ISSN NO: 2770-2936
Date of Publication:18-12-2022

Uncommon Presentation of Second Branchial Cyst Compression on the Carotid Bifurcation: A Case Report

Enas Abdulmaged [1]*, Hanan Amanuel Qas Marrogy [2]
[1, 2] Specialist Oral and maxillofacial Surgeon, Department of Oral and Maxillofacial Surgery, Al-Wasity Teaching Hospital, Muthanna Al Shaibani Street, Baghdad, 10089, Baghdad, Iraq
Emails: drenas999@yahoo.com [1], Hanan_emmanuel95@yahoo.com [2]
* *Corresponding author*

Abstract: The aim of this case report is to disclose atypical manifestations of non-epileptic seizures caused by cerebral ischemia as a result of a progressively large infected second branchial cyst compressing over the carotid bifurcation for the past three years. A female patient who presented to our clinic with symptoms of a constantly growing neck mass, pain, headache, difficulty turning the head, convulsions, teeth clenching, and syncope as a result of delayed appropriate treatment. After taking a thorough history of her signs and symptoms, along with laboratory tests and an electroencephalogram (EEG), we come to the definite diagnosis of the reason for her seizure-like symptoms, which were caused by the pressure exerted by the enlarged, infected second branchial cyst.

Key words: Second branchial cyst, Non-epileptic seizures, Iraq

Highlights

- Second branchial cysts remain asymptomatic in the absence of infection. However, if secondary infection occurs, the cystic cavity may become an infection-hosting space.
- This expansion of the cyst wall causes compression and irritation to the vital structures surrounding the cyst, such as the carotid arteries.
- Non-epileptic seizures can be caused by persistent compression or irritation of the carotid artery that leads to cerebral ischemia.
- Approximately four months of postoperative follow-up showed improvement in signs and symptoms.

Introduction

Branchial cleft cysts, which are congenital anomalies, may be inherited as an autosomal dominant trait. originating at any level of the mandible (first branchial cleft) to the supraclavicular region (fourth branchial cleft). Depending on the degree of incomplete obliteration of the cervical sinus or the epithelial rests of the branchial clefts during the fourth week of embryogenesis, they appear at any age with equal frequency in males and females. They can be found at any depth between the skin and the pharynx. Second branchial cleft cysts are the most common benign branchial cleft cysts, representing approximately 40–95% of branchial anomalies (1, 2). They commonly occur along the anteromedial border of the sternocleidomastoid muscle (3), posterolateral to the submandibular gland, and anterolateral to the carotid sheath (4). They typically present clinically as painless, mobile, and fluctuant neck cysts, and they frequently follow an upper respiratory tract infection.Secondary infection and inflammation can occur, leading to a sudden, slow increase in the size of the cyst with symptoms of pain, swelling, tenderness, and fever (5). An infected branchial cleft cyst will demonstrate peripheral rim enhancement, which occurs when there is no external opening and fluid cannot drain out of the neck. Then, a collection of fluid forms (6). This exerts pressure on the vital structures present around the cyst. Therefore, some patients may experience symptoms of ipsilateral carotid artery compression secondary to the enlargement of the cyst. In rare instances, non-epileptic seizures can be caused by persistent compression or irritation of the carotid artery that leads to cerebral ischemia. Non-epileptic seizures are events that resemble epileptic attacks but lack their clinical and electrographic features (7).

In this case, we report a second branchial cleft cyst in a 36-year-old female patient with a history of non-epileptic seizures caused by the enlargement and compression of the cyst on the carotid bifurcation.

ISSN NO: 2770-2936
Date of Publication:18-12-2022

Presentation of Case

A 36-year-old female patient presented to our clinic in the Oral and Maxillofacial Surgery department at our institute a year prior, complaining of a recurrent, progressive, painful swelling in the left side of her neck (Fig. 1). The swelling was initially noticed as a small, asymptomatic mass at 14 years of age, remained constant over the years, and an enlargement was noticed 3 years ago with associated symptoms of pain, headache, difficulty turning the head, convulsions, clenching of teeth, and syncope. Before presenting at our hospital, the patient reported multiple previous attempts done at an outside clinic to drain the swelling with a systemic antibiotic prescription, but no improvement of signs and symptoms occurred. She was referred to a neurological department for assessment of her seizures, and anti-epileptic drugs were prescribed to her to alleviate her symptoms. A cardiovascular consultation was also obtained to determine the relationshipbetween the enlarged cyst and the external and internal carotid arteries.

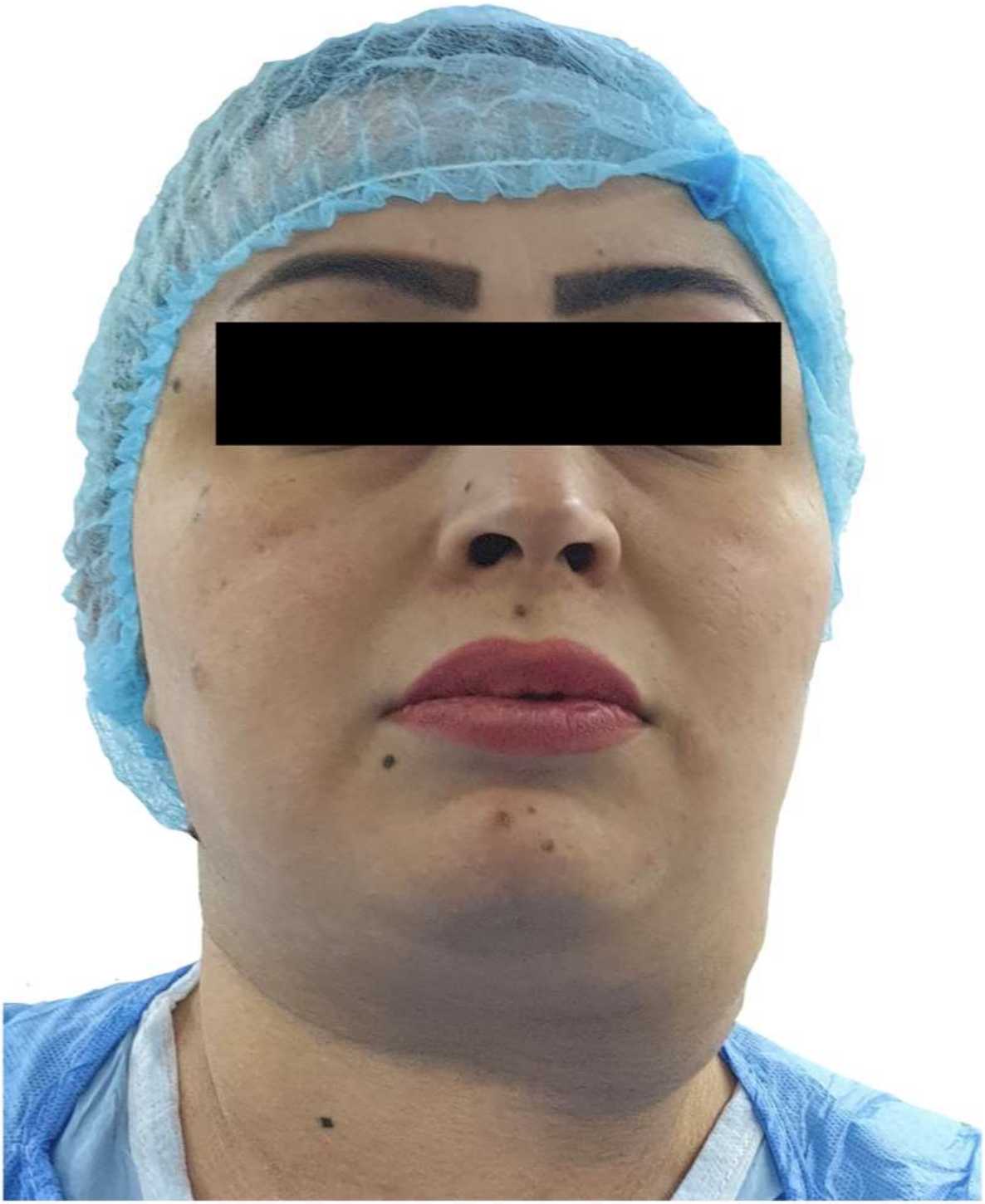

Fig. 1. Left-side neck swelling This image is published with the patient's consent.

ISSN NO: 2770-2936
Date of Publication:18-12-2022

Upon physical examination, the mass extended from the lower left border of the mandible to half the length of the neck; the skin over the mass was normal in color with no ulceration or sinus opening. Pain and fluctuation were felt during palpation, but no intraoral findings were found. Preoperative conventional laboratory investigations were normal, except an elevation in WBC level was noticed. A FNA was performed, and the cellular smear of the turbid fluid revealed a mixed inflammatory cellular infiltrate consisting of mature lymphocytes, plasma cells, polymorphonuclear neutrophils, and foamy macrophages, as well as a large number of squamous epithelial cells. The cytological picture suggested a branchial cleft cyst with chronic suppurative inflammation. One of the FNA blood samples accidentally revealed tuberculous lymphadenitis; thus, the IGRA test (interferon gamma release assay) and polymerase chain reaction (PCR) were both performed and resulted in positive tuberculosis.
In addition, cellular smears indicated heavy chronic inflammatory cells, necrotic debris, and a large number of foamy macrophages. The overall picture consists of tuberculous abscess collections (cold abscess).
The computed tomography (CT) scan demonstrated a well-defined hypodense mass with a thickened cell wall extending from the lower left border of the mandible at the C3 level to the C5 level of the neck, anteromedial to the sternocleidomastoid muscle, and lateral to the pharynx and larynx, pushing the vessels medially. The largest anteroposterior, mediolateral, and superoinferior dimensions were 38 mm, 45 mm, and 50 mm, respectively.
The electroencephalogram (EEG) showed normal results, ruling out any changes in the electrical activity of the brain that are indicative of epileptic seizures. (Fig. 2)

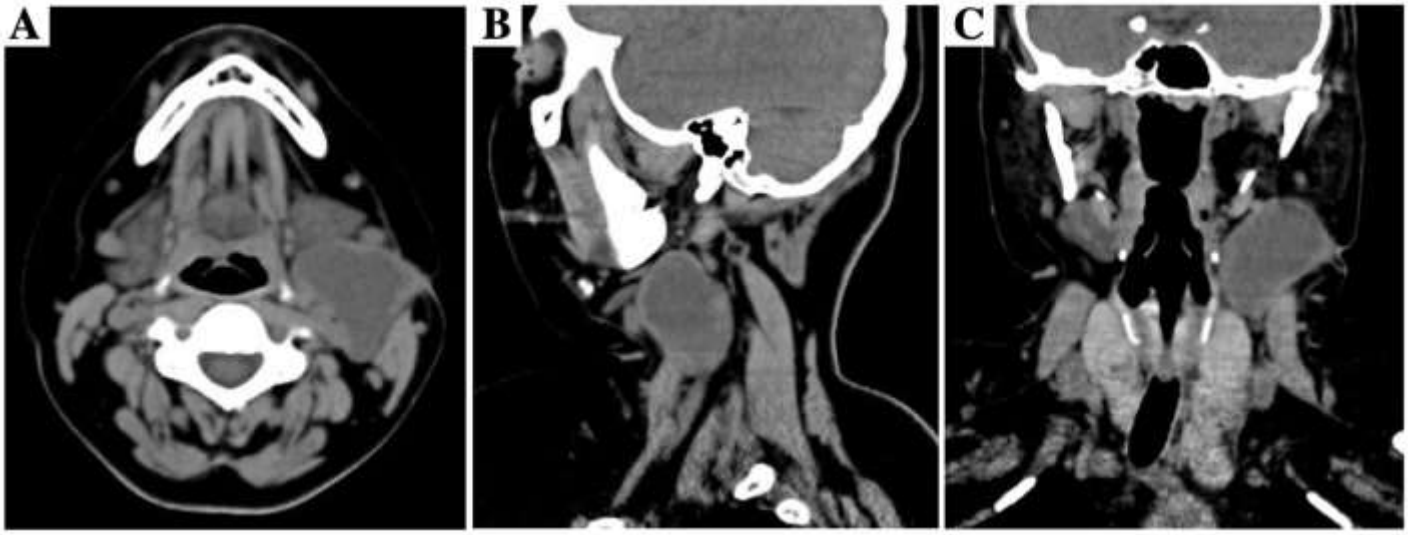

Fig. 2. (A) axial CT view demonstrates a well-defined hypodense mass compressing the vital structures medially. (B) sagittal view of the CT demonstrates the superioinferior enlargement of the cyst. (C) coronal view of the CT demonstrates the depth of extension of the cyst.

Before preparing the patient for surgery, anti-tubercular therapy was started at a specialized facility for respiratory diseases. And after six months, IGRA findings were negative for tuberculosis.
The patient was operated on under general anesthesia with oral intubation and in the supine position, and surgical access to expose the mass was achieved via a submandibular incision. Dissection was carried out to remove the cyst without damaging the surrounding vital structures. Unfortunately, the cyst ruptured intraoperatively, and the turbid-colored fluid was washed using normal saline. The dissection was not completed until the entire cyst lining was excised and a drain was placed. (Fig. 3)

ISSN NO: 2770-2936
Date of Publication:18-12-2022

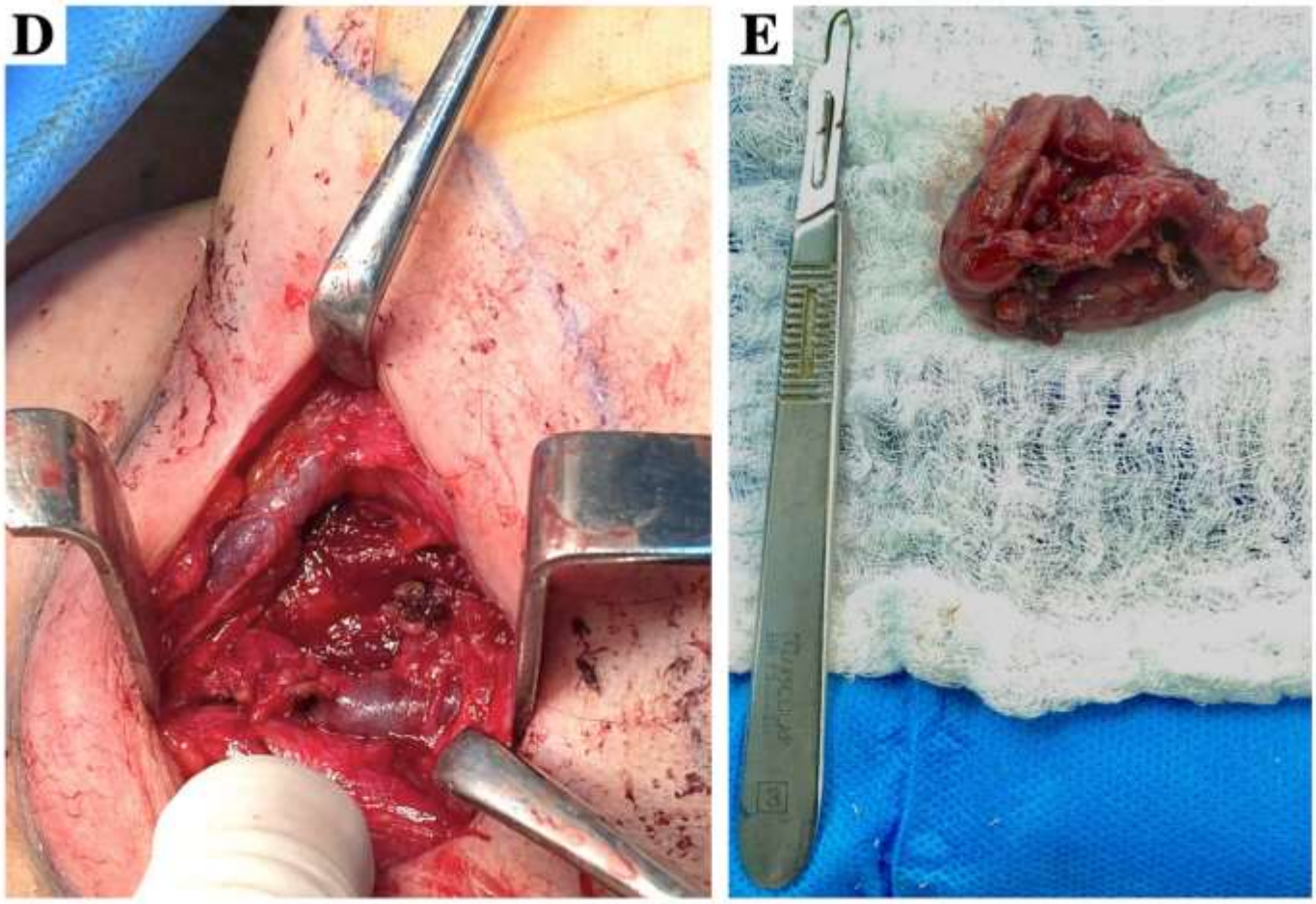

Fig. 3. (D) Intraoperative image after removal of the second branchial cyst. (E) the capsule of the cyst after complete excision

Approximately six months of postoperative follow-up showed improvement in her signs and symptoms.

Discussion

Second branchial cleft cysts represent the most frequent type of lateral neck masses; they are congenitally benign anomalies that appear in childhood and develop in adults as a mass or infection in the neck. Histologically, they are lined by squamous epithelium and contain inflammatory lymphocytes (1). Radiographically, they present as well-circumscribed, fluid-like masses with uniformly thin walls; some extend medially from the bifurcation of the internal and external carotid arteries to the lateral pharyngeal wall and anteromedial to the sternocleidomastoid muscle, as presented in our case (8). Frequently, they remain asymptomatic in the absence of infection. However, if secondary infection arises, the cystic cavity can form a potential space that hosts infection; hence, episodic swelling can occur, causing a rapid increase in the size of the cyst along with a marked thickening of the cyst wall following repeated infection (9, 10). The cyst wall's expansion compresses and irritates the vital structures surrounding the cyst, such as the carotid arteries in this case. Any compression on the carotid arteries could result in cerebral ischemia due to disruption of blood supply from the enlarged, infected cyst over the carotid bifurcation. The chronic interruption of blood supply to the brain leads to damage to part of the brain and the consequent appearance of seizure-like symptoms (11).

The diagnosis of non-epileptic seizures was made in consultation with neurology, and it is based on obtaining a thorough history and conducting radiological and electrographic investigations. Clinically, they may appear to be generalized convulsions, similar to grand mal epileptic seizures. However, unlike epileptic seizures, non-epileptic seizures are not caused by electrical disruptions in the brain, and the diagnosis is confirmed by an EEG, which records the electrical activity of the brain. Treatment of the underlying cause can often relieve the symptoms (12).

ISSN NO: 2770-2936
Date of Publication:18-12-2022

Fortunately, the patient sought a second opinion when her symptoms persisted and the size of the cyst continued to grow. In this case, a non-epileptic seizure is conclusively diagnosed based on the improvement of signs and symptoms following complete excision of the cyst.

Because this condition is rare, the exact etiology of the secondary infection of the second branchial cyst is unknown. Nevertheless, it is possible that the untreated tuberculous lymphadenitis caused the secondary infection.

Conclusion

Due to the persistence of secondary infection and the second branchial cyst's unique location and extension of its wall over the carotid bifurcation, the infected cyst continued to encroach on the carotid arteries, applying constant pressure and irritation that caused disruption of the blood supply and, thus, a subsequent decrease in oxygen to the brain. This persistent effect over a period of three years led to the occurrence of non-epileptic seizures. The fact that surgical excision is the curative option and the improvement of her signs and symptoms imply that the seizure-like symptoms she had been experiencing were from the pressure exerted by the enlarged, infected cyst.

References

1. Coste, Anouchka H. "Branchial Cleft Cyst – Stat Pearls - NCBI Bookshelf." National Center forBiotechnology Information, National Library of Medicine, 14 Apr. 2022.
2. Fonseca, et al. Fonseca Oral and Maxillofacial Surgery 3rd Edition Volume 2.
3. Shahrokh C. Bagheri, et al. Neck Mass: Diagnosis and Management - ScienceDirect. Elsevier, 2012.
4. Current Therapy In Oral and Maxillofacial Surgery | ScienceDirect." ScienceDirect.Com | Science,Health and Medical Journals, Full Text Articles and Books., Elsevier, 2012.
5. Susan Muller, et al."Second Branchial Cleft Cyst - PMC.'" PubMed Central (PMC), NationalLibrary of Medicine, 2015.
6. Jorge A Soto, and Brian Lucey. "Emergency Radiology Science Direct." ScienceDirect.Com Science,Health and Medical Journals, Full Text Articles and Books., Elsevier, 2009.
7. Imitators of epilepsy: the physiologic nonepileptic seizures." J. epilepsy clin. neurophysiol. 2007.
8. Gökçe, Erkan, and Murat Beyhan. "CT and MRI Findings of Branchial Cleft Cysts. EasternJournal Of Medicine, no. 1, LookUsBilisim A.S., 2021, pp. 47-52. Crossref.
9. Su Hu, Chun-Hong Hu, et al. "Atypical Imaging Observations of Branchial Cleft Cysts.Spandidos Publications, Spandidos Publications, 2013.
10. Marotta, Dario A., et al. "Branchial Cleft Cyst Harbors Metastatic Papillary Thyroid Carcinoma. Cureus, Cures, Inc., Mar. 2021.
11. David, Joshua, et al. "Stylocarotid Artery Syndrome." Journal of Vascular Surgery, no. 6, ElsevierBV, Dec. 2014.
12. LaFrance, W. Curt, et al. "Minimum Requirements for the Diagnosis of Psychogenic Nonepileptic Seizures: A Staged Approach." Epilepsia, no. 11, Wiley, Sept. 2013.