity for TTF-1 and negative for GATA-3 (H and E, $\times 100$)

H and E: Hematoxylin and eosin

and independent malignancies or breast carcinoma with lung metastasis than bronchial carcinoma with breast metastasis. In this context, the correct diagnosis of breast metastasis from bronchial carcinoma requires a high index of suspicion and the application of an appropriate profile of immunohistochemical findings. One of the key diagnostic features in this case was the histopathological appearance of the tumor in breast biopsy. The histologic appearance of carcinoma in the breast was not typical for triple-negative breast carcinoma (TNBC). TNBC accounts for approximately 10-20% of all breast cancer cases and is associated with a poor prognosis [1]. Except for specific triple-negative low-grade tumors of the breast (adenoid-cystic carcinoma of the breast and micro-glandular adenosis), most triple-negative breast cancers are high-grade tumors with necrosis, show aggressive biological behavior and have a higher rate of distant metastases compared to other subtypes of breast cancer [6-8]. In one study, 18% of TNBCs showed lung metastasis in 5-year follow-up, compared to 7% for non-TNBCs [9].

The clinical presentation of breast metastases from lung cancer can vary widely and may include palpable masses, nipple discharge, and skin changes. In some cases, breast metastases may be the first sign of underlying lung cancer. In most cases, metastatic disease of the breast occurs after the primary tumor has been diagnosed. However, in approximately 25% of patients, a breast mass is the initial mode of presentation [10]. Breast metastases from extramammary sites can often present as solitary breast masses and can be challenging to distinguish from primary breast malignancies or benign diseases. A potential list of usual sources of metastasis to the breast includes lung, ovarian, gastrointestinal, and soft tissue tumors [11,12].

The use of immunohistochemical staining in this case played a crucial role in confirming the diagnosis of metastatic lung adenocarcinoma. This finding is consistent with previous studies showing that the use of immunohistochemical markers can help differentiate between primary and metastatic tumors [1]. Several markers can be helpful in differentiating metastatic breast tumors from primary breast carcinoma. TTF-1 and napsin-A are helpful in identifying lung adenocarcinoma, whereas PAX-8 and CA-125 are useful for ovarian cancer. Gastrointestinal tumors can be identified using CK20, CDX-2, and villin, and soft tissue tumors can be identified using markers such as S-100, smooth muscle actin, and desmin [13,14].

In addition, molecular profiling using next-generation sequencing (NGS) can be useful in identifying the origin of metastatic tumors [15,16]. Overall, the use of IHC and NGS can aid in the diagnosis of metastatic breast tumors and help differentiate them from primary breast carcinoma, ultimately guiding appropriate treatment strategies.

In conclusion, breast metastasis from non-mammary sites is a rare occurrence that can pose diagnostic challenges for clinicians. Metastasis of lung adenocarcinoma to the breast represents a triple-negative carcinoma and should be considered in differential diagnosis by a pathologist, particularly when the histological findings are not completely typical for triple-negative breast carcinoma. Accurate diagnosis is important and helps prevent unnecessary breast surgery or additional tumor therapy.

Ethics

Informed Consent: Informed consent was obtained from the patient.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: R.R., M.M., I.P., Concept: R.R., M.M., I.P., Data Collection or Processing: R.R., M.M., I.P., Analysis or Interpretation: R.R., M.M., I.P., Literature Search: R.R., M.M., I.P., Writing: R.R., M.M., I.P.

Conflict of Interest: No conflict of interest was declared by the authors.

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

REFERENCES

1. Travis WD, Brambilla E, Nicholson AG, Yatabe Y, Austin JHM, et al. The 2015 World Health Organization Classification of Lung Tumors: Impact of Genetic, Clinical and Radiologic Advances Since the 2004 Classification. *J Thorac Oncol.* 2015;10:1243-60.
2. Lee AH. The histological diagnosis of metastases to the breast from extramammary malignancies. *J Clin Pathol.* 2007;60:1333-41.
3. Lee SK, Kim WW, Kim SH, Hur SM, Kim S, et al. Characteristics of metastasis in the breast from extramammary malignancies. *J Surg Oncol.* 2010;101:137-40.
4. Yeh CN, Lin CH, Chen MF. Clinical and ultrasonographic characteristics of breast metastases from extramammary malignancies. *Am Surg.* 2004;70:287-90.
5. Riihimäki M, Hemminki A, Fallah M, Thomsen H, Sundquist K, et al. Metastatic sites and survival in lung cancer. *Lung Cancer.* 2014;86:78-84.
6. Foulkes WD, Smith IE, Reis-Filho JS. Triple-negative breast cancer. *N Engl J Med.* 2010;363:1938-48.
7. Mhamdi HA, Kourie HR, Jungels C, Aftimos P, Belbaraka R, et al. Adenoid cystic carcinoma of the breast - an aggressive presentation with pulmonary, kidney, and brain metastases: a case report. *J Med Case Rep.* 2017;11:303.
8. Guerini-Rocco E, Piscuoglio S, Ng CK, Geyer FC, De Filippo MR, et al. Microglandular adenosis associated with triple-negative breast cancer is a neoplastic lesion of triple-negative phenotype harbouring TP53 somatic mutations. *J Pathol.* 2016;238:677-88.
9. Lin NU, Claus E, Sohl J, Razzak AR, Arnaout A, et al. Sites of distant recurrence and clinical outcomes in patients with metastatic triple-negative breast cancer: high incidence of central nervous system metastases. *Cancer.* 2008;113:2638-45.
10. Gupta D, Merino MI, Farhood A, Middleton LP. Metastases to breast simulating ductal carcinoma in situ: report of two cases and review of the literature. *Ann Diagn Pathol.* 2001;5:15-20.

11. Babu KS, Roberts F, Bryden F, McCafferty A, Downer P, et al. Metastases to breast from primary lung cancer. *J Thorac Oncol.* 2009;4:540-2.
12. Malek D, Buccheri S, Dey CB, Samli B, Plemmons J. Lung cancer metastasis to the breast mimicking inflammatory breast carcinoma on imaging. *Radiol Case Rep.* 2019;14:1500-5.
13. Moreno-Astudillo L, Villaseñor-Navarro Y, Sánchez-Goytia V, Porras-Reyes F, Lara-Mercado A, et al. A case series of breast metastases from different extramammary malignancies and their literature review. *Case Rep Radiol.* 2019 Jan 8;2019:9454201.
14. Semba R, Horimoto Y, Arakawa A, Saito M. Metastatic breast tumors from extramammary malignancies: a case series. *Surg Case Rep.* 2021;7:154.
15. Fernandes GS, Marques DF, Girardi DM, Braghiroli MIF, Coudry RA, et al. Next-generation Sequencing-based genomic profiling: fostering innovation in cancer care? *Clinics (Sao Paulo).* 2017;72:588-94.
16. Yu B, Wang Q, Liu X, Hu S, Zhou L, et al. Case report: molecular profiling assists in the diagnosis and treatment of cancer of unknown primary. *Front Oncol.* 2022;12:723140.

A Case of Esophageal Leiomyoma Resected After Selective Circular Myotomy in Submucosal Tunneling Endoscopic Resection

Abdullah Murat Buyruk

Ege University Faculty of Medicine, Department of Internal Medicine, Division of Gastroenterology, İzmir, Turkey

ABSTRACT

Submucosal tunneling endoscopic resection (STER) is one of the recommended treatment modalities in patients with subepithelial lesions (SEL). There is limited information in the literature about the applicability of STER in SEL originating from the deep muscularis propria (MP) layer. This report presents a case of SEL originating from the deep MP layer that underwent STER in which challenging dissection at the base of the lesion was facilitated by selective circular myotomy proximal to the lesion.

Keywords: Submucosal tunneling endoscopic resection, selective circular myotomy, subepithelial lesion

INTRODUCTION

Advances in endoscopy have increased the detection rate of subepithelial lesions (SEL) [1]. The majority of SELs are found in the upper gastrointestinal tract (most frequently found in the esophagus, esophagogastric junction, gastric cardiac) [2]. The treatment for symptomatic SEL is uncertain, and the main options are endoscopic resection [endoscopic mucosal resection, endoscopic submucosal excavation, endoscopic submucosal dissection (ESD), endoscopic full-thickness resection, and submucosal tunneling endoscopic resection (STER)] and surgery [3]. Among these methods, STER is a safe and less invasive approach in the treatment of SEL, especially that originating from the deep muscularis propria (MP) layer. When submucosal tunneling is feasible, STER is the safest technique because mucosal flap entry is easier to close than ESD or EFTR. The most suitable locations for the STER method are the middle or lower esophagus and gastric cardiac [3,4].

This report presents a case of SEL originating from the deep MP layer that underwent STER in which challenging dissection at the base of the lesion was facilitated by selective circular myotomy (SCM) proximal to the lesion.

CASE PRESENTATION

A 52-year-old female patient was admitted to our gastroenterology clinic with complaints of dysphagia for the past 10 months. Esophagogastroduodenoscopy showed a SEL

approximately 25 cm in length that caused partial esophageal obstruction on the anterior wall of the proximal esophagus (Figure 1). Endosonographic examination revealed a 28×20 mm hypoechoic SEL originating from the MP layer diameter. Informed consent was obtained from the patient after which STER was performed. A 3 mm flush knife was used for tunneling



Figure 1. Esophagogastroduodenoscopy showed a SEL approximately 25 cm in length that caused partial esophageal obstruction on the anterior wall of the proximal esophagus

SEL: Subepithelial lesion



Address for Correspondence: Asst. Prof. Abdullah Murat Buyruk, Ege University Faculty of Medicine, Department of Internal Medicine, Division of Gastroenterology, İzmir, Turkey

Phone: +90 232 343 43 43 E-mail: amuratbuyruk@gmail.com ORCID ID: 0000-0002-7233-2042

Received: 02.07.2023 Accepted: 27.07.2023



©Copyright 2023 by the Azerbaijan Gastroenterology Invasive Endoscopy Society / Caucasian Medical Journal published by Galenos Publishing House. This journal is licensed under a Creative Commons Attribution-NonCommercial 3.0 International (CC BY-NC 3.0).

at 5 cm proximal to the SEL to access the lesion. Submucosal dissection was first performed to separate the lesion from the mucosal layer, but the lesion was found to originate from the deep MP, which complicated the dissection at the base of the lesion. Bleeding during blunt dissection at the base of the lesion using an insulation-tipped knife further blurred the already limited view at the base of the lesion. To locate the bleeding site, a 1 cm SCM was performed immediately proximal to the lesion, which made the base of the lesion visible. STER + proximal SCM not only made the bleeding site visible but also facilitated dissection (Figure 2). Following this procedure, which lasted approximately 28 min, SEL was resected en bloc (Figure 3). No complications occurred after the procedure. Histopathological examination confirmed leiomyoma diagnosis.

DISCUSSION

STER was inspired by peroral endoscopic myotomy for resection of upper gastrointestinal SEL, especially those arising from the MP layer [4]. This technique is highly effective (en bloc resection: 94.6%, complete resection: 97.5%) but its success may be influenced by tumor depth. Procedures for tumors originating from the deep MP layer may last longer, have lower rates of en bloc resection, and have an increased risk of complications [5]. The width of the tunneling space may facilitate submucosal dissection, but damage to the tumor capsule and bleeding may occur during the deep muscular dissection. Cai et al. [6] reported in a limited case series that STER is also a feasible, safe, and effective endoscopic technique to achieve curative resection for extraluminal SEL. In their case series, full-thickness myotomy was performed above the estimated location of the



Figure 2. One cm selective circular myotomy was performed immediately proximal to the SEL, which made the base of the lesion visible
SEL: Subepithelial lesion

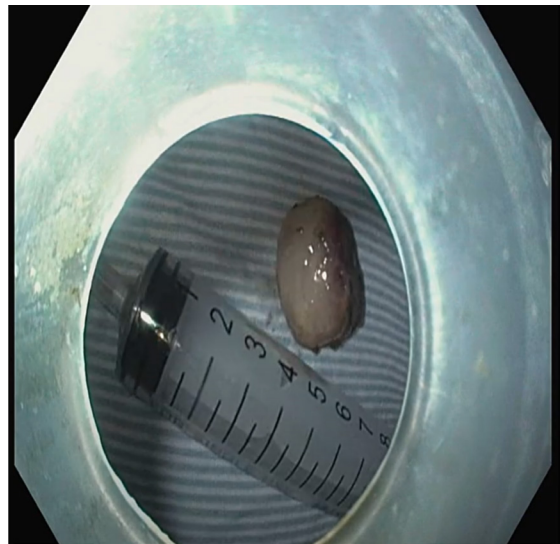


Figure 3. SEL was resected en bloc
SEL: Subepithelial lesion

extraluminal tumor [6]. In this study, since the SEL was located with the deep MP, proximal selective myotomy was sufficient. As in our case, when the source of bleeding cannot be found from the deep layer of the SEL or deep muscle dissection becomes difficult, STER + proximal SCM may be an effective and rescue method.

In our clinic, we have recently gained a greater experience of third-space endoscopic procedures. We have been using the STER procedure more frequently than before. As a result, we believe that using the STER + proximal SCM technique in SELs originating from the deep MP layer can facilitate dissection at the base of the lesion and reduce the risk of complications.

Ethics

Informed Consent: Informed consent was obtained.

Peer-review: Externally peer-reviewed.

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

REFERENCES

1. Lim YJ, Son HJ, Lee JS, Byun YH, Suh HJ, et al. Clinical course of subepithelial lesions detected on upper gastrointestinal endoscopy. *World J Gastroenterol.* 2010;16:439-44.
2. Menon L, Buscaglia JM. Endoscopic approach to subepithelial lesions. *Therap Adv Gastroenterol.* 2014;7:123-30.
3. Standards of Practice Committee; Faulx AL, Kothari S, Acosta RD, Agrawal D, et al. The role of endoscopy in subepithelial lesions of the GI tract. *Gastrointest Endosc.* 2017;85:1117-32.
4. Xu MD, Cai MY, Zhou PH, Qin XY, Zhong YS, et al. Submucosal tunneling endoscopic resection: a new technique for treating upper GI submucosal tumors originating from the muscularis propria layer (with videos). *Gastrointest Endosc.* 2012;75:195-9.

5. Dellatore P, Bhagat V, Kahaleh M. Endoscopic full thickness resection versus submucosal tunneling endoscopic resection for removal of submucosal tumors: a review article. *Transl Gastroenterol Hepatol.* 2019;4:45.
6. Cai MY, Zhu BQ, Xu MD, Qin WZ, Zhang YQ, et al. Submucosal tunnel endoscopic resection for extraluminal tumors: a novel endoscopic method for en bloc resection of predominant extraluminal growing subepithelial tumors or extra-gastrointestinal tumors (with videos). *Gastrointest Endosc.* 2018;88:160-7.

Carbon Dioxide Cholangiography in Diagnosis of Gallbladder Perforation: Deciphering Black, and White

İsmail Caymaz¹, Anar Mammadov², Mahmut Bilal Doğan³, Ayşenur Buz Yaşar⁴

¹Istanbul Medeniyet University Faculty of Medicine, Göztepe Training and Research Hospital, Department of Interventional Radiology, Istanbul, Turkey

²Muğla Sıtkı Koçman University Faculty of Medicine, Muğla, Turkey

³Istanbul Medeniyet University Faculty of Medicine, Göztepe Training and Research Hospital, Department of Radiology, Istanbul, Turkey

⁴Vezirköprü State Hospital, Clinic of Radiology, Samsun, Turkey

ABSTRACT

Gallbladder perforation is a rare and serious complication of acute cholecystitis that requires prompt diagnosis and treatment. Current imaging techniques, such as the use of iodinated contrast agents, have limitations in certain patient populations. Carbon dioxide (CO₂) contrast media has emerged as a potential alternative due to its non-nephrotoxic and non-allergenic properties. However, there is limited research on the use of CO₂ cholangiography for assessing gallbladder perforation. In this case presentation, we describe the use of CO₂ cholangiography in a 72-year-old woman with suspected acute cholecystitis and gallbladder perforation. The patient underwent percutaneous cholecystostomy because of her unsuitable general condition for surgery. CO₂ cholangiography revealed loss of biliary tract integrity and obstruction of the cystic canal. CO₂ cholangiography offers the advantages of decreased cholangitis risk and improved patient comfort. Although this case demonstrates the feasibility of CO₂ cholangiography for gallbladder perforation assessment, further studies are needed to validate the safety and efficacy of this technique. Overall, CO₂ contrast media may provide high-quality imaging with a lower risk of complications, highlighting the potential benefits of its use in percutaneous interventions for biliary leakage evaluation.

Keywords: Carbon dioxide (CO₂), cholangiography, gallbladder perforation

INTRODUCTION

Gallbladder perforation is a relatively rare complication that occurs most frequently as a result of acute cholecystitis, with a relatively high mortality [1]. In patients with suspected acute cholecystitis, imaging is important for two main reasons; first, to confirm the diagnosis of acute cholecystitis, the latter is to detect complications that require urgent surgery such as gangrenous, emphysematous cholecystitis, and perforation [2]. Iodinated contrast agents are accepted as the gold standard for percutaneous and angiographic imaging worldwide [3]. A better alternative is carbon dioxide (CO₂) contrast, which is used for vascular interventions in patients with renal impairment or iodine hypersensitivity due to non-nephrotoxic and non-allergenic characteristics [4]. There are few studies in the literature on using CO₂ contrast media in biliary imaging. There is no study published about gallbladder perforation assessment using CO₂ cholangiogram.

CASE PRESENTATION

A 72 years old woman presented to our hospital emergency department with acute onset fever, severe abdominal pain, nausea, and vomiting. The patient was referred to the radiology department for imaging purposes with prediagnosis of acute cholecystitis, according to the findings of the physical examination and laboratory findings. Ultrasonographic examination revealed that the gallbladder wall of the patient was markedly edematous and thickened, the sac was hydroptic, and the luminal sludge was full of gallbladder calculi were also monitored at neck level approximately 2 cm in diameter. It has also been suspected to be a sustained integrity of the gallbladder wall. Heterogeneous hypoechoic solid lesions were observed in the liver parenchyma with a multiplicity of which the greatest is about 6 cm in diameter and primarily interpreted in favor of metastasis. In subsequent examinations, cholangiocarcinoma was detected as the primary pathology.



Address for Correspondence: Asst. Prof. İsmail Caymaz, Istanbul Medeniyet University Faculty of Medicine, Göztepe Training and Research Hospital, Department of Interventional Radiology, Istanbul, Turkey

Phone: +90 216 280 33 33 E-mail: dr.ismailcaymaz@gmail.com ORCID ID: 0000-0002-4426-9672

Received: 17.07.2023 Accepted: 02.08.2023



©Copyright 2023 by the Azerbaijan Gastroenterology Invasive Endoscopy Society / Caucasian Medical Journal published by Galenos Publishing House. This journal is licensed under a Creative Commons Attribution-NonCommercial 3.0 International (CC BY-NC 3.0).

Percutaneous cholecystostomy was performed with sonographic guidance in our interventional radiology unit owing to the fact that the general condition of the patient is not suitable for surgery due to a history of dementia and heart failure. For this patient, CO₂ cholangiography was chosen as a control imaging technique because of the low risk of cholangitis [5]. CO₂ cholangiography showed that the integrity of the biliary tract was lost and the cystic canal was obstructed (Figure 1).

DISCUSSION

Perforation of the gallbladder, which was first reported in 1844 by Duncan, is a rare serious complication of cholecystitis [5]. Clinical manifestations of acute gallbladder perforation can be similar to those of acute cholecystitis, and this could account for the delay in the diagnosis [6].

Rates of gallbladder perforation have fallen because of widespread imaging methods and better treatment approaches. Nonetheless, accurate diagnosis is crucial to decrease rates of mortality and complications such as peritonitis [7]. For this purpose, CO₂ cholangiography can be used as a safe imaging technique because of the decreasing rate of cholangitis.

Initially, CO₂ was used for retroperitoneal insufflation [8]. More recently, it has been safely injected intraarterial digital subtraction angiography [9]. Recently, CO₂ retrograde cholangiography reduces the risk of cholangitis and therefore can be used for percutaneous imaging of gallbladder perforation [10]. CO₂ is being used as a contrast material for a long time in endoscopic retrograde cholangiopancreatography (ERCP) imaging of the biliary tree and is increasingly preferred due to its

safety, less painful, and discomfort complaints [11]. In addition, the utilization of CO₂ in cholangiography significantly reduced the incidence of cholangitis and the mean hospital stay time after ERCP [10]. Even though there was not enough experience in anterograde cholangiography with CO₂, perforation was clearly observed in this study. The accuracy of the method should have been investigated with a greater number of patients. The use of CO₂ during percutaneous interventions in the evaluation of biliary leakage can be superior to other methods to provide higher quality images with lower risk of cholangitis and allergic reaction and higher postoperative comfort of the patient. Further studies are needed to advance this method and elaborate the safety and validity of the technique.

Ethics

Informed Consent: Informed consent was obtained from the patient.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: İ.C., Concept: İ.C., Design: İ.C., Data Collection or Processing: İ.C., M.B.D., A.B.Y., Analysis or Interpretation: İ.C., Literature Search: İ.C., Writing: İ.C., A.M., M.B.D., A.B.Y.

Conflict of Interest: No conflict of interest was declared by the authors.

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

References

1. Gunasekaran G, Naik D, Gupta A, Bhandari V, Kuppusamy M, et al. Gallbladder perforation: a single center experience of 32 cases. *Korean J Hepatobiliary Pancreat Surg*. 2015;6:10.
2. Smith EA, Dillman JR, Elsayes KM, Menias CO, Bude RO. Cross-sectional imaging of acute and chronic gallbladder inflammatory disease. *AJR Am J Roentgenol*. 2009;192:188-96.
3. Mendes Cde A, Martins Ade A, Teivelis MP, Kuzniec S, Varella AY, et al. Carbon dioxide contrast medium for endovascular treatment of ilio-femoral occlusive disease. *Clinics (Sao Paulo)*. 2015;70:675-9.
4. Hawkins IF, Cho KJ, Caridi JG. Carbon dioxide in angiography to reduce the risk of contrast-induced nephropathy. *Radiol Clin North Am*. 2009;47:813-25, v-vi.
5. Anderson BB, Nazem A. Perforations of the gallbladder and cholecystobiliary fistulae: a review of management and a new classification. *J Natl Med Assoc*. 1987;79:393-9.
6. Isch JH, Finneran JC, Nahrwold DL. Perforation of the gallbladder. *Am J Gastroenterol*. 1971;55:451-8.
7. Malik HS, Cheema HA, Fayyaz Z, Hashmi MA, Parkash A, et al. Spontaneous perforation of bile duct, clinical presentation, laboratory work up, treatment and outcome. *J Ayub Med Coll Abbottabad*. 2016;28:518-22.
8. Burko H, Klatte EC. Renewed interest in gases for contrast roentgenography. *Am J Roentgenol Radium Ther Nucl Med*. 1967;99:645-59.
9. Hawkins IF. Carbon dioxide digital subtraction arteriography. *AJR Am J Roentgenol*. 1982;139:19-24.

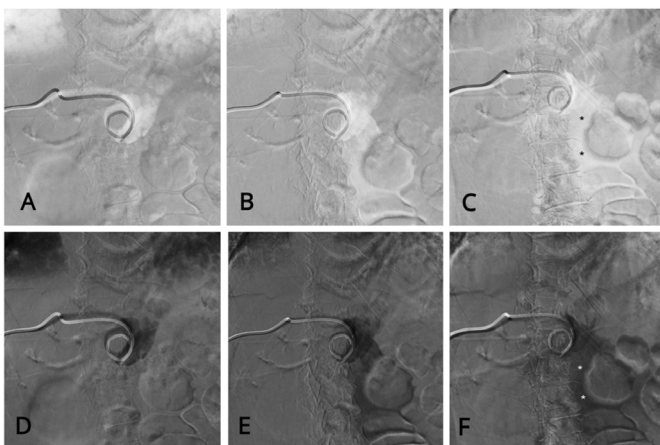


Figure 1. A-C images were obtained with injection of CO₂ contrast material. D-F are negative images of these images. As indicated by the stars (*) in the C and F images, the contrast material in the carbon dioxide cholangiography is more expanse

CO₂: Carbon dioxide

10. Zhang R, Zhao L, Liu Z, Wang B, Hui N, et al. Effect of CO₂ cholangiography on post-ERCP cholangitis in patients with unresectable malignant hilar obstruction - a prospective, randomized controlled study. *Scand J Gastroenterol.* 2013;48:758-63.
11. Cheng Y, Xiong XZ, Wu SJ, Lu J, Lin YX, et al. Carbon dioxide insufflation for endoscopic retrograde cholangiopancreatography: a meta-analysis and systematic review. *World J Gastroenterol.* 2012;18:5622-31.