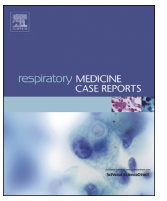
Respiratory Medicine Case Reports 39 (2022) 101730

Contents lists available at ScienceDirect

Respiratory Medicine Case Reports

journal homepage: www.elsevier.com/locate/rmcr



Case Report

A case of malignant stroke requiring hemicraniectomy following bronchial artery embolisation

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ARTICLE INFO

Keywords:
Bronchial artery embolisation
Malignant stroke
Hemicraniectomy
Post-stroke recovery

1. Introduction

Large volume haemoptysis is a recognised complication of cystic fibrosis (CF) lung disease and usually occurs due to bronchial artery hypertrophy. Haemoptysis in such instances is often treated with bronchial artery embolisation (BAE), which involves either coiling or administrating embolic agents to targeted vessels via an arterial microcatheter. Stroke is a recognised rare complication of the procedure [1] and its risk may be higher in CF patients as a result of vascular anastomoses that form due to chronic pulmonary inflammation [2]. Malignant strokes are associated with cerebral and/or cerebellar oedema, may lead to significant rises in intracranial pressure and result in cerebral and/or brain stem herniation. The patient in this report suffered a malignant stroke following BAE and required emergency neurosurgical decompression. To our knowledge no other reported cases of malignant stroke post-BAE have been reported. This case report highlights the need for all clinicians to recognise the possibility of malignant stroke following BAE. Alongside other research this report also suggests that the risk of embolic events post-BAE may be higher in CF patients.

2. Case report

A 41-year-old male with CF (*Phe508del/Gly551Asp)* presented acutely to a tertiary adult Cystic Fibrosis (CF) centre with a 5-day history of large volume haemoptysis, shortness of breath and chest pain. He had a prior history of significant haemoptysis treated via bronchial artery embolisation (BAE) in 2012. His medical history also included multiple superior vena cava (SVC) stenting procedures for catheter induced SVC stenosis, *Pseudomonas aeruginosa* colonisation, pancreatic insufficiency and CF-related diabetes. The patient's clinical trajectory substantially improved following the initiation of ivacaftor therapy in 2013, with a marked reduction in admission frequency but he continued to have severe lung disease with a baseline FEV_1 of 38% predicted.

At presentation his vital signs were normal and a chest radiograph showed changes consistent with severe CF lung disease with no acute changes. The patient continued to expectorate over 100mL of blood daily despite intravenous antibiotics, vitamin K and tranexamic acid. An arterial phase CT-thorax with contrast showed a markedly enlarged main bronchial artery with numerous enlarged, tor-

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https://doi.org/10.1016/j.rmcr.2022.101730
Received 26 May 2022; Received in revised form 5 August 2022; Accepted 15 August 2022
Available online 19 August 2022

tuous mediastinal bronchial arteries, a large abnormal blood supply arising from the right subclavian artery and that his SVC stents were patent (Fig. 1a and b). The scan also highlighted the patient's extensive lung disease secondary to his CF (Fig. 2).

Fig. 1A displays a coronal thick Maximum Intensity Projection (MIP) image from the patient's initial CT-thorax. The most superior arrow highlights a significantly hypertrophied bronchial artery as it descends and enters the mediastinum. The other two arrows demonstrate that the artery continued to be grossly enlarged after it descended and deviated laterally to supply the left lung parenchyma. Fig. 1B also displays a thick MIP image from the patient's initial CT-thorax and highlights a separate grossly enlarged artery originating from the right subclavian artery. This vessel is shown to descend into the mediastinum where it then supplies the right lung.

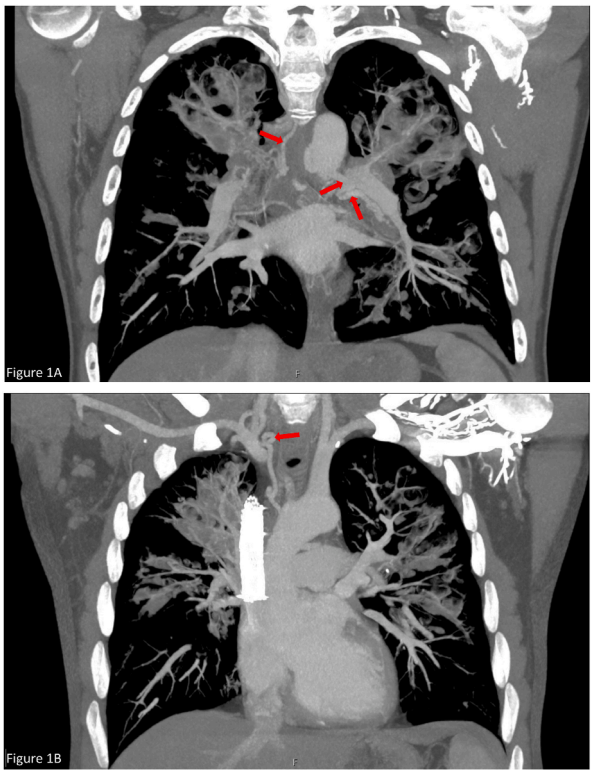

Fig. 1. CT-thorax demonstrating enlarged and tortuous arteries.

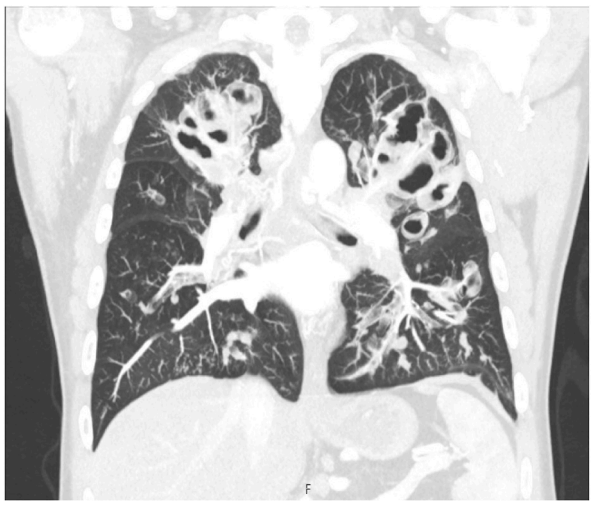

Fig. 2. CT-thorax highlighting the lung windows.

Fig. 2 is an image taken from the patient's initial CT-thorax highlighting the lung windows. The image shows extensive, thick-walled cystic changes throughout both lungs and widespread enlargement in bronchial wall thickness consistent with severe CF lung disease. The pleural spaces and central airways are clear.

The patient displayed intractable haemoptysis despite medical management and a decision to proceed to BAE was made. Full informed consent for the procedure was provided by the patient following discussions regarding its risks and benefits. The affected bronchial artery was super selectively cannulated under sedation, a microcatheter was advanced into each branch and embolisation was achieved using 350–500 μm and 500–700 μm of polyvinyl alcohol (PVA). Two further accessory bronchial arteries were identified and embolised. Due to the length of the procedure and volume of contrast (300mLs) used, angiography of the vasculature arising from the right subclavian artery was deferred. A further procedure three days later identified two additional tortuous vessels, which were selectively catheterised with angiographic confirmation of position and embolised. The only procedure related complication noted was a headache presumed to be secondary to glyceryl trinitrate (GTN) use during the procedure, which responded to simple analgesia.

Contrast was used during both procedures to identify collateral vessels connecting the bronchial arteries targeted for embolisation to systems supplying other parts of the lung and elsewhere. The interventional radiology team was able to identify a connection between the bronchial circulation and a vessel that most likely originated from the right subclavian artery and supplies the apex of the right lung (Fig. 3).

Fig. 3 shows the microcatheter inserted within the main bronchial trunk and contrast being injected into the bronchial artery system. Contrast is also shown within a more superior vessel close to the apex of the lungs that runs laterally and likely arises from the right subclavian artery. Smaller collateral vessels can be seen connecting these two systems. This image therefore shows that collateral vessels were present joining the bronchial circulation to this separate vascular system. No connections between the bronchial circulation and vessels supplying the cerebrum or cerebellum were identified during angiography.

Within 24 h of the second procedure, the patient developed a new severe occipital headache associated with flashing lights, photophobia, nausea and vomiting. There was no disturbance of consciousness or focal neurological signs. An urgent CT head (Fig. 4A) demonstrated a large infarct measuring 32mm × 32mm x 22mm in the right cerebellar hemisphere as well as three smaller but nevertheless extensive infarcts in the left cerebellar hemisphere. The CT also demonstrated at least 3 distinct infarcts within the occipital lobes, the largest of which was 17mm × 10mm x 7mm, and one much smaller infarct in the right caudate nucleus. The scan showed no evidence of midline shift or hydrocephalus. Given these findings, the patient's acute symptoms and his recent history, a diagnosis of embolic cerebrovascular accident (CVA) secondary to the BAE procedure was made.

The following day the patient deteriorated and demonstrated a reduction in his Glasgow Coma Scale (GCS = 12: Eyes 4, Voice 4, Motor 2). A repeat CT-head (Fig. 4B) showed progressive dilatation of the supratentorial ventricular systems associated with trapped temporal horns. The cerebellar, occipital and right caudate nucleus infarcts seen previously showed no significant changes. The patient was therefore diagnosed with obstructive hydrocephalus and urgently transferred to the regional neurosurgical unit where a burr hole procedure was performed to achieve cranial decompression. A post-operative CT however showed that the degree of hydrocephalus showed little to no improvement and that tonsillar herniation had developed. An emergency hemicraniectomy was therefore

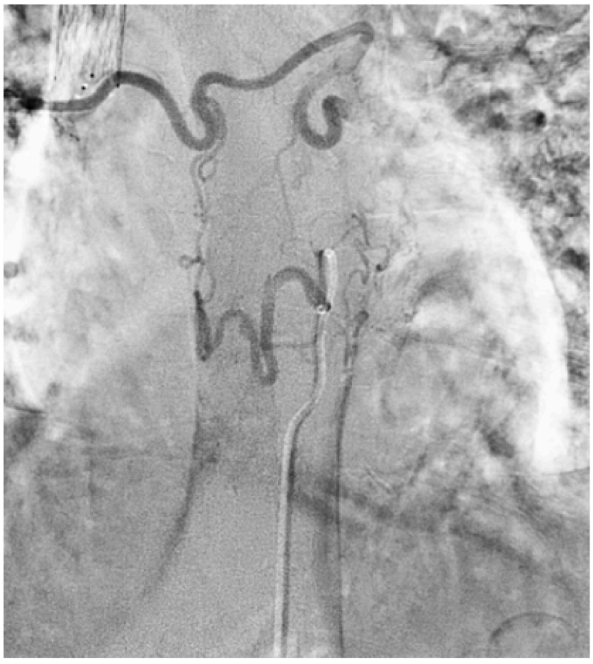

Fig. 3. Collateral vessels connecting the bronchial and right subclavian artery networks.

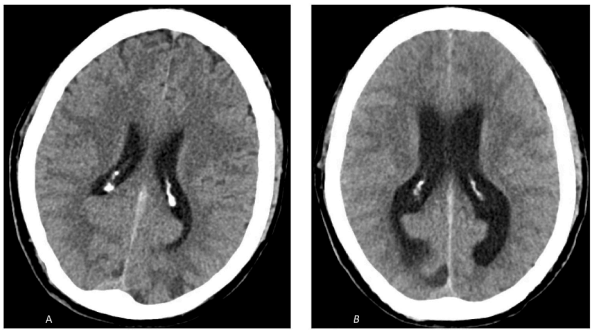

Fig. 4. Ct head imaging showing obstructive hydrocephalus.

performed and successfully achieved cranial decompression. The patient's post-operative recovery was prolonged and complicated by hallucinations, severe nausea and vomiting and disabling vertigo. A continuous subcutaneous infusion of anti-emetics facilitated discharge to home and a full physical recovery was achieved over a period of 6 months. A follow up MR brain scan 5–6 months after the stroke showed evolution of the radiological abnormalities in keeping with expected recovery, with encephalomalacia and minimal herniation of the cerebellar hemispheres through the craniectomy defect.

Fig. 4A displays an image of the lateral ventricles and surrounding cerebral cortex and subcortical areas taken from the patient's initial CT head. Fig. 4B displays a CT image from the same region of the brain 24 hours later following a decline in his Glasgow Come Scale. Fig. 4A demonstrates no evidence of disruption of the supratentorial ventricular system whereas Fig. 4B shows that the lateral ventricles became significantly more dilated, especially within the temporal horns. The trapping of CSF within the temporal horns seen in Fig. 4B suggests that the patient developed obstructive hydrocephalus, most likely as a complication of the multiple infarcts in the cerebellum, occipital lobes and right caudate nucleus.

3. Discussion

We describe a case of malignant stroke following BAE that required neurosurgical decompression via a burr hole procedure and a subsequent emergency hemicraniectomy. Although embolic CVA is a well described potential complication of this procedure, there are aspects of this case which make it unusual. Both the number of infarcts and the patient's younger age may have contributed to the significant cerebral oedema seen in this case. It is also noteworthy that despite the degree of abnormality identified on initial brain imaging, the patient had minimal and easily controllable symptoms during and immediately after the procedure.

Hemicraniectomy is a surgical procedure where a large section of the skull is removed and the dura is opened. The procedure therefore provides space for the swollen brain to expand and reduces the intracranial pressure. CVA with associated cerebral oedema requiring hemicraniectomy is relatively uncommon, particularly in the absence of the occlusion of a large intracranial artery. A retrospective study that examined 2227 ischaemic strokes in adults showed that just 0.3% of cases would have qualified for hemicraniectomy as per the criteria used in recent clinical trials [3]. Given the significant survival and functional outcome benefits in treated patients, more studies are needed to determine whether additional subgroups of patients with ischemic stroke may benefit from hemicraniectomy. To our knowledge, no other cases where patients have required neurosurgical decompression have been published. This may be a result of under reporting of similar episodes, a lack of rapid access to neuroimaging and/or an inability to rapidly transfer patients to neurosurgical centres.

Previous studies suggest that symptomatic post-BAE strokes are rare. A systematic review by Panda et al. showed that the risk of stroke, TIA and cortical blindness following BAE was 0.6–2.2% [1]. Additionally, Frood et al. reported just 1 out of 68 of patients suffered a stroke over a 2-year period following BAE, and this patient made a complete recovery within 1 month [4]. Headache is well-recognised but often overlooked symptom of stroke and is more common in patients with posterior circulation stroke syndromes [5]. Without prompt recognition of his acutely deteriorating symptoms by the clinical team this patient may have been diagnosed at a much later stage or misdiagnosed completely, especially as his initial headache was relatively mild. Knowledge of this rare complication post-BAE was therefore essential in managing this patient effectively.

It has been suggested that post-BAE strokes occur due to the passage of emboli via anastomoses between bronchial arteries and pulmonary veins or between bronchial and vertebral arteries [6]. Other possible mechanisms include the reflux of embolic agents used in BAE from targeted bronchial arteries into the systemic circulation [7]. Although just 0.02% of the population have anastomoses between pulmonary arteries and veins, such malformations are more likely to arise in pulmonary inflammation [2]. CF lung disease is associated with an inflammatory state, bronchial artery hypertrophy and collateral vessel formation. People with CF may therefore be at greater risk of CVA following BAE. In this case no abnormal anastomoses between the bronchial circulation and vessels supplying the brain were found, including during a retrospective review of the images at a multidisciplinary team meeting. It is possible however, that connections between these systems existed but were below the imaging resolution. Although rare events such as this should not prevent clinicians from recommending BAE to treat haemoptysis, this case suggests that knowledge of the risk of embolic stroke and how it may present is essential to ensure rapid diagnosis and treatment.

4. Conclusion

- Knowledge of the risk of CVA post BAE enabled prompt diagnosis and management.
- Headache during and/or following BAE should lead clinicians to suspect posterior circulation stroke as a possible complication and consider urgent neurological assessment and investigations.
- Given that CF patients may be at higher risk of CVA post BAE due to arterial and arteriovenous anastomoses, caution is necessary when considering the procedure in this cohort.

Consent statement

Full informed patient consent was acquired from the patient prior to the submission of this case report.

Declaration of competing interest

There have been no competing interests during the writing and submission of this manuscript.

References

[1] A. Panda, A.S. Bhalla, A. Goyal, Bronchial artery embolization in hemoptysis: a systematic review, Diagn Interv Radiol 23 (4) (2017) 307–317, https://doi.org/10.5152/dir.2017.16454.

[2] M. Hoffman, J.P. Gerding, J.B. Zuckerman, Stroke and myocardial infarction following bronchial artery embolization in a cystic fibrosis patient, J. Cyst. Fibros. 16 (1) (2017 Jan) 161–162 https://doi.org/10.1016/j.jcf.2016.11.007, Epub 2016 Dec 6. PMID: 27939048.

[3] R. Rahme, R. Curry, D. Kleindorfer, J.C. Khoury, A.J. Ringer, B.M. Kissela, K. Alwell, C.J. Moomaw, M.L. Flaherty, P. Khatri, D. Woo, S. Ferioli, J. Broderick, O. Adeoye, How often are patients with ischemic stroke eligible for decompressive hemicraniectomy? Stroke 43 (2) (2012 Feb) 550–552 https://doi.org/10.1161/STROKEAHA.111.635185, Epub 2011 Oct 27. PMID: 22034001; PMCID: PMC3265663.

[4] R. Frood, S. Karthik, S. Mirsadraee, I. Clifton, K. Flood, S.J. McPherson, Bronchial artery embolisation for massive haemoptysis: immediate and long-term outcomes-A retrospective study, Pulm Ther 6 (1) (2020) 107–117, https://doi.org/10.1007/s41030-020-00112-x.

[5] A. Harriott, F. Karakaya, C. Ayata, Headache after ischemic stroke, Neurology 94 (1) (2019) e75–e86.

[6] J.H. Park, D.S. Kim, J.S. Kwon, D.H. Hwang, Posterior circulation stroke after bronchial artery embolization, Neurol. Sci. 33 (4) (2012 Aug) 923–926 https://doi.org/10.1007/s10072-011-0855-2, Epub 2011 Nov 29. PMID: 22124851.

[7] T. Nisar, A rare case of posterior circulation stroke caused by bronchial artery embolization, J. Stroke Cerebrovasc. Dis. 27 (8) (2018 Aug) e153–e155 https://doi.org/10.1016/j.jstrokecerebrovasdis.2018.02.040, Epub 2018 Mar 21. PMID: 29571759.