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CASE REPORT

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Dieulafoy's disease of the bronchus: rare but potentially fatal: a case report and a review of literature

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Abstract

Background Dieulafoy's disease of the bronchus can cause massive and even fatal hemoptysis. Even though it is rare, it should be considered by physicians all over the world. This paper reports a case of bronchial Dieulafoy's disease and summarizes the data of similar cases reported in literature.

Methods We report a case of bronchial Dieulafoy's disease (BDD) in Tunisia. We also present a review of literature related to BDD from 1995 to 2022 using the PubMed, Google Scholar, web of science and Chinese National Knowledge Infrastructure Databases. Clinical characteristics, chest imaging, bronchoscopic and angiographic findings were summarized. Treatment courses were identified as well as patients' outcome.

Results We report the case of a 41-year-old man, so far in good health, presenting with massive hemoptysis. Bronchoscopy showed blood clots and a protruding lesion covered by mucosa with a white pointed cap at the entrance of the right upper lobe. Biopsies were not attempted. Embolization of bronchial artery was first realized and was not successful, with post procedure complications. Surgical intervention stopped the bleeding and pathological examination of the resected specimen confirmed Dieulafoy's disease of the bronchus. Ninety cases of BDD were reported from 1995 to 2022. The main symptom was hemoptysis. Chest imaging findings were not specific. The diagnosis of BDD was mainly based on the bronchoscopy, branchial angiography and pathological findings or surgical specimens. Bronchoscopy findings were mostly nodular or prominent lesions (52.4%). Twenty-eight patients underwent bronchoscopic biopsies, 20 had massive bleeding and 10 died. Bronchial angiography mainly showed tortuous and dilation of bronchial artery, and the lesions were mainly located in the right bronchus. Selective bronchial artery embolization (SBAE) was performed in 32 patients and 39 patients underwent surgery.

Conclusion To our knowledge, this is the first case of bronchial Dieulafoy's disease to be reported in Tunisia and North Africa. When the diagnosis is suspected, bronchoscopic biopsy should be avoided as it might lead to fatal hemorrhage. Selective bronchial artery embolization can stop the bleeding, but surgery can be required.

Keywords Hemoptysis, Dieulafoy's disease, Bronchi, Embolization, Surgery

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Background

Massive hemoptysis is a medical emergency which is still feared by most physicians. It presents several diagnostic and therapeutic challenges. Determining the origin of bleeding and underlying etiology is a cornerstone of the treatment plan. However, it may not be immediately apparent and a thorough investigation must be lead. We present the case of a young patient suffering from massive hemoptysis due to bronchial Dieulafoy's disease, as well as a review of literature of similar cases in order to improve the understanding of the disease.

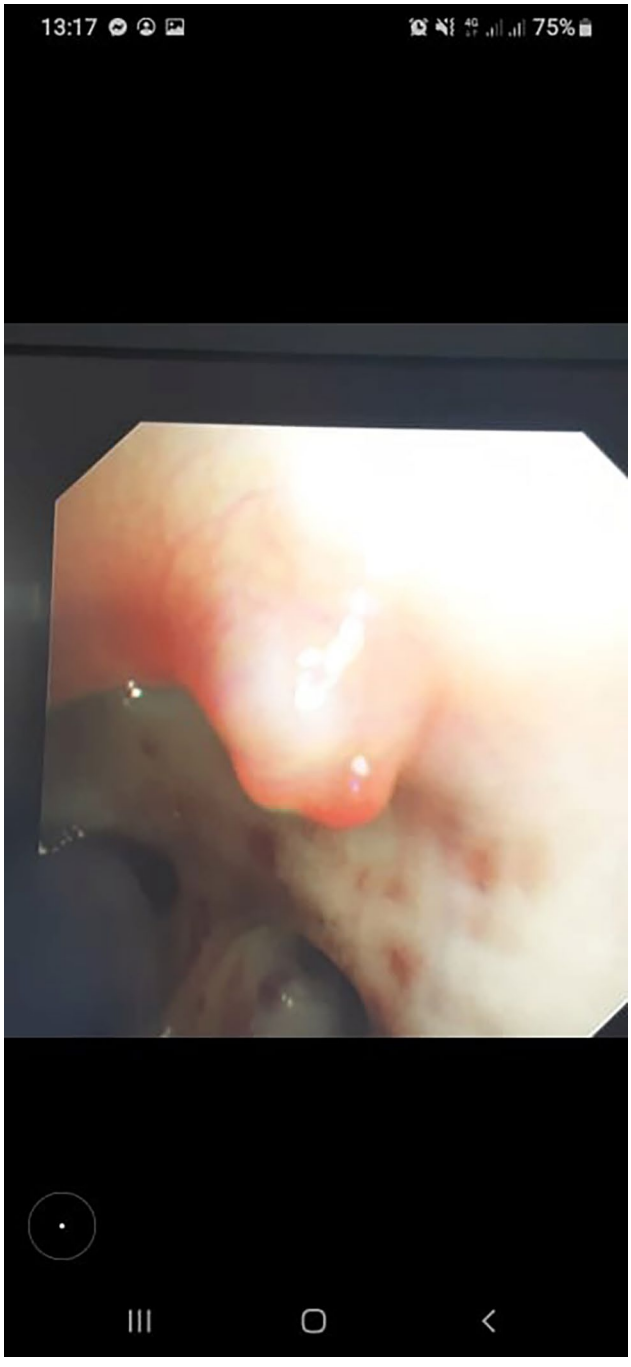

Fig. 1 Nodular protrusion from the mucosa at the entrance of the right upper lobe

Case presentation

A 41-year-old man was admitted in February 2022 to the pneumology department with sudden onset of massive hemoptysis.

He had a less severe episode of hemoptysis one year ago, concomitant to dental extraction, but was otherwise healthy. He worked as a university professor and was an occasional smoker. Initial clinical examination was normal, aside from sinus tachycardia. Blood biochemistry parameters showed a decrease in hemoglobin level from 14 g/dl to 9 g/dl, indicating blood transfusion.

Bronchoscopy showed bleeding stigma in the right main bronchi and a protruding lesion at the entrance to the right upper lobe. The surface was covered by mucosa and had a white pointed cap (Fig. 1). Blood clots were also noted in the right lower lobe bronchus. Computed tomography (CT) scans revealed ground glass opacities at the upper, middle and lower lobes of the right lung (Fig. 2). The right bronchial artery had an ectopic origin from the aortic arche. Further workup with bronchial arteriography revealed no tortuous arteries nor any vascular blush. However, the patient was still coughing up important amounts of fresh blood, approximately 300 ml in an episode, despite prescribing systemic hemostatic treatment.

All of the investigations performed found no evidence of any etiology and vasculitic screen was negative.

Given the findings of the bronchoscopy, we performed an arterial embolization of the right bronchial artery. The left intercostal artery, the left cervico-thoracic artery and the 5th right intercostal artery gave vascular branches to the right hilum and embolization of these arteries was also performed (Fig. 3). However, the procedure failed to prevent the recurrence of his bleeding. The patient developed complications afterwards. He suffered from splenic infarction, bilateral renal infraction and a posterior inferior cerebellar artery stroke, due to spilling of the embolizing agent. The patient also developed acute respiratory failure due to bilateral pulmonary embolism, proximal on the left side. Ultimately, he required hemostasis surgery.

He had a right pneumonectomy on veno-venous extracorporeal membrane oxygenation with simple surgical follow up. Curative heparin was prescribed to treat his pulmonary embolism.

The pathological examination of the resected specimen had found abnormally dilated, sinuous and anastomotic vessels extending into the bronchial mucosa consistent with the diagnosis of bronchial Dieulafoy's syndrome (Fig. 4).

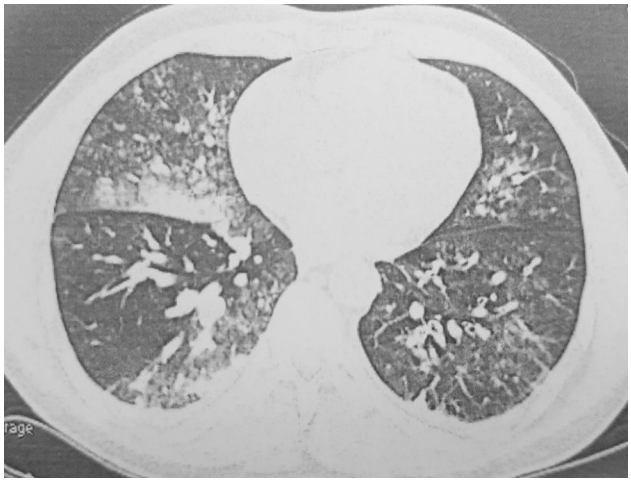

Fig. 2 Ground glass opacities of both lungs in computed tomography scans

The postoperative course was uneventful aside from a chest wall hematoma. A follow-up 4 months later, the patient was well with no further episodes of hemoptysis.

Discussion

Dieulafoy's disease has been first reported in 1898 by Georges Dieulafoy usually affecting the digestive tract [1]. The bronchial location of this disease (BDD) has been first reported by Sweerts et al. in 1995 [2]. It's an extremely rare affection which may manifest by massive hemoptysis. Over the past decade, cases of the disease have been increasingly outlined. A recent systematic review of the literature published by Qian et al. collected 73 cases from 1995 to 2019 [3]. We reported a case of BDD and searched the databases of PubMed, Google Scholar, Web of science and Chinese National Knowledge Infrastructure using the terms "bronchial Dieulafoy's disease or Dieulafoy's disease of the bronchus". After removing repeated cases and incomplete ones, we identified 62 articles and 90 cases [2–64] from January 1995 to December 2022. To our knowledge, this case would be the first one reported in Tunisia and North Africa.

The cause of the disease is still unknown. Theories vary from congenital vascular malformations to bronchial injury secondary to previous infections. Parrot et al. [15] suggested a possible association with inflammatory

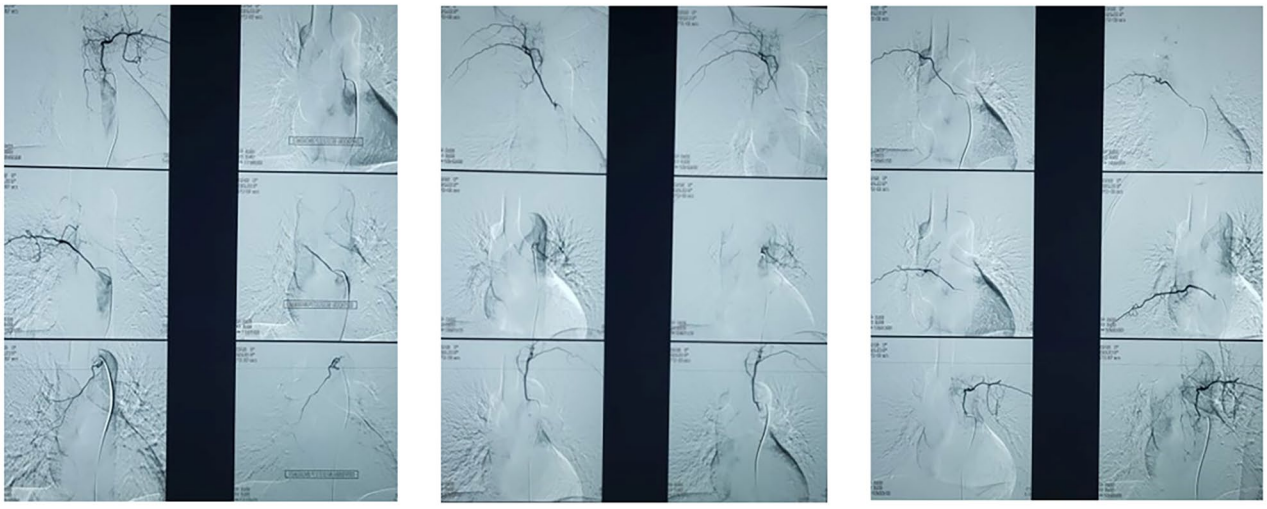

Fig. 3 Angiographic arterial embolization

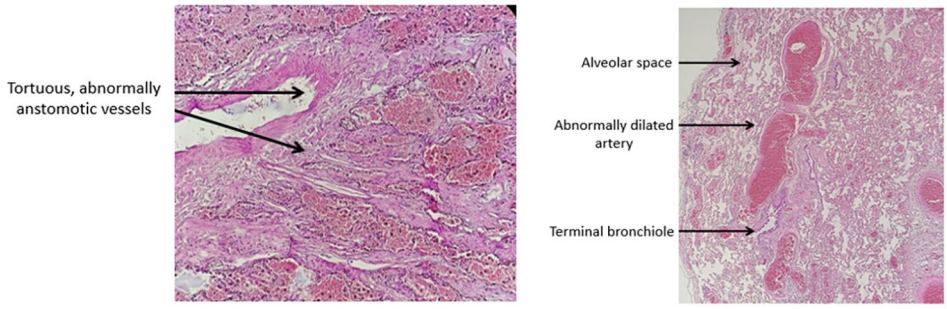

Fig. 4 Pathological findings: Abnormally dilated, sinuous and anastomotic vessels extending into the bronchial mucosa

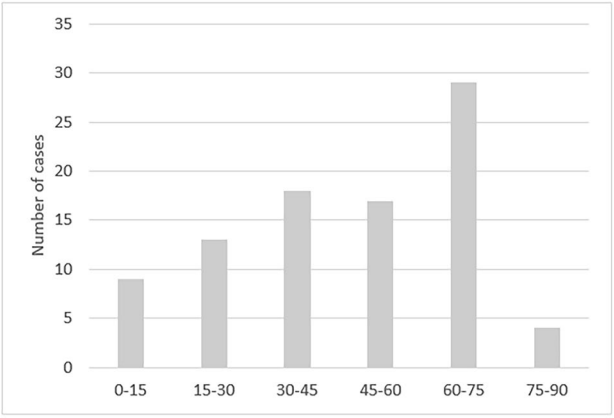

Fig. 5 Distribution by age of 90 cases of Dieulafoy's disease

Table 1 General information of patients (N = 90)

General information	Number of cases	Proportion (%)
Smoking history		
Not mentioned	21	23.3
Yes	38	42.2
No	31	34.4
Respiratory diseases		
Tuberculosis	12	13.3
Chronic obstructive pulmonary disease	9	10
Repeated infection	8	8.9
Bronchiectasis	6	6.7
Asthma	2	2.2
Pulmonary hypertension	1	1.1
Pulmonary fibrosis	1	1.1
Clinical manifestations		
Hemoptysis	74	82.2
Cough	19	21.1
Dyspnea or acute respiratory failure	11	11.1
Pulmonary infection	4	4.4
Fever	4	4.4
Chest pain	1	1.1

lesions in tuberculosis or stretching and dilation of the bronchial artery. Advanced age and tobacco smoking have been implicated in the increase of bleeding-related complications [27]. However, the disorder may affect people at every age especially middle-aged adults [3], and also non-smokers. There were 34 females and 56 males in 90 patients reported in this review with a male to female ratio of 1.6. Their age varied form 9 months to 85 years old (Fig. 5). History of smoking was found in 38 cases and previous respiratory diseases are summarized in Table 1.

Clinical manifestations are non-specific, but the most common one was recurrent hemoptysis (Table I). Massive and even fatal hemoptysis may occur especially while performing bronchoscopy guided biopsy. Other symptoms such as a cough [24, 30, 45, 57], chest pain [28], infection [17, 25, 45] or respiratory failure [4, 41, 43, 46] can be reported by patients.

Table 2 Bronchoscopy findings (N = 84)

Bronchoscopy findings	Number of cases	Proportion (%)
Endobronchial hemorrhage	7	8.3
Blood clots or thrombus	16	19
Nodular or prominent lesions	44	52.4
Only a white cap	3	3.6
Non pulsatile process	8	9.5
Normal	8	9.5

Establishing the diagnosis of Dieulafoy's disease can be quite difficult. An exhaustive evaluation was lead with chest X-rays, computed tomography (CT) scans, bronchoscopies, biopsies and bronchial angiographies in historical cases.

In Dieulafoy's disease of the bronchus, chest X-rays and chest CT are rarely contributive to the diagnosis. The relevance of this exam is its contribution to excluding other lung diseases causing the bleeding. In this review, Chest X-ray was performed for 38 patients. Whereas, 79 patients had chest CT. They showed mostly manifestations of an intrapulmonary hemorrhage with ground glass opacities (24 cases) and inflammatory changes (34 cases). Other findings were atelectasis (10 cases), consolidation (5 cases), bronchiectasis (10 cases) and endobronchial nodes or masses (6 cases). Due to lack of specificity and sensitivity, most authors agree that chest X-rays and CT scans are not the best modality to diagnose Dieulafoy's disease.

Bronchoscopy was performed for most patients (84 cases). It mainly showed a mucosal protrusion in the site of the bleeding bronchus, which diameter can be only a few millimeters [3, 64]. The mucosa covering the protruding surface may look like a "white cap", without pulsations. The surrounding mucosa can be normal or congested and other lesions can be found (Table 2).

These bronchoscopic findings in BDD are not always diagnostic since the abnormal vessel is usually pinpoint mucosal defect surrounded by normal-looking mucosa. Moreover, a small lesion can be undetected due to pooling of blood or clots within bronchial lumen or to a distal localization below the subsegmental bronchus. Bronchial angiography can contribute to the diagnosis as it can show a rich blood supply to the corresponding site of the lesion; a deformed, tortuous and dilated artery with signs of bleeding [23]. Also, when detected, these abnormalities can indicate a selective bronchial embolization, which has an important therapeutic value. However, lesions of the arteries may not be visualized, as it was the case for 2 cases in the literature [3] and for our patient. The bronchial arteries usually originate from the proximal descending thoracic aorta. Arteries that originate elsewhere in the aorta or from other vasculature are termed ectopic [65]. Right bronchial artery occasionally

originates from the aorta but more commonly shares its origin with another artery, usually an intercostal artery. Choi et al. [66] evaluated in their study the spectrum of variations in bronchial artery and among ectopic origins, concavity of the aortic arch was the most common.

In most cases, the abnormal lesions were located in the right bronchus (64 cases), which may be due to its anatomical structure. The diversity of embryonic development of the right bronchial artery accounts for a higher risk of abnormalities causing the congenital BDD [5]. Cases of BDD of left bronchus and bilateral bronchi were also reported (Table 3). In most cases, bleeding originated from the bronchial artery system [3].

Among 90 cases reported, biopsies were attempted for 28 patients whom presented a nodular lesion without a typical vascular lesion. It was primarily suspected to be an endobronchial mass or carcinoid tumor. Twenty patients had severe hemoptysis [4, 6, 11, 17–19, 21, 24, 25, 30, 36, 37, 45, 48, 49, 52, 57, 61–63] and ten patients did not survive [4, 6, 17, 24, 25, 30, 45, 49, 52, 61].

Bronchial biopsies in such diseases entail the risk of triggering fatal hemoptysis. Since 2014, with a better understanding of Dieulafoy's disease, biopsies have been avoided for nodules suspected to be caused by Dieulafoy's disease [26], which has reduced the risks of massive hemorrhage.

In 2010, Guiroli et al. [19] have demonstrated the clinical utility of endobronchial ultrasound EBUS in the evaluation of bronchial alteration suspicious of Dieulafoy's lesion. This technique can be helpful to clarify the nature of the nodular lesion and contributes to the diagnosis, avoiding potentially disastrous interventions [32]. The major manifestation is a fluid echo-free zone in the submucosal lesion. The Doppler mode can be used to detect blood flow. However, convex probe EBUS cannot reach the upper lobe bronchus nor segmental bronchus. Radial probe EBUS can be used instead but it has no doppler mode and cannot determine blood flow within the lesion.

To make a definite diagnosis, many researchers consider that pathological examination of biopsies, surgical or autopsy specimens is required. However, there are no uniform diagnostic criteria and due to risks involved, the need for pathological diagnosis remains controversial. In some cases, the diagnosis was only based on the findings of bronchoscopy and bronchial angiography [28, 29, 31].

The pathological exam usually shows an arterial malformation in the bronchial submucosa. The tortuous, dilated and deformed artery forms small nodules coated with bronchial mucosa and protruding from the bronchial lumen [10, 15]. Diagnosis is confirmed when a dysplastic artery is identified in the bleeding territory without evidence of other underlying lung disease, vasculitic changes or neoplasm.

Table 3 Lung segment localization of bronchial Dieulafoy's disease (N = 90)

Lung segment localization	Number of cases	Proportion (%)
Right bronchus	64	71.1
• Right main bronchus	3	3.3
• Right upper lobe bronchus	16	17.8
• Right intermediate bronchus	7	7.8
• Right middle lobe bronchus	21	23.3
• Right lower lobe bronchus	24	26.7
Left bronchus	25	27.8
• Left main bronchus	5	5.6
• Left upper lobe bronchus	10	11.1
• Left lower lobe bronchus	10	11.1
Bilateral bronchus	5	5.6

Table 4 Treatment of bronchial Dieulafoy's disease (N = 90)

Treatment plan	Number of cases	Proportion (%)
Medical treatment	9	10
Selective bronchial artery embolization only	32	35.6
Selective bronchial artery embolization (unsuccessful) then surgery (lobectomy)	15	16.7
Selective bronchial artery embolization (successful) then surgery (lobectomy)	9	10
Surgery only	15	16.7
Bronchoscopic treatment		
Freeze + place silica gel stent	1	1.1
Argon ion coagulation	2	2.2
Endotracheal intubation + mechanical ventilation	4	4.4

Treatment options include medical treatment, surgical lung resection, selective bronchial artery embolization (SBAE) and bronchoscopic ablation. Among 90 patients reported, 32 patients were treated with only SBAE, whereas, 15 patients had pulmonary lobectomy (Table 4). Two patients were successfully treated with argon plasma coagulation through bronchoscope [29, 39]. One patient failed to receive cryotherapy, and then placed silica gel stent [14].

Conservative treatment and the use of hemostatic agents is rarely efficient in stopping the hemorrhage. Niu al. [40] have reported that pituitrin and thrombin may occasionally have good therapeutic effect in some cases of infantile bronchial Dieulafoy's disease. Bronchoscopic ablation has been tried in a minority of cases [3] and is not without perils. Mediastinitis, esophageal injuries, and broncho esophageal fistulas are all potential complications [46]. Selective bronchial artery embolization is often performed as a first-line treatment and is efficient in most patients [64]. But hemoptysis may reoccur after the procedure. Among 56 patients treated with first-line SBAE, 24 underwent surgery afterwards due to unsuccessful embolization or to prevent reoccurrence of bleeding. In cases where intercostal artery embolization failed, authors reported

that the abnormal blood vessels originated from the pulmonary artery, requiring pulmonary lobectomy to control the bleeding. Up to date, surgery has been the main definitive treatment with a success rate of nearly 100% in all reports [3]. Recurrence of hemoptysis in unlikely after resection of the diseased lung lobe.

Conclusions

Dieulafoy's disease of the bronchus is a rarely reported, and possibly underdiagnosed, cause of life-threatening hemoptysis. It should be included in the differential diagnosis of patients with massive hemoptysis with no other evident etiology. Bronchial angiography and EBUS may be highly suggestive of this disease. While SBAE is a less invasive procedure, surgical treatment remains a lifesaving approach that reduces the probability of recurrence. Therefore, it is the best choice which also allows an accurate histopathological diagnosis.

List of abbreviations

BBD	bronchial Dieulafoy's disease
CT	Computed tomography
EBUS	endobronchial ultrasound
SBAE	selective bronchial artery embolization

Authors' contributions

Daboussi Salsabil: conceived the work, reviewing and finalization of the manuscript. Kacem Marwa: collected clinical details, analysis and interpretation of data, contriubuted to writing. Boubaker Nouha: approval of the final version. Chaabene Mariem: approval of the final version. Aichaouia Chiraz: approval of the final version. Mhamdi Samira: approval of the final version. Moatemri Zied: approval of the final version.

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Yes.

Declarations

Ethical approval

We obtained the approval of **the local committee for the protection of individuals** of the military hospital of Tunis.

Competing interests

No competing interests.

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