Bronchial artery enlargement may be the cause of recurrent haemoptysis in Behçet's syndrome patients with pulmonary artery involvement during follow-up

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ABSTRACT

Objective. *Haemoptysis occurring in a Behçet's syndrome (BS) patient with pulmonary artery involvement (PAI) during follow-up is usually regarded as PAI relapse. However, bronchial artery enlargement (BAE) may be the source of haemoptysis in some patients.*

Methods. *A chart review at the end of December 2014 revealed 118 patients with PAI in our centre since 1979. Nine (all men) had recurrent haemoptysis during follow-up which could not be explained with relapse of PAI.*

Results. *Haemoptysis recurred a median of 1.5 years (IQR: 9 months-5 years) during follow-up. Thorax CT scans did not show relapse of PAI or emergence of BAE. The patients were treated empirically but continued to complain of occasional haemoptysis thereafter. BAE was detected in 8 patients after a median follow-up of 9 years (IQR: 5-12 years). Six patients underwent bronchial artery embolisation that was repeated in 3. One patient with severe pulmonary hypertension died 3 weeks later. The remaining 5 are under follow-up for between 5 months-9 years. Pulmonary infarction and mild hemiparesis occurred in 2 patients after embolisation. One patient died with haemoptysis before undergoing embolisation. Another one with small BAE is under follow-up for 8 years without embolisation. The source of bleeding could not be determined in 1 patient who is now haemoptysis free for 5 years.*

Conclusion. *BAE may be the source of recurring and fatal haemoptysis in BS patients with PAI during follow-up. Embolisation appears to be a life-saving procedure.*

Introduction

Behçet's syndrome (BS) is a systemic vasculitis of unknown aetiology characterised by recurrent oral and genital ulcers, papulopustular skin lesions, a sight-threatening uveitis, gastrointestinal involvement, central nervous system involvement, and arthritis (1, 2). Blood vessels of any size and type may also be involved leading to the formation of thrombosis including intracardiac thrombus, occlusions, and aneurysms (3-5).

The most feared type of vascular involvement is the pulmonary artery involvement (PAI) due to its high mortality risk (6). The frequency of PAI has been reported as between 1-4% in retrospective series with men making up the vast majority of affected patients (7). Pulmonary artery thrombosis (PAT) and pulmonary artery aneurysms (PAA) are the two clinical forms of PAI. Both may occur simultaneously in the same patient; PAT may evolve to PAA over time and PAA may be totally or partially thrombosed (8). In contrast to peripheral arteries, PAI is often multiple and is detected in both lungs with a predilection to involve large proximal to medium-sized lower lobe pulmonary arteries (7). On the other hand, additional imaging findings in thorax computed tomography (CT) scans of patients with PAI like infiltrations, nodules and cavities and their histological examinations suggest that the scope of PAI is much more extensive and includes small vessels as well (8).

The lungs have a dual blood circulation supplied both by the pulmonary and bronchial arteries (9). Pulmonary arteries carry deoxygenated blood for gas exchange at the alveolar level and provide 99% of the blood flow. The bronchial arteries deliver oxygenated blood to the airways, lymph nodes, visceral pleura, vasa vasorum of the aorta, pulmonary arteries, and veins. There are rich anastomoses between the pul-

Table I. The demographic, clinical and treatment characteristics of the patients at the time of PAI diagnosis.

Case no	Age at BS diagnosis (years)	Age at PAI diagnosis (years)	Co-existing vascular involvement	Type of PAI	Consolidation therapy/duration	Maintenance therapy/duration
1	25	25	DVT, ICT	PAT bilateral	CYC-Pred/10 months	AZA/4.5 years
2	31	31	None	PAA bilateral	CYC-Pred/8 months	AZA/5.5 years
3	19	25	None	PAA bilateral	CYC-Pred/18 months	AZA - IFX[1]/7 years
4	35	36	None	PAA bilateral	CYC-Pred/10 months	AZA/4 years
5	17	20	STM, DVT, DST, VCI	PAA bilateral	CYC*-Pred/5 years	AZA/9 years
6	29	34	DVT, ICT	PAT right	CYC-Pred/14 months	AZA-IFX/2 years
7	31	33	STM, ICT	PAA bilateral, PAT right	CYC-Pred/15 months	AZA/1 year
8	28	34	DVT, DST, ICT	PAA bilateral	CYC-Pred/1 year	AZA/4 years
9	23	28	DST, DVT, coronary artery involvement	PAA left	CYC-Pred/ 2 years	AZA/ 3 years

[1]Infliximab for 2.5 years; *Oral cyclophosphamide. AZA: azathioprine; BS: Behçet's syndrome; CYC: cyclophosphamide; DST: dural sinus thrombosis; DVT: deep vein thrombosis; ICT: intracardiac thrombus; IFX: infliximab; PAA: pulmonary artery aneurysm; PAI: pulmonary artery involvement; PAT: pulmonary artery thrombosis; Pred: prednisolone; STM: superficial thrombophlebitis.

monary and bronchial circulation at the capillary level, and any pathology reducing blood flow to the pulmonary arteries may result in a compensatory enlargement of these anastomoses and eventually of the bronchial arteries (10). In fact, these enlarged vessels have been proposed as the main source of haemoptysis in most cases because their thin and fragile walls may rupture easily with increased blood flow and pressure (11, 12).

PAI is still the leading cause of mortality in BS, but its prognosis nowadays is considerably better than before as a result of earlier diagnosis and earlier initiation of immunosuppressive therapy. In our last series published in 2012, the survival rate among our 47 patients was 74% after a mean follow-up of 7 years (8). On the other hand, only 46% of the survivors in that study were symptom-free during their follow-ups and the remaining had some residual symptoms including recurrent haemoptysis. Haemoptysis does not always originate from pulmonary arteries and may also emerge from enlarged bronchial arteries or pulmonary-bronchial anastomoses and bronchial artery enlargement (BAE) as the source of haemoptysis has been reported in a few case reports of BS patients with PAI before (13-15). In recent years, we have the impression that we diagnose BAE as a cause of haemoptysis more frequently than before. This situation is most probably related to the advances in imaging technologies, but it may also well be due to the prolonged survival of PAI patients. This retrospective report summarises our experience with BAE in BS patients having PAI.

Materials and methods

The multidisciplinary Behçet's syndrome Outpatient Clinic at the Cerrahpasa Medical Faculty was established in 1979 and currently has approximately 10000 registered patients. A chart review at the end of December 2014 revealed 118 patients (111 men and 7 women) diagnosed as having PAI. A further search revealed that 9 of them had experienced haemoptysis during their follow-up that was not explained with a PAI relapse. We retrieved the information on the clinical history, follow-up, management and outcome of these patients from the charts. All available radiologic examinations were re-evaluated by our radiologist when needed. The ethics committee of Cerrahpasa Medical Faculty (345986/2015) approved the study.

Results

Initial presentation of the patients with PAI

All 9 patients were male and fulfilled the International Study Group Criteria at the time of the diagnosis of PAI (16) (Table I). Their mean age at the onset of BS and the diagnosis of PAI was 26.4±6 SD years and 30±5.4 SD years, respectively. The median disease duration at the time of PAI diagnosis was 5 years (IQR: 9 months - 6 years). Haemoptysis was the presenting symptom in all with 3 patients also having high fever. The levels of C-reactive protein (CRP) and erythrocyte sedimentation rate (ESR) were elevated in all patients. PAI was in the form of PAA in 6 patients, isolated PAT in 2, and the combination of PAA and PAT in one patient. BAE was evident in the initial thorax CT of a patient who was referred to us because of haemoptysis of 4 months duration. This patient had multiple partially thrombosed PAA in both lungs and multiple PAT in the right pulmonary tree. Upon diagnosis of PAI, all patients initially received 3 pulses of 1 g methylprednisolone followed by prednisolone 1 mg/kg/d and monthly boluses of 1 g cyclophosphamide as consolidation treatment. The dose of prednisolone was then tapered down according to the clinical response. After a mean duration of 1.6±1.3 SD years, cyclophosphamide was changed to azathioprine 2.5 mg/kg/d in all patients, which was given for a mean duration of 4.1±2 SD years. Two patients also received infliximab 5 mg/kg combined with azathioprine for 2 and 2.5 years, respectively.

Recurrence of haemoptysis

The first recurrence of haemoptysis was a median of 1.5 years (IQR: 9 months - 5 years) during the follow up. At this time, 5 patients were on cyclophosphamide for a mean duration of 10±4.3 SD months, 3 were receiving azathioprine as maintenance (for 4 years in 2 patients and 8 years in 1 patient) and 1 pa-

Table II. Treatment and outcome of patients after bronchial artery enlargement.

Case no, initial diagnosis	Re-evaluation of thorax CT	Bronchial or non-bronchial artery embolisation	Complication	Medical treatment	Outcome/ follow-up time
1, PAT bilateral	Chronic thrombotic changes in right pulmonary artery	Bronchial artery embolisation	None	AZA	No haemoptysis, 1.5 year
2, PAA bilateral	PAA disappeared chronic thrombotic changes in both pulmonary arteries	Bronchial artery embolisation	None	AZA	No haemoptysis, 1.5 years
3, PAA bilateral	PAA disappeared, chronic thrombotic changes in both pulmonary arteries	Bronchial artery embolisation	Lung infarction	AZA	Small amounts of haemoptysis, 6 months
4, PAA bilateral	PAA disappeared, Thrombus formation in right pulmonary artery due to prior embolisation	Non-bronchial artery (IMA) embolisation 2 times	Hemiparesis	No treatment	No haemoptysis, 9 years
5, PAA bilateral	PAA disappeared	Bronchial and non-bronchial artery (epigastric) embolisation (2 years after)	None	No treatment	Occasional and small haemoptysis, 5 years
6, PAT right	No change	Bronchial artery embolisation 2 times	None		Died due to right heart failure
7, PAA bilateral, PAT right	PAA disappeared, chronic thrombotic changes in right pulmonary artery	Not done	Not done		Died due to bleeding
8, PAA bilateral	PAA disappeared, chronic thrombotic changes in both pulmonary arteries	Not done	Not done	No treatment	No haemoptysis, 8 years
9, PAA left	PAA disappeared	Not done	Not done	No treatment	No haemoptysis, 5 years

tient was free of medical treatment for 4 years. Four patients had active mucocutaneous lesions whereas 5 patients had free of symptoms attributable to BS. CRP levels were available in 8 patients and were normal in 7. The patient with elevated CRP had an organising pneumonia. There were no signs of the development of any new PAI nor any worsening of existing PAI in thorax CT scans. The patients were treated empirically based on the amount of haemoptysis. Four patients with large amounts of haemoptysis received 1 g/d methylprednisolone pulses, 1 underwent embolisation of pulmonary arteries in addition to pulse steroids and no additional treatment was given to 4 patients complaining of small amounts of haemoptysis. These 9 patients subsequently complained of occasional recurrence of small bouts of haemoptysis and this had not led to any intervention or changes in their management.

Bronchial artery enlargement

After a median follow-up of 9 years (IQR: 5–12 years) BAE was diagnosed in 8 patients (Table II). At the time of diagnosis, 2 patients had superficial thrombophlebitis of the legs whereas the remaining 7 were free of any BS symptom. CRP and/or ESR levels were also normal in all. BAE was evident in thorax CT scans of 5 patients and additional 3 patients have been diagnosed with bronchial artery angiography.

Six patients underwent embolisation of the bronchial arteries (Figs. A, B and C). Two patients became haemoptysis free for 18 months following embolisation and are still using azathioprine. The third developed pulmonary infarction after embolisation that resolved with symptomatic treatment. Small amounts of haemoptysis recurred in this patient 4 months later and his last thorax CT showed non-bronchial collaterals originating from coeliac artery supplying the embolised segment of the lung. He is still on azathioprine therapy. The fourth patient had been treated with pulmonary artery embolisation 6 years ago. He underwent embolisation of internal mammary artery supplying the previously embolised pulmonary artery. The embolisation was repeated 4 months later because of re-bleeding. Now, after 9 years following the last embolisation, this patient is free of haemoptysis but has mild hemiparesis as a complication. The fifth patient initially underwent embolisation of the bronchial arteries and was symptom-free for 2 years. He then experienced massive haemoptysis that was treated with the embolisation of the superior epigastric artery. Now, at 6 years following the embolisation, he complains of occasional haemoptysis in small amounts and is off treatment for 5 years. The sixth patient had a longstanding history of clinically symptomatic pulmonary hypertension (WHO Class III) and was receiving bosentan and sildenafil. He underwent embolisation that was repeated 1 week later because of increased amounts of haemoptysis but died with right heart failure 3 weeks later. The seventh patient died of massive haemoptysis at the emergency unit of another centre before undergoing embolisation. The eighth patient had small sized BAE at

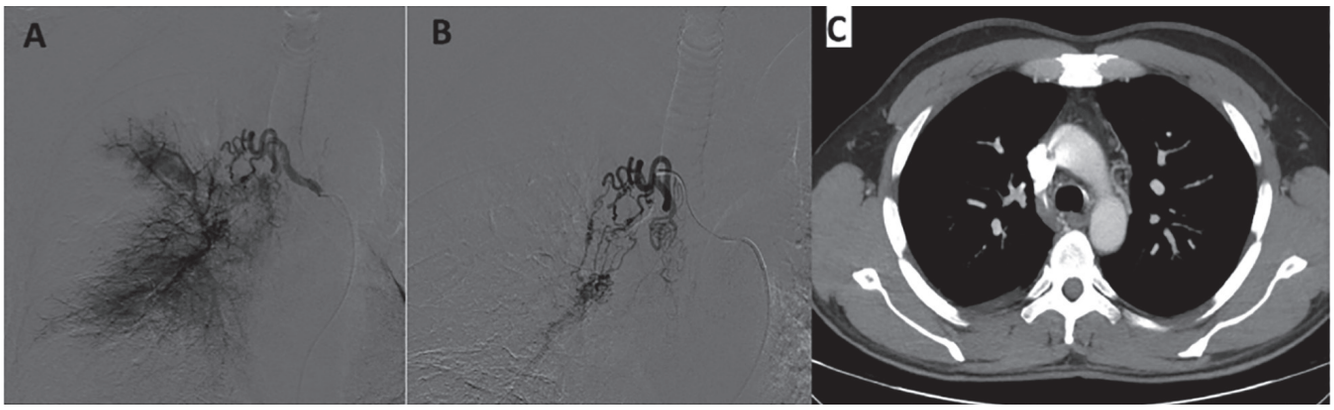

Fig. 1. The images of digital subtraction angiography before and after embolisation and MIP reconstruction of thorax CT.
Digital subtraction angiography images show enlarged and tortuous bronchial artery supplying the right middle lobe before (A) and after embolisation (B). The hypertrophic bronchial arteries can be also seen in the thick-section MIP (maximum intensity projection) reconstruction of thorax CT (C).

thorax CT and did not undergo embolisation. He is still reporting occasional small bouts of haemoptysis now being at the 8th year after the diagnosis of BAE and is off treatment for the last 2 years. In the last patient, the source of bleeding could not be demonstrated despite repeated thorax CT scans, bronchoscopies, and pulmonary angiography. Bronchial artery angiography was not performed in this patient but at the time of this writing, he was completely haemoptysis free for 5 years and was off treatment.

Discussion

BAE is the main source of haemoptysis in general (17). In the developing world, the leading cause of BAE related haemoptysis is pulmonary tuberculosis, whereas bronchiectasis and lung cancer are the most frequent causes in developed countries (18-20). BS is also counted among the causes of BAE associated haemoptysis (18). However, we could not find any reference to BAE neither in a comprehensive review on the pulmonary involvement in BS nor in a book on BS (21). These suggest that BAE in BS does not get due attention.

In 1959, Hughes and Stovin reported 2 male patients with widespread thrombosis and PAA who died with haemoptysis (22). Postmortem examination of these patients showed organised and recanalised thrombi in the systemic veins, heavy inflammatory infiltration of pulmonary arteries and degenerative and thickened bronchial arteries that had no inflammatory cell infiltration (22). The presence of BAE was also shown radiologically in subsequent reports of patients with Hughes-Stovin syndrome which is today considered to be an incomplete form of BS due to many clinical and histopathological similarities between the two conditions (23-25).

BS is known to involve arteries and veins of all sizes, but no single case of isolated BAE can be found in the literature. All of the few published BAE cases (13-15) and the patients in this report had concomitant PAI suggesting that BAE develops as a consequence of the compromised pulmonary blood flow leading to increased pressure in bronchial arteries rather than being the result of vessel wall inflammation. The late occurrence of BAE during the course of PAI as well as the lack of elevated acute phase response at the time of the diagnosis of BAE further support this view.

Some disease characteristics such as the male sex predominance, age at developing BS, disease duration until the emergence of PAI and the frequency of venous thrombosis were similar between the patients developing BAE and the 47 patients with PAI that we had reported before (8). However, patients with BAE differ from those with isolated PAI by having more frequent bilateral PAI (78% *vs*. 57%), intracardiac thrombosis (44% *vs*. 28%), dural sinus thrombosis (33% *vs*. 15%) and chronic thrombotic changes in the pulmonary arteries at thorax CT scans (78% *vs*. 60%) (8).

If a patient with PAI starts to complain of re-occurrence of haemoptysis during follow-up after initial clinical response to treatment, clinicians give most attention to the exclusion of relapsing PAI or an emerging infection in the light of the immunosuppressive therapy. Our experience now suggests that bleeding from BAE should also be put high on the list as the possible source of haemoptysis. The enlarged bronchial arteries are visualised as round, linear or nodular enhanced structures measuring more than 2 mm in diameter in thorax CT, CT angiography (Fig. C) or MRI angiography and their presence should be regarded as a sign of diverse pathologies affecting pulmonary circulation (26, 27).

Embolisation of the bronchial arteries is considered as an effective non-surgical treatment for the immediate control of haemoptysis (28, 29). Immediate and long-term control of haemoptysis was achieved in 5 out of 6 patients undergoing embolisation. The fate of the patient who died of massive haemoptysis before undergoing embolisation also underlines the life-saving efficacy of this intervention. Our single patient who died despite embolisation had advanced right-heart failure due to long-standing and severe pulmonary hypertension (mean pulmonary artery pressure: 90 mmHg). It is to be noted that systolic pulmonary artery pressure (sPAP) was mildly elevated in additional 4 patients (mean sPAP: 29±8 SD mmHg) supporting our previous observation that PAI may lead to elevations in estimated pulmonary artery pressure (30). Our experience also tells us that embolisation procedure can be effectively and safely

repeated when haemoptysis recurs. In fact, repeat embolisation for patients with recurrent haemoptysis has been reported to increase the non-recurrence rates for 2–5 years (31, 32). Hemiparesis, as we have observed in 1 patient is the most serious complication of embolisation occurring at a frequency of 1–6% (33).

The main limitation of our study is its retrospective design that allows us to assess only those patients complaining of haemoptysis during follow-up. A prospective study is definitely needed to understand what percentage of PAI patients develops BAE over time.

In conclusion, haemoptysis during follow-up of a BS patient with PAI may be related to BAE. Embolisation of the bronchial arteries appears to be a life-saving procedure and can be repeated in the same patient when haemoptysis recurs.

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