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Diffuse alveolar haemorrhage with suspected idiopathic pulmonary hemosiderosis and decrease in lung diffusing capacity and chronic respiratory failure

Kotaro Iwasaki , Yasuo Matsuzawa, Hiroki Wakabayashi, Kotaro Kumano

Department of Internal Medicine, Toho University Medical Center Sakura Hospital, Sakura, Japan

Correspondence to

Dr Kotaro Iwasaki;
kotaro.iwasaki@med.toho-u.ac.jp

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SUMMARY

Idiopathic pulmonary hemosiderosis (IPH) is a rare disease of unknown aetiology that causes recurrent episodes of diffuse alveolar haemorrhage (DAH). A male patient in his 50s had repeatedly experienced hemoptysis for the past 6 years, along with a decrease in the pulmonary diffusing capacity and chronic respiratory failure. After a 6-year follow-up, the patient experienced sudden exacerbation of hemoptysis and respiratory failure, and he was hospitalised. A CT of the chest revealed diffuse pulmonary infiltrates, whereas the bronchoalveolar lavage revealed hemosiderin-laden macrophages. Thus, the patient was diagnosed with DAH. As all diseases that cause DAH other than IPH were negative, the patient was suspected of IPH. He was treated with a combination of glucocorticoids and azathioprine, and his hemoptysis and chronic respiratory failure improved; however, the decrease in the pulmonary diffusing capacity did not improve. Treating adult-onset IPH with glucocorticoids and azathioprine might not improve pulmonary diffusing capacity.

BACKGROUND

Idiopathic pulmonary hemosiderosis (IPH) is a rare disease of unknown aetiology that causes recurrent episodes of diffuse alveolar haemorrhage (DAH). IPH is extremely rare, and its incidence in children is estimated to be 0.24–1.23 per 1 000 000.^{1,2} Recently, a previous study has reported IPH in adults aged ≥ 30 years.³ However, there are not many reports on adult-onset IPH. Many cases of adult-onset IPH have a chronic history; however, there are few reports on acute exacerbation in long-term chronic respiratory dysfunction.⁴

A previous study has confirmed respiratory dysfunction in IPH, such as restrictive ventilatory and pulmonary diffusing capacity impairments.⁵ To the best of our knowledge, there is no report on adult-onset IPH with only pulmonary diffusing capacity impairment. Here, we report the case of a patient suspected of having an adult-onset IPH in his 50s who presented with recurring hemoptysis, a decrease in pulmonary diffusing capacity and chronic respiratory failure, and was treated with glucocorticoids and azathioprine. We examined the patient's disease course before and after treatment, along with respiratory function. Our case report anonymised the patient to protect his privacy and obtained written informed consent.

A male patient in his 50s presented with recurring hemoptysis of unknown aetiology and chronic

respiratory failure for 6 years before he was hospitalised. Until the onset of hemoptysis 6 years prior, the patient smoked 30 cigarettes a day for 30 years. Before hospitalisation, a CT of the chest was performed; however, no pulmonary opacity other than emphysematous changes was observed. Pulmonary function tests performed before hospitalisation, when there were no symptoms of hemoptysis, showed a forced expiratory volume in 1 s (FEV₁) of 2.37 L, forced vital capacity (FVC) of 3.07 L, total lung capacity (TLC) of 4.67 L, residual volume (RV) of 1.55 L, FEV₁/FVC ratio of 77.2%, percentage-predicted FVC (FVC_{90pred}) of 81.2% and percentage-predicted TLC (TLC_{90pred}) of 82.5% (table 1). Obstructive and restrictive ventilatory impairment was not confirmed; therefore, chronic obstructive pulmonary disease (COPD) was not considered. However, the percentage-predicted diffusing capacity of the lungs for carbon monoxide (DLCO_{90pred}) was 27.1% and the DLCO/alveolar volume (VA) ratio was 2.32 mL/min/mm Hg/L, confirming notable pulmonary diffusing capacity impairment (table 1). Home oxygen therapy (oxygen via the nasal cannula at 2.5 L/min) was administered to treat the patient's chronic respiratory failure of unknown aetiology; however, dyspnoea and hemoptysis exacerbated, and he was transported for emergency hospitalisation.

On admission, the patient's consciousness level was normal. His body height was 173 cm, weight 93 kg, body temperature 38.5°C, pulse 133 beats/min, respiratory rate 32 breaths/min, blood pressure 158/87 mm Hg and oxygen saturation 64% on room air. Coarse crackles were audible in the chest, and anaemia was confirmed in the palpebral conjunctiva. There were no abnormal findings in any other physical examinations. Other than dyspnoea and hemoptysis, there was no significant symptom, such as arthralgia, peripheral neuropathy, rash, muscle pain, chest pain or abdominal pain. Laboratory findings revealed an inflammatory response and iron-deficiency anaemia (table 2). Chest X-ray revealed patchy opacity from the middle lung field to the lower lung field (figure 1). Contrast-enhanced CT of the chest did not reveal abnormality in chest blood vessels; however, diffuse pulmonary infiltrates were disseminated across the whole lung field, in addition to the existing emphysematous changes (figure 2). Bronchoalveolar lavage (BAL) was performed by wedging a bronchofiberscope into the right middle lobe (B4), and three serial aliquots of 50 mL of physiological saline



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Table 1 Pulmonary function tests

	Pre-treatment		1-year treatment		1.5-year treatment	
	Patient	Normal	Patient	Normal	Patient	Normal
FEV ₁ (L)	2.37	3.26	2.67	3.10	2.67	3.01
FVC (L)	3.07	3.78	3.48	3.69	3.70	3.64
TLC (L)	4.67	5.66	5.38	5.61	5.71	5.55
FEV ₁ /FVC ratio (%)	77.2	Within 5% of the predicted ratio	76.7	Within 5% of the predicted ratio	72.2	Within 5% of the predicted ratio
FEV _{1%} pred (%)	72.7	80–120	86.1	80–120	88.7	80–120
FVC _{%pred} (%)	81.2	80–120	94.3	80–120	101.6	80–120
TLC _{%pred} (%)	82.5	80–120	95.9	80–120	102.9	80–120
DLCO _{%pred} (%)	27.1	80–120	30.0	80–120	30.7	80–120
DLCO/VA ratio (mL/min/mm Hg/L)	2.32	5.06	1.76	4.91	1.80	4.88
RV (L)	1.55	1.54	2.03	1.6	1.93	1.58

DLCO_{%pred}, percentage-predicted diffusing capacity of the lungs for carbon monoxide; FEV₁, forced expiratory volume in 1 s; FEV_{1%}pred, percentage-predicted FEV₁; FVC, forced vital capacity; FVC_{%pred}, percentage-predicted FVC; Normal, normal value; Patient, patient's value; RV, residual volume; TLC, total lung capacity; TLC_{%pred}, percentage-predicted TLC; VA, alveolar volume.

were instilled and withdrawn. BAL fluid (BALF) was bloody, and Berlin blue stain revealed a large number of hemosiderin-laden macrophages (figure 3).

Non-invasive positive pressure ventilation (NPPV) was performed, and treatment with pulse steroid (intravenous methylprednisolone in a pulse dose, 1000 mg/day for 3 days) was started. On day 4 of hospitalisation, treatment was changed to intravenous administration of 80 mg of methylprednisolone per day (1 mg/kg/day) for 9 days. On day 10 of hospitalisation, NPPV was removed. Subsequently, the patient's hemoptysis improved. On day 13 of hospitalisation, treatment was changed to oral administration of 50 mg of prednisolone per day, with a gradual decrease. On day 20 of hospitalisation, the patient's respiratory failure improved, and he was discharged.

DIFFERENTIAL DIAGNOSIS

The patient was diagnosed with DAH, and a differential diagnosis was made for diseases that cause DAH. There was no physical finding of collagen diseases or vasculitis, such as arthralgia, peripheral neuropathy, rash, muscle pain or abdominal pain. Laboratory findings did not reveal significant detection of haematuria, proteinuria, renal dysfunction or autoantibodies (table 2). Based on physical and laboratory findings, antineutrophil cytoplasmic antibody associated vasculitis, immunoglobulin A vasculitis, Goodpasture syndrome (anti-glomerular basement membrane disease) and collagen diseases were negative. Bleeding time, prothrombin time, activated partial thromboplastin time and platelet counts were normal. No organ other than the lung had any bleeding; thus, blood disease-associated bleeding was also negative. Contrast-enhanced CT of the chest did not reveal pulmonary infarction or pulmonary embolism. Cardiac ultrasound excluded heart diseases such as mitral valve stenosis. Cardiac catheterisation did not present any finding indicative of pulmonary hypertension and heart failure. The mean pulmonary artery pressure was 19 mm Hg, whereas the mean pulmonary capillary wedge pressure was 13 mm Hg. There was no history of bone marrow transplantation, lung transplantation, radiation therapy or medication that could cause DAH. Infections such as leptospirosis and infective endocarditis, which could cause DAH, were excluded from the medical history of conditions that were recurring in the past 6 years. Bacteria and fungi were not detected in blood culture. Only indigenous bacteria were

isolated in the BALF culture. Given that the patient's symptom improved without the administration of anti-infective drugs, infection was excluded. IPH is diagnosed when three main symptoms (anaemia, hemoptysis and diffuse pulmonary infiltrates) are found and when other diseases that can cause DAH are excluded. Our patient experienced hemoptysis with iron-deficiency anaemia and diffuse pulmonary infiltrates intermittently in the past 6 years and was hospitalised. As all diseases that cause DAH other than IPH were negative, the patient was suspected of IPH.

OUTCOME AND FOLLOW-UP

After being discharged, the patient's hemoptysis and dyspnoea recurred when the oral administration of prednisolone was gradually reduced to 12 mg/day. Adding oral administration of azathioprine at 100 mg/day improved symptoms. Ultimately, the prednisolone dose could be reduced to 8 mg/day. Although diffuse pulmonary infiltrates improved, the emphysematous change and dyspnoea on exertion remained (figure 4). One year after hospitalisation and continuous administration of 8 mg/day of prednisolone and 100 mg/day of azathioprine, pulmonary function tests were performed when the patient was not experiencing hemoptysis. Results showed that FEV₁ was 2.67 L, FVC 3.48 L, TLC 5.38 L, RV 2.03 L, FEV₁/FVC ratio 76.7%, FVC_{0%pred} 94.3%, TLC_{0%pred} 95.9%, DLCO_{0%pred} 30.0% and DLCO/VA 1.76 mL/min/mm Hg/L (table 1). Although FVC_{0%pred} and TLC_{0%pred} significantly improved, improvements in FEV₁ and DLCO_{0%pred} were limited, and pulmonary diffusing capacity impairment persisted. However, the patient no longer required oxygen during rest, and his inflammatory response and anaemia improved. At the time of conducting this study, more than 1.5 years after hospitalisation, oral administration of 8 mg/day of prednisolone and 100 mg/day of azathioprine was continued. The pulmonary function test results were FEV₁ of 2.67 L, FVC 3.70 L, TLC 5.71 L, RV 1.93 L, FEV₁/FVC ratio 72.2%, FVC_{0%pred} 101.6%, TLC_{0%pred} 102.9%, DLCO_{0%pred} 30.7% and DLCO/VA 1.80 mL/min/mm Hg/L (table 1). Although pulmonary diffusing capacity did not improve, the patient continued outpatient treatment without hemoptysis or recurrence of respiratory failure.

Table 2 Laboratory findings on admission

Test	Patient's value	Normal value (male)
Red blood cells ($\times 10^{12}/L$)	2.4	4.3–5.7
Haemoglobin (g/L)	105	135–175
Hematocrit (%)	29.7	39.7–52.4
White blood cells ($\times 10^9/L$)	12.1	3.3–9.0
Neutrophils (%)	87	35–77
Eosinophils (%)	1	0–8
Basophils (%)	0	0–2
Monocytes (%)	3	2–10
Lymphocytes (%)	8	8–49
Platelets ($\times 10^9/L$)	172	140–340
Total protein (g/dL)	7.2	6.7–8.3
Albumin (g/dL)	4.0	3.8–5.2
AST (IU/L)	25	10–40
ALT (IU/L)	13	5–45
LDH (IU/L)	1240	120–240
BUN (mg/dL)	14.5	8.0–20.0
Creatinine (mg/dL)	0.97	0.61–1.04
Na (mEq/L)	140	137–147
K (mEq/L)	4.2	3.5–5.0
Cl (mEq/L)	107	98–108
Fe ($\mu g/dL$)	40	50–200
Ferritin (ng/dL)	19.3	21.81–274.66
C reactive protein (mg/dL)	4.49	≤ 0.30
PT (s)	14.0	11.7
APTT (s)	26.2	29.4
D-dimer ($\mu g/mL$)	7.2	< 1.0
BNP (pg/mL)	27.1	≤ 18.4
Immunoglobulin G (mg/dL)	1370	870–1700
Immunoglobulin A (mg/dL)	192	110–410
Immunoglobulin M (mg/dL)	19	33–190
C3 (mg/dL)	88	65–13
C4 (mg/dL)	28	13–35
CH50 (mL)	32	30–46
ANA (times)	< 40	< 40
dsDNA (IU/mL)	2	≤ 12
aCL (U/mL)	< 8	< 10
RF (IU/mL)	< 3	≤ 15
CCP (IU/mL)	< 0.5	< 4.5
RS	–	–
Scl-70	–	–
ACA	–	–
Top 1	–	–
RNP	–	–
SS-A	–	–
SS-B	–	–
MPO-ANCA (IU/mL)	< 0.5	< 3.5
PR3-ANCA (IU/mL)	< 0.5	< 2.0
Anti-GBM (U/mL)	1.5	< 7.0
Urinalysis		
Protein	–	–
Sugar	–	–
Blood	–	–
Blood gas analysis (room air)		
pH	7.506	7.350–7.450
PaO ₂ (mm Hg)	22.6	75.0–100.0
PaCO ₂ (mm Hg)	28.2	35.0–45.0
HCO ₃ ⁻ (mmol/L)	17.5	20.0–26.0

ARS: anti-aminocyl-tRNA synthetase antibody; ACA, anti-centromere antibody; aCL, anti-cardiolipin antibody; ALT, alanine aminotransferase; ANA, anti-nuclear antibody; anti-GBM, anti-glomerular basement membrane antibody; APTT, activated partial thromboplastin time; AST, aspartate aminotransferase; BNP, brain natriuretic peptide; BUN, blood urea nitrogen; C3, complement component 3; C4, complement component 4; CCP, anti-cyclic citrullinated peptide; CH50, 50% haemolytic unit of complement; dsDNA, anti-double-stranded DNA; LDH, lactate dehydrogenase; MPO-ANCA, myeloperoxidase-antineutrophil cytoplasmic antibody; PaCO₂, arterial carbon dioxide pressure; PaO₂, arterial oxygen pressure; PR3-ANCA, proteinase-3-antineutrophil cytoplasmic antibody; PT, prothrombin time; RF, rheumatoid factor; RNP, anti-ribonucleoprotein; Scl-70, anti-Scl-70 antibody; SS-A, anti-SS-A antibody; SS-B, anti-SS-B antibody; Top 1, anti-topoisomerase 1 antibody.



Figure 1 Chest X-ray on admission. Patchy opacity is revealed from the middle lung field to the lower lung field.

DISCUSSION

IPH refers to recurring alveolar haemorrhage where hemosiderin deposition in the lungs is confirmed; however, its aetiology is unknown. Glucocorticoids and immunosuppressive agents are reportedly effective in treating IPH, and recurrent alveolar haemorrhage and structural destruction of pulmonary alveoli are likely caused by immunological abnormalities.⁶ A recent study reported that the pathogenesis of IPH most probably involves an immunologic mechanism with a genetic predisposition.⁷

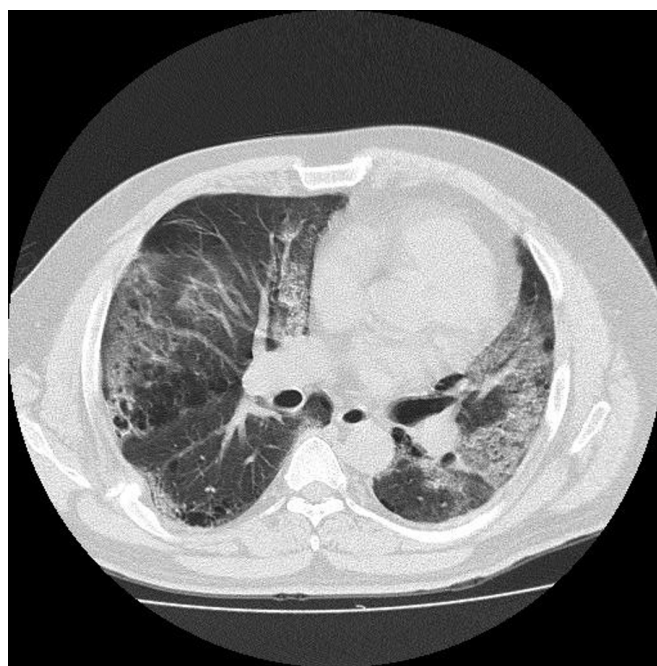


Figure 2 CT of the chest on admission. In addition to existing emphysematous changes, diffuse pulmonary infiltrates were disseminated throughout the overall lung field.

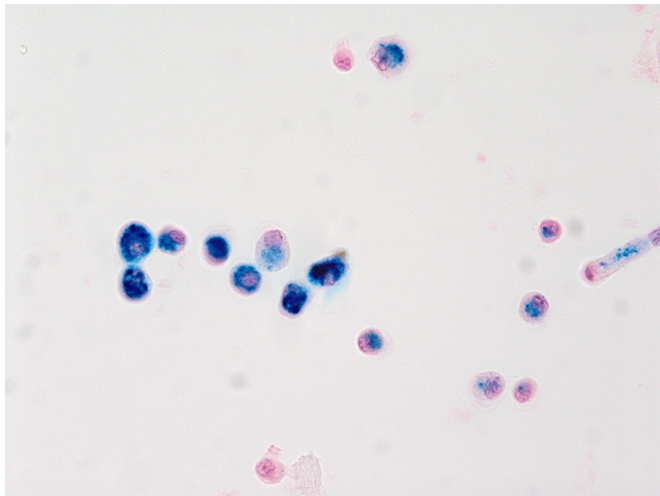


Figure 3 Bronchoalveolar lavage fluid (BALF) analysis. After inserting a bronchofiberscope into the right middle lobe (B4), 150 mL of physiological saline was administered and 44 mL of BALF was recovered. In BALF analysis (Berlin blue stain, $\times 60$), hemosiderin-laden macrophages were revealed. BALF cell counts: $2.0 \times 10^5/\text{mL}$ (macrophages 97%, neutrophils 1% and lymphocytes 2%).

To diagnose IPH, other diseases that cause DAH must be excluded. Although no pathological finding of the lungs leads to an IPH diagnosis, lung biopsy is essential for excluding other diseases.⁸ However, even if IPH is diagnosed by a lung biopsy, a future diagnosis of vasculitis is possible. Thus, continuous vigilant observation is necessary for all cases.⁹ Our patient presented with severe respiratory failure at the time of admission, and lung biopsy could not be performed at that time. Although lung biopsy was possible after treatment, his symptoms had improved, and since the risk of biopsy was higher than the benefit, treatment continued without a lung biopsy. Since we were unable to



Figure 4 CT of the chest after administering 8 mg/day of prednisolone and 100 mg/day of azathioprine. There is residual emphysematous change, but diffuse pulmonary infiltrates have disappeared.

perform a lung biopsy, we could not make a definitive diagnosis of IPH; however, vasculitis was clinically unlikely and IPH was strongly suspected.

IPH is extremely rare. A Swedish retrospective study reported that annually, there are 0.24 incidences of IPH per 1 000 000 children aged ≤ 11 years.¹ A Japanese retrospective study reported that annually, there are 1.23 incidences of IPH per 1 000 000 children aged ≤ 14 years.² Generally, 80% of patients with IPH are childhood-onset, whereas 20% are adult-onset.¹⁰ Whereas the incidence is low in children, it is even lower in adults. Thus, a large-scale randomised controlled trial cannot be conducted on adult-onset IPH, and treatment must be determined based on case reports and case series. To prevent IPH recurrence, glucocorticoids and immunosuppressive agents (eg, hydroxychloroquine, azathioprine and cyclophosphamide) are sometimes used together.^{11 12} Saeed *et al* administered glucocorticoids in 17 children and achieved a 5-year survival rate of 86% regardless of the use of immunosuppressive agents (hydroxychloroquine and azathioprine).¹³ A previous report had stated that the prognosis of IPH was about 2.5 years.¹⁴ These reports indicate that glucocorticoids and immunosuppressive agents are effective in treating IPH. The combined use of corticosteroids and azathioprine has been reported as the ideal method of preventing IPH exacerbation.³ In our case, when the prednisolone dose was gradually reduced from 15 mg to 12 mg, hemoptysis and respiratory failure recurred. Adding 100 mg of azathioprine improved hemoptysis and respiratory failure, and prednisolone could be reduced to 8 mg. However, the pulmonary diffusing capacity impairment did not improve as observed on pulmonary function tests. Thus, we discussed the cause of decrease in DLCO in the present case. Initially, we suspected that COPD or pulmonary hypertension caused severe decrease in DLCO. However, since restrictive ventilatory impairment was not confirmed and decrease in FEV₁ was limited, COPD was excluded.¹⁵ Moreover, we performed cardiac catheterisation, but pulmonary hypertension was not confirmed. Since COPD was excluded, we considered the possibility that the emphysematous changes in the CT of the chest was due to hemosiderin deposition through structural destruction of

Patient's perspective

I had been suffering from occasional hemoptysis since 6 years before the hospitalisation. I had a busy work schedule and was unable to get a detailed testing done. My breathing became gradually difficult for some unknown reason, and I started with home oxygen therapy. A month before, I was hospitalised and hemoptysis became consistent, but I persevered. However, on the day of hospitalisation, hemoptysis and difficulty breathing became suddenly exacerbated, so I requested an emergency transport. Once I was hospitalised, several tests were performed, the cause was identified and hemoptysis and difficulty breathing improved. After the symptoms improved, the doctor proposed a lung biopsy, but as it had a risk of pneumothorax and reduced lung function, I refused the biopsy proposal. Although I did not want the lung biopsy, I wish I got other tests done sooner. Subsequently, when the dose of prednisolone was reduced, hemoptysis and breathing problems recurred. However, administering azathioprine improved the symptoms. Today, there is no recurrence of hemoptysis, and as long as I am resting, there is no need for home oxygen therapy. I am very grateful to the doctor for diagnosing this extremely rare disease and providing an appropriate treatment.

Learning points

- ▶ In patients with recurring hemoptysis and chronic respiratory failure, idiopathic pulmonary hemosiderosis (IPH) must be included in the differential diagnosis, even for adults.
- ▶ Adult-onset IPH may cause acute exacerbation after long-term chronic respiratory failure.
- ▶ On performing pulmonary function tests, adult-onset IPH might manifest only pulmonary diffusing capacity impairment and not restrictive or obstructive ventilatory impairment.
- ▶ Treating adult-onset IPH with decreased pulmonary diffusing capacity and chronic respiratory failure with glucocorticoids and azathioprine might not improve pulmonary diffusing capacity.

pulmonary alveoli. DAH can cause increase in DLCO, but we measured DLCO when DAH was not confirmed. As a result, we considered long-term and repetitive DAH caused by IPH, which led to emphysematous change and thereby a decrease in DLCO. However, smoking may be the possible cause of emphysema instead of COPD, which resulted in decreased values for DLCO only. Therefore, careful differential diagnosis is needed in the future. Some IPH reports have confirmed the impairment of restrictive ventilatory and pulmonary diffusing capacities on performing pulmonary function tests.⁵ Hemosiderin deposited in the interstitial space causes irreversible lung damage,¹⁶ which, in turn, is assumed to cause the impairment of restrictive ventilatory and pulmonary diffusing capacities. Our patient did not present with restrictive or obstructive ventilatory impairment, but presented with pulmonary diffusing capacity impairment and chronic respiratory failure. Generally, decreased pulmonary diffusing capacity does not cause respiratory failure; however, if extreme, it can lead to respiratory failure. In our case, irreversible lung damage developed long after DAH onset, and the patient's chronic respiratory failure may be attributed to a drastic decrease in pulmonary diffusing capacity. Although FVC_{90pred} and TLC_{90pred} significantly improved by the treatment, improvements in FEV₁ and DLCO_{90pred} were limited, and pulmonary diffusing capacity impairment persisted. IPH treatment with glucocorticoids and azathioprine might be difficult for the improvement of the decreased pulmonary diffusing capacity caused by irreversibly advanced lung damage. Limited number of reports exists on pulmonary function tests on adult-onset IPH, and further investigations are necessary.

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ORCID iD

Kotaro Iwasaki <http://orcid.org/0000-0003-0901-9966>

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