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A RARE CASE OF FUNGAL BALL IN GRANULOMATOUS POLYANGITIS

Medicine

Dr. Astha. A. Jha 3rd Year Resident, Department of Medicine, Smt. N.H.L. Medical College, Ahmedabad

Dr. Bhargav H. Patel* 3rd Year Resident, Department of Medicine, Smt. N.H.L. Medical College, Ahmedabad *Corresponding Author

Dr. Jayesh J. Dutt Professor and Head of Unit, Department of Medicine, Smt. N.H.L. Medical College, Ahmedabad

Dr. Khushali L. Patel Associate Professor, Department of Medicine, Smt. N.H.L. Medical College, Ahmedabad

Dr. Jaydev S. Mod Assistant Professor, Department of Medicine, Smt. N.H.L. Medical College, Ahmedabad

ABSTRACT

Granulomatous polyangitis is a rare autoimmune disorder causing vasculitis of small vessels. It usually presents with upper and lower respiratory with renal involvement and other systemic manifestation such as uveitis. This is a case of 42year old female came with complains of low grade fever since 8 months with chronic cough and bilateral red eye since 6 months. After ruling out systemic infection like tuberculosis patient was considered to have Granulomatous polyangitis in view of a systemic complains with laboratory evidence. Patient was started on systemic steroids for the same which subsided the symptoms except cough which didn't resolve. HRCT was done for the same which revealed fungal ball and culture of sputum KOH mount for the same showed few branched septate fungus with positive aspergillus antigen. Steroids were withheld in view of acute fungal infection and was discharged on antifungals for the same. Patient improved with antifungals , however was lost to follow up in subsequent visits.

KEYWORDS

INTRODUCTION:

Granulomatosis with polyangiitis (formerly Wegner's granulomatosis) is a small-to-medium vessel vasculitis which affects multiple organs. Pathologically, there is a focal necrotizing vasculitis involving the small arteries and veins of the ear, nose, and throat, the lungs, the kidneys, the heart, the nervous system, the eyes, the skin, and, infrequently, other organs [1–3]. The term mycetoma is used to describe a mobile fungus ball within a pre existing, usually fibrotic, lung cavity. Mycetomas are usually caused by Aspergillus (aspergilloma). This is a very interesting case of Granulomatosis with Polyangiitis (GPA) presenting with chronic cough and bilateral red eye. Following treatment with steroids the symptoms regressed however she developed a fungal infection due to immunosuppression by steriods.

CASE VIGNETTE:

A 42 year old female came with complain of Low grade fever since 8 months and dry cough since 7 months.

She also had complaint of bilateral eye pain, excessive lacrimation and visual blurring since 5 months

The patient was relatively asymptomatic before 8months. Then she developed low grade fever with chills, without rigors, daily, usually evening rise, which was relieved on medication. She showed to some private practitioner for the same, where she was diagnosed as viral fever, and was treated for the same. Her CBC at that time (10/02/18) showed Hemoglobin- 9.7, Total count 12,000 and Platelet count of 2.04 lacs. ESR was 126mm.

She had persistent fever, along with which she also developed dry cough. Cough was non productive, not associated with change in posture or time of the day. She consulted a physician for the same. At that time she had Reduced air entry and crepitations, based on which a Chest X ray was done, which showed, Bilateral Lower zone consolidation. She was treated as Pneumonia during that time. But she did not improve for 2 months.

Therefore, further investigations were done. An HRCT was done, which was suggestive of diffuse areas of consolidation with bilateral upper and lower lobe with few areas of cavitation. Also there were multiple centrilobular opacities, involving bilateral upper and lower lobe, with tree in bud appearance. These findings were interpreted as Pulmonary tuberculosis by the radiologist.

In order to confirm pulmonary kochs, a sputum sample was sent, which did not show presence of acid fast bacilli. But as patient was not improving on antibiotics, and patient had an ESR of 166, she was empirically started on AKT. Her symptoms did not reduce, also she developed redness, photophobia, pain and lacrimation in the right eye.She consulted an ophthalmologist for the same, and she was given some medication during that period, but no documents are available.After 15 days, she developed similar complains in the other eye also.She then showed to another ophthalmologist, who kept the impression of corneal ulcer in the right eye with progressive thinning of margins. Left eye was normal. She was started on systemic as well as topical antibiotics and symptomatic treatment.

Since May 2018 she was on oral Tab Prednisolone 8mg. after starting steroids, her condition improved considerably. Her systemic symptoms resolved, but her ocular symptoms persisted. She also developed loss of vision in both the eyes.

She then consulted a cornea specialist, where she was diagnosed bilateral peripheral ulcerative keratitis. Patient was referred to a rheumatologist as she had systemic symptoms along with bilateral peripheral ulcerative keratitis.

Patient consulted a rheumatololgist on 25th October 2018, with complete loss of vision in the right and and partial loss of vision in the left eye. There was no ear discharge, nasal discharge, fever, breathlessness, epistaxis, musculoskeletal complains. Her systemic examination was normal. Respiratory and CNS examination was normal. Her reports done during that time showed:

Haemoglobin	10	Creatinine	0.63
Total count	13,840	Potassium	3.5
N/L	11230/1610	Calcium	8.6
Platelets	2.05 lacs	SGPT	28
PS	Microcytic, hypochromic RBCs	ANCA by IF	++++ (showing cANCA pattern)
ESR	126	PR3 by ELSIA	100 (0-10)
CRP	18.0	2D ECHO	Normal

In view of high ESR, history of respiratory involvement along with bilateral peripheral ulcerative keratitis, response to steroids and ANCA & PR3 positivity she was considered to have Granulomatous polyangitis.

Patient was then given a pulse of 500mg methylprednisolone for 7 days. Patient was planned to start cyclophosphamide.She then developed fungal infection within the lung cavity, thus cyclophosphamide was not given.HRCT done during that time (30/10/18) showed cavitatory changes with fungal ball noted within. In view of the fungal infection, immunosuppression was stopped and Bronchoalveolar lavage was done. The sample for then sent for culture and routine examination.BAL fluid showed fungal hyphae. So aspergillus antigen was sent which came back positive. Antifungals were added according to sensitivity.On follow up the cavity size reduced with improvement in the respiratory symptoms. Patient was planned to start on immunosupressives again, but she was then lost to follow up.

DISCUSSION:

Granulomatosis with polyangiitis (formerly Wegner's granulomatosis) is a small-to-medium vessel vasculitis which affects multiple organs. Pathologically, there is a focal necrotizing vasculitis involving the small arteries and veins of the ear, nose, and throat, the lungs, the kidneys, the heart, the nervous system, the eyes, the skin, and, infrequently, other organs [1–3]. The term mycetoma is used to describe a mobile fungus ball within a preexisting, usually fibrotic, lung cavity. Mycetomas are usually caused by Aspergillus (aspergilloma). The cavity wall has a rich blood supply from the bronchial and other branches of the systemic circulation, and thus has a propensity to bleed. Pulmonary aspergillomas clinically present as chronic productive cough or hemoptysis, which can be life threatening. Pulmonary aspergillomas have been identified in association with a wide variety of lung diseases, including tuberculosis, sarcoidosis, asbestosis, histoplasmosis, ankylosing spondylitis, bronchiectasis, bronchial cysts and malignancy. The presence of a pre-existing lung cavity appears to be the most common predisposing factor to the formation of an aspergilloma (Pennington, 1980). In Wegener's granulomatosis, excavation of the lung infiltrates occurs in roughly 50% of cases (Fauci and Wolff, 1973). Unless complete resolution is achieved, vegetation of Aspergillus in these cavities may occur, as illustrated in this case. The development of an aspergilloma, causing haemoptysis and changing the radiological appearance of the lung infiltrate, should not be confused with disease activity of Wegener's granulomatosis. Surgical resection of an aspergilloma is the only certain method of cure. [4] Such opportunistic infections following immunosuppression warrants strict maintainance of asepsis and regular screening for the same.

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