Original Article

Pulmonary siderosis cases diagnosed with minimally invasive surgical technique: A retrospective analysis of 7 cases

Erkan Akar, Tekin Yildiz[1], Safak Atahan[2]

Departments of Thoracic Surgery and [1]Chest Diseases, Bursa Yuksek Ihtisas Training and Research Hospital, [2]Department of Pathology, Faculty of Medicine, Biruni University, İstanbul, Turkey

Address for correspondence:
Dr. Erkan Akar, Department of Thoracic Surgery, Bursa Yuksek Ihtisas Training and Research Hospital, Bursa, Turkey.
E-mail: drerkanakar@hotmail.com

Submission: 16-05-2017
Accepted: 07-03-2018

Abstract:

OBJECTIVE: The aim of the present study was to report our patients with pulmonary siderosis (PS) who were exposed to iron oxide and diagnosed with the minimally invasive surgical technique (video-assisted thoracic surgery [VATS]), and to provide a discussion of the relevant literature.

METHODS: Hospital records of seven patients who were diagnosed with PS by VATS between 2008 and 2016 were retrospectively reviewed. VATS was performed for seven patients for whom no definitive diagnosis could be made with other diagnostic techniques.

RESULTS: All 7 patients included in our study were male, and their mean age was 54 years. As for the profession, two patients were founders, two were grinders, and three were welders. All patients were operated with VATS. The patients were followed with chest radiograms and spirometric tests for an average of 24 (15–36) months. At the follow-up, the patients were not only free of progression but also they even showed regression.

CONCLUSION: VATS is currently an established technique used for many diagnostic and therapeutic procedures, notably in chest surgery practice. We also advocate that VATS technique is an ideal method for making the pathological diagnosis of pneumoconioses when other methods fail to do so.

Keywords:

Diagnosis, minimally invasive surgery, pneumoconiosis, pulmonary siderosis

Pneumoconiosis is the general term used for pulmonary inorganic dust accumulation and tissue reactions developing against it. When there is a pure exposure to inert materials such as iron, antimony, barium, silver, tin, and cerium, cessation of exposure will usually reverse tissue reaction within 3–6 months, without resulting fibrosis. These types of pneumoconioses are termed as benign pneumoconioses.[1] Pulmonary siderosis (PS) is also benign pneumoconiosis that develops as a result of exposure to metallic iron or iron oxide dusts.[2,3] The occupations that lead to the development of PS include mining, welding, steel manufacturing, iron oxide manufacturing, grinding wheel manufacturing, and silver jewelry manufacturing.[2]

The study aimed to review our PS cases who were exposed to iron oxide dust and who were diagnosed with the help of a minimally invasive surgical technique and to provide a review of the relevant literature.

Methods

The hospital records of seven patients who were diagnosed with having PS with a minimally invasive technique between 2008 and 2016 were retrospectively reviewed.

Access this article online

Quick Response Code:

Website: www.thoracicmedicine.org

DOI: 10.4103/atm.ATM_152_17

How to cite this article: Akar E, Yildiz T, Atahan S. Pulmonary siderosis cases diagnosed with minimally invasive surgical technique: A retrospective analysis of 7 cases. Ann Thorac Med 2018;13:163-7.

Data regarding age, sex, occupation, admission complaint, family history of a genetic disorder, and smoking status were explored. All patients underwent posterolateral chest X-ray imaging and thoracic computerized tomographic (CT) imaging. They also underwent respiratory function testing and blood biochemistry and complete blood count analyses. Flexible bronchoscopy and bronchoalveolar lavage were carried out for making a pathological diagnosis. Video-assisted thoracic surgery (VATS) was carried out for seven patients for whom no definitive diagnosis could be made with other methods. This retrospective study was approved by the Institutional Ethics Committee, and the requirement for informed consent was waived because of the retrospective nature.

Operative technique

All patients were operated under general anesthesia at the operating room. After placing patients on the operating table and inducing anesthesia, they were intubated using a double lumen tube. They were placed on the operating table as if a standard posterolateral thoracotomy incision would be made, and they were scrubbed. In lateral decubitus position, the arm was suspended in abduction so that the scapula was maximally elevated. The table was tilted, and a 10 cm high roll pillow was placed under the thoracic cage to maximally expose intercostal spaces. Using a single incision and 0° telescope (Karl Storz, Hopkins II telescopes diameter 5 mm, length 29 cm trocar size 6 mm) imaging, a biopsy sample (Echelon Flex 60 mm Endo Stapler 4.1 mm) was taken with wedge resection from the site where the lung lesion was located. Biopsy samples were taken from two separate sites in patients with bilateral diffuse lung lesions. Entire pleura and pulmonary surfaces were reviewed after the operation. After performing a check for air leaks and hemostasis, a 28F chest tube was placed through the incision site. All patients were extubated at the operating table and transferred to the regular ward.

Results

All 7 patients were male, and their mean age was 54 years. The admission complaints were a cough and chest pain in two patients, whereas five were asymptomatic. Four patients were active smokers for 15 years. Two patients were founders, two were grinders, and 3 were welders; and they had been working for an average of 11 (8–15) years [Table 1]. The respiratory system examination of two patients showed minimal crepitation in the basal parts of both lungs at the end of expiration, but the examination of other systems was normal. The spirometric examination revealed modest obstructive defects in two patients. In the conventional radiograms of the chest, the most common pathological sign was diffuse, widespread reticulonodular opacities [Figure 1]. Thoracic CT revealed multiple small nodular opacities, honeycomb appearances, and patchy ground-glass opacities, usually confined to middle-upper zones [Figure 2]. The fibrotic changes that we detected in two patients were mostly in the lower zones of the lungs. All patients who underwent the procedure for diagnostic purposes formerly underwent transbronchial biopsy and lavage at the Chest Diseases Department, but could not be diagnosed. Having been referred to our clinic, these patients were operated with the VATS technique. Intraoperatively, entire lung surface was found to be covered by a black anthracotic material [Figure 3]. The pathological examination of the tissue samples taken with wedge resection revealed hemosiderin-laden macrophages that were positively stained with Perl's Prussian blue, a finding which was consistent with siderosis [Figure 4]. Three patients were found to have elevated serum iron and ferritin levels when studied after obtaining the pathological diagnosis. The time to chest tube removal was 2 (1–4) days on an average. No postoperative complication or death occurred. The patients were followed for an average of 24 (15–36) months with chest radiograms and spirometry tests. During the follow-up, the patients were not only free of progression but also they even showed regression during that time.

Table 1: Demographic characteristics of patients

Variables	Number of patients, *n* (%)
Gender	
Female	0
Male	7 (100)
Average age	54±0.21
Profession	
Founder	2 (29)
Grinder	2 (29)
Welder	3 (42)
Smoking history	4 (57)
Leading symptoms	
Cough and chest pain	2 (29)
No symptom	5 (71)
Accompanying diseases	
COPD	2 (50)
Diabetes	1 (25)
Hypertension	1 (25)
Radiological findings	
Upper lobe	5 (71)
Upper + lower lobe	2 (29)

COPD=Chronic obstructive pulmonary disease

Discussion

Heavy inhalation of iron compounds is common in mining, foundry, steel manufacturing, iron oxide manufacturing, grinding wheel production, and jewelry. Welder's siderosis was first defined by Doig

Figure 1: Posteroanterior chest X-ray most commonly showed diffuse, widespread reticulonodular opacities

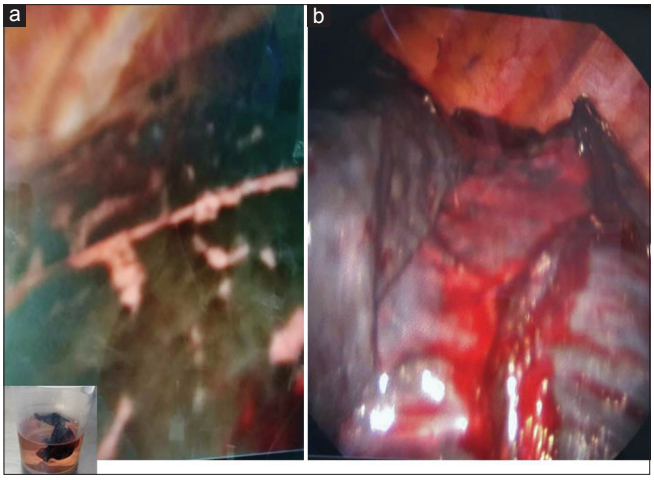

Figure 3: Intraoperatively, entire lung surface was found to be covered by a black anthracotic material (a,b)

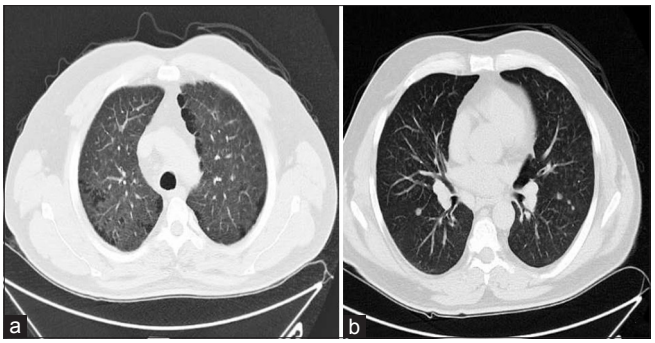

Figure 2: Thoracic computerized tomography revealed multiple small nodular opacities, honeycomb appearances (a), and patchy ground-glass opacities (b), which were usually confined to middle-upper zones

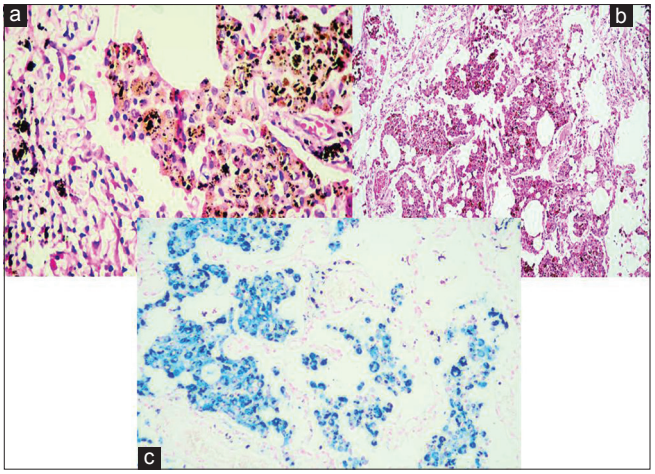

Figure 4: (a) There are hemosiderin-laden macrophages in the alveolar spaces. (H and E, ×40) (b) In the alveolar spaces are hemosiderin-laden macrophages. Anthracosis pigments are in the septa. There is no evidence of inflammation (H and E, ×200,). (c) Staining for iron demonstrates alveoli filled with hemosiderin-laden macrophages. (×100, Perls' Prussian blue)

and McLaughlin[4] in 1936. They prospectively assessed 16 welders with respect to their clinical examination findings and radiological properties. After a 9-year follow-up of 15 cases, they showed the absence of symptoms, and they were even able to reveal that the disease did not progress. Since then, the debate has begun whether siderosis is a benign pneumoconiosis since fibrosis could not be definitively shown in the pathological examination of the lung.[5] Buckell and Garrad,[6] in 1946, reviewed siderosis and asserted that fibrosis may actually exist in these lesions. Further, the existence of fibrosis was reported by subsequent studies in multiple case series with spirometric defects.[7-9]

Long-term pure iron oxide exposure has no known detrimental effect with permanent functional impairment. However, symptomatic interstitial fibrosis may occur, although rarely, when persons exposed to iron dust or smoke are also simultaneously exposed to other silicates, notably silica, and asbestos.[10] In this study, we revealed minimal fibrosis in two cases. These patients were welders. However, even though we could not demonstrate any exposure to silicates pathologically, we believe that fibrosis is still possible given that dust from many compounds pollute working places during welding. There may be no symptoms at all in PS, but affected patients may also present with a cough, sputum, and effort dyspnea, all depending on exposure time and any lesion developed in response.[11]

Two of our patients who were smokers had cough and chest pain. Given that siderosis usually elicits no symptoms at all, those complaints may be attributed to smoking.

The main diagnostic tests to make a diagnosis of PS include PA chest X-Ray, high-resolution CT, and thoracic CT. PA chest X-Ray is characterized by bilateral diffuse reticulonodular opacities.[12] In a study performed by Akira in 1995,[13] thoracic CT findings of 21 welders mainly were mainly micronodules in the centrilobular

region. This finding was subsequently corroborated by others.[14] However, studies, later on, revealed that lesions of siderosis were mainly concentrated in upper lobes.[15] Since siderosis is a benign form of pneumoconiosis, unlike other collagen pneumoconioses, its radiological signs may partially or near completely reverse after exposure is eliminated. Respiratory function tests are the second most widely used tests after radiological examinations used for evaluating occupational diseases. Although respiratory function tests are nonspecific occupational diseases, they are valuable for assessing dyspnea, distinguishing obstructive and restrictive disorders, and making a clinical evaluation of the disease.[16] Whereas exposure to iron dust is known to rarely cause obstructive airway disease, smoking further increases such risk.[7-9,17] In conclusion, we are of the opinion that obstructive disease that occurred in two patients was a consequence of the combined effect of smoking and iron dust inhalation.

To diagnose diffuse lung disorders of different etiology, one of the invasive techniques should be selected, such as transbronchial biopsy and lung biopsy. The use of VATS has been currently increasing in every field of medicine. It has been used as an established technique for many diagnostic and therapeutic procedures.[18] Some authors recommend using thoracotomy and thoracoscopy to diagnose PS cases that cannot be diagnosed pathologically with transbronchial biopsy.[19,20] Ferguson,[21] Mack *et al*.[22] advocated the use of thoracoscopic biopsy to diagnose diffuse lung disorders, and stressed that with this technique diagnostic accuracy reached 100%. Thoracoscopic wedge resection is a reliable tool for the diagnosis of infiltrative and interstitial parenchymal diseases.[21,22] Moreover, biopsy sampling with this approach is simple and highly successful because plaural and parenchyma can be adequately visualized and the best site for biopsy sampling can be determined. In addition, postoperative pain is less severe with this approach.

The most striking feature of pathology examination of PS is the presence of hemosiderin-laden macrophages in the peribronchovascular space and interstitium. In pure siderosis, fibrosis is either completely absent or minimal.[19] Alveolar structures are spared; the stromal reaction is minimal, and it primarily consists of reticulin fibers. Iron granules stained by Prussian blue are also visualized. The reaction is potentially reversible.[1]

Ferritin is the main iron storage protein, and it is a sensitive indicator of total iron depots in the absence of systemic inflammatory conditions.[23] As an inflammatory marker, ferritin has been shown to increase in various chronic lung diseases.[24,25] However, ferritin level in PS is substantially higher than those encountered in inflammatory lung disease.[24] Serum ferritin level increases parallel to bronchoalveolar lavage fluid level.[26] We determined significant elevation in serum ferritin levels of three patients although we failed to reveal the same elevation in bronchoalveolar lavage fluid.

Excessive iron deposits are seen in heterozygous hemochromatosis with H63D mutation, persons undergoing multiple blood transfusions, and persons with thalassemia and sideroblastic anemia. Nevertheless, this accumulation is of a lower extent than that seen with PS.[27] Our extensive review of past histories and laboratory investigations of the patients revealed no such conditions.

Conclusion

PS is benign pneumoconiosis that develops as a result of the exposure to metallic iron or iron oxide dust. One of the available invasive techniques such as transbronchial biopsy or lung biopsy may be selected to make the diagnosis of PS. We also advocate that VATS is an ideal technique for making the pathological diagnosis of pneumoconioses when other methods fail to do so, because this technique is characterized by a simpler biopsy sampling, a better visualization of pleura and lung parenchyma, an ability to spot the best site for biopsy sampling, and a higher success rate.

Financial support and sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest.

References

1. Şimşek C. Lung diseases related to toxic inhalation. Clinical progress, special issue for occupational diseases. Clin Dev Occup Dis Spec Number0 2011;23:71-8.
2. Gothi D, Satija B, Kumar S, Kaur O. Interstitial lung disease due to siderosis in a lathe machine worker. Indian J Chest Dis Allied Sci 2015;57:35-7.
3. Doherty MJ, Healy M, Richardson SG, Fisher NC. Total body iron overload in welder's siderosis. Occup Environ Med 2004;61:82-5.
4. Doig AT, McLaughlin AI. X-ray appearance of the lungs of electric arc welders. Lancet 1936;1:771-5.
5. Doig AT, McLaughlin AI. Clearing of X-ray shadows in welders' siderosis. Lancet 1948;1:789-91.
6. Buckell M, Garrad J. The incidence of siderosis in iron turners and grinders. Br J Ind Med 1946;3:78-82.
7. Skloot G, Goldman M, Fischler D, Goldman C, Schechter C, Levin S, *et al*. Respiratory symptoms and physiologic assessment of ironworkers at the world trade center disaster site. Chest 2004;125:1248-55.
8. Johnson A, Moira CY, MacLean L, Atkins E, Dybuncio A, Cheng F, *et al*. Respiratory abnormalities among workers in an iron and steel foundry. Br J Ind Med 1985;42:94-100.
9. Gomes J, Lloyd OL, Norman NJ, Pahwa P. Dust exposure and impairment of lung function at a small iron foundry in a rapidly developing country. Occup Environ Med 2001;58:656-62.

10. Ji C, Chen G, Cai HR, Meng FQ, Chen YB, Guo LC, *et al.* An unusual case of welder's siderosis with local massive fibrosis: A case report. Chin Med J (Engl) 2012;125:552-4.
11. Khalid I, Khalid TJ, Jennings JH. A welder with pneumosiderosis: A case report. Cases J 2009;2:6639.
12. Morgan WK. Other pneumoconiosis. In: Morgan WK, Seaton A, editors. Occupational Lung Diseases. Pennsylvania: WB Saunders; 1995. p. 407-56.
13. Akira M. Uncommon pneumoconioses: CT and pathologic findings. Radiology 1995;197:403-9.
14. Fidan F, Esme H, Unlu M, Acar M, Albayrak R, Dilek FH, *et al.* Welder's lung associated with pneumothorax. J Thorac Imaging 2005;20:120-2.
15. Glazer CS, Newman LS. Occupational interstitial lung disease. Clin Chest Med 2004;25:467-78, vi.
16. Gulati M, Redlich CA. Occupational lung disorders: General principles and approaches. In: Fishman AP, editor. Fishman's Pulmonary Diseases and Disorders. 4th ed. New York: McGraw Hill; 2008. p. 933-42.
17. Kuo HW, Chang CL, Liang WM, Chung BC. Respiratory abnormalities among male foundry workers in central Taiwan. Occup Med (Lond) 1999;49:499-505.
18. Handy JR Jr., Asaph JW, Douville EC, Ott GY, Grunkemeier GL, Wu Y, *et al.* Does video-assisted thoracoscopic lobectomy for lung cancer provide improved functional outcomes compared with open lobectomy? Eur J Cardiothorac Surg 2010;37:451-5.
19. McCormick LM, Goddard M, Mahadeva R. Pulmonary fibrosis secondary to siderosis causing symptomatic respiratory disease: A case report. J Med Case Rep 2008;2:257.
20. Şimşek C, Kalaycıoğlu O, Beder S, Tunç M, Evirgen O, Ertürk Ş. Welder's siderosis and pulmonary alveoler proteinosis (A case report). Turkiye Klin J Med Sci 1992;12:250-3.
21. Ferguson MK. Thoracoscopy for diagnosis of diffuse lung disease. Ann Thorac Surg 1993;56:694-6.
22. Mack MJ, Aranoff RJ, Akuff TE, Douthit MB, Bowman RT, Ryan WH. Present role of thoracoscopy in the diagnosis and treatment of disease of the chest. Ann Thorac Surg 1992;54:403-9.
23. Prieto J, Barry M, Sherlock S. Serum ferritin in patients with iron overload and with acute and chronic liver diseases. Gastroenterology 1975;68:525-33.
24. Schneerson JM, Jones BM. Ferritin, finger clubbing and lung disease. Thorax 1981;36:688-92.
25. Kell DB, Pretorius E. Serum ferritin is an important inflammatory disease marker, as it is mainly a leakage product from damaged cells. Metallomics 2014;6:748-73.
26. Yoshii C, Matsuyama T, Takazawa A, Ito T, Yatera K, Hayashi T, *et al.* Welder's pneumoconiosis: Diagnostic usefulness of high-resolution computed tomography and ferritin determinations in bronchoalveolar lavage fluid. Intern Med 2002;41:1111-7.
27. Bulaj ZJ, Griffen JM, Jorde JB, Edwards CQ, Kushner JP. Clinical and biochemical abnormalities in people heterozygous for haemochromatosis. N Engl J Med 1996;335:1799-805.