CASE REPORT

Strongyloidiasis: an unusual cause of septic shock with pneumonia and enteropathy in western countries

Florent Montini,¹ Frederic Grenouillet,² Gilles Capellier,^{1,3} Gaël Piton^{1,3}

¹Intensive Care Unit, University Hospital of Besançon, Besançon, France

²Parasitology Unit, University Hospital of Besançon, Besançon, France

³EA3920 and SFR FED 4234

Correspondence to

Dr Gaël Piton,
piton.gael@wanadoo.fr

Accepted 2 March 2015

SUMMARY

We report a case of invasive strongyloidiasis in a patient from the French Antilles who had been living in France for many years, with no history of immunosuppression, and who was hospitalised in the intensive care unit for septic shock with multimicrobial hypoxaemia pneumonia and exudative enteropathy. Initiation of systemic corticosteroid therapy for septic shock seems to have precipitated onset of the parasitic infection, with recurrence of hypoxaemic pneumonia complicated by hypoxic cardiac arrest. The diagnosis was confirmed after roundworm larvae were found on bronchoalveolar lavage. Treatment with ivermectin was initiated, but the patient died in a context of postanoxic encephalopathy.

BACKGROUND

A diagnosis of invasive strongyloidiasis is rare in western European countries, but should be considered in patients originating from tropical countries, even a long time after travel to zones at risk, when they present with severe sepsis and both respiratory and intestinal symptoms, particularly when multiple germs are identified. Given that systemic corticosteroid therapy is the main risk factor for progression to disseminated disease, whose prognosis is dismal, any possibility of a diagnosis of strongyloidiasis should incite physicians to withhold corticosteroids until parasitic infection is ruled out.

CASE PRESENTATION

A 57-year-old man was treated at his home by the mobile emergency medical unit for problems of consciousness associated with a state of shock. His only antecedents were active smoking, estimated at 30 pack-years, and alcohol poisoning. He lived alone as a recluse, and had had no contact with his family for many years. Clinical examination revealed blood pressure at 70/30 mm Hg, a heart rate of 100 bpm, SpO₂ at 92% in room air and a Glasgow score of 6/15 without signs of focalisation. There was also diffuse oedema of the lower limbs and lumbar region. Lastly, there were signs of profuse diarrhoea and vomiting around the patient. Vascular filling was initiated and continued until his admission to the intensive care unit of our hospital, where he was treated by norepinephrine for a shock state that appeared to be septic. Despite restoring haemodynamic status, the consciousness disorders persisted and the patient was intubated. Biological tests showed very low total blood protein, at 33 g/L, and severe hypoalbuminaemia at

7 g/L. A cerebral CT scan without injection and a lumbar puncture were performed to investigate causes of the coma, but both were normal.

Chest X-ray showed pneumonia in the lower right lobe. The pneumococcal urinary antigen test was positive, and we retained a diagnosis of septic shock with pneumonia caused by pneumococcus. Antibiotic therapy (ceftriaxone and metronidazole) was initiated, in view of the vomiting and the high likelihood of aspiration. We also initiated treatment with hydrocortisone hemisuccinate (100 mg bolus followed by 200 mg/day) for the septic shock, and high doses of norepinephrine were required. In terms of bacteriological examinations, a blood culture performed at admission was positive for *Proteus mirabilis*, two other blood cultures were positive for drug-susceptible *Staphylococcus epidermidis*, and tracheal bronchoaspiration was positive for two additional germs, namely *Escherichia coli* and *Haemophilus influenzae*, both drug-susceptible. Transoesophageal echography did not find any signs of infectious endocarditis. The patient's clinical course was favourable under antibiotic therapy, and the patient was extubated a few days after admission. His state of consciousness was normal. However, he had persistent diarrhoea. Stool tests to investigate the presence of *Clostridium difficile* came back negative. Two days after extubation, the patient again presented with pneumonia that caused complete atelectasis of the right lung, complicated by hypoxic cardiac arrest with asystole. Cardiopulmonary resuscitation lasting 25 min was necessary. Further bacteriological tests revealed a blood culture positive for *Enterococcus faecalis* and tracheal bronchoaspiration positive for *Myroides odoratimimus*, a telluric germ that is not normally pathogenic. In view of the multiplicity of germs and the atypical nature of the last germ identified, bronchoalveolar lavage was performed, and revealed larvae compatible with strongyloides (figure 1). Stool testing also revealed the presence of high numbers of *Strongyloides* larvae. The diagnosis of disseminated strongyloidiasis was confirmed and treatment with ivermectin was initiated. Since the patient failed to regain consciousness after the cardiac arrest, we ruled out neuromeningeal localisation of the infection by performing lumbar puncture, which showed that the spinal fluid contained no bacteria and no *Strongyloides* larvae. Neurological testing confirmed severe postanoxic encephalopathy further to the hypoxic cardiac arrest. The patient died 2 weeks later.

CrossMark

To cite: Montini F, Grenouillet F, Capellier G, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2014-209028

Figure 1 Direct examination of bronchoalveolar lavage fluid showed *Strongyloides* larvae (May Grunwald Giemsa staining). Note the size of the larva in comparison with the polynuclear neutrophils in the microscopic field.

OUTCOME AND FOLLOW-UP

Death after a follow-up of 15 days.

DISCUSSION

We report a case of disseminated strongyloidiasis at the origin of septic shock with recurring bacterial pneumonia caused by multiple germs, with exudative enteropathy, in a patient from the French Antilles.

Strongyloidiasis is caused by a roundworm, of which two varieties can infect humans, namely *Strongyloides stercoralis* and *Strongyloides fullborni*. It is a tropical disease that mainly affects sub-Saharan Africa, the Antilles, Central and South America, the Indian Ocean and South-East Asia.¹ Humans can be infected by larvae in soil polluted with faecal material, mainly by penetration of the larvae through the skin. Mucous and sexual contamination is also possible. The larvae are transported in the blood and lymphatic circulation to the right heart and the lungs, where they ascend the pulmonary tree and are swallowed, thereby reaching the digestive tract. Some larvae are excreted, but others can infest the intestinal wall and mature in the intestine, perpetuating the parasitic cycle. Indeed, this cycle explains the durable nature of strongyloidiasis. The presence of immunodepression, or treatment with corticosteroids, precipitates an acceleration of the cycle of reinfection, leading to clinical evidence of disseminated disease. Symptoms in non-immunodepressed patients are mainly digestive (eg, diarrhoea, malabsorption), cutaneous (rash, larva currens) or rheumatismal. At the larval migration phase, Loeffler's syndrome may be observed, with pulmonary symptoms, such as coughing and asthma-like dyspnoea, as well as eosinophils. In immunodepressed patients, particularly in patients treated with corticosteroids or in those infected with the human T-cell leukaemia virus type 1, disseminated strongyloidiasis may lead to multiorgan failure, with the presence of larvae in all organs.² Dissemination of the larvae can be complicated by secondary pulmonary infections and neuromeningeal infection with microbes originating from the digestive tract.^{3 4}

The diagnosis of strongyloidiasis was confirmed rather late in our patient. The clinical presentation was misleading, since the digestive symptoms were not particularly salient at admission, contrary to the respiratory symptoms, which were predominant. In addition, strongyloidiasis is a rare disease in western countries, which could explain why this diagnosis is not

systematically considered. Nevertheless, several elements could have led us to this diagnosis sooner. First, since strongyloidiasis is a common infection in the French Antilles, the patient's origin was suggestive of this diagnosis. Second, the association of diarrhoea, oedema and severe hypoalbuminaemia at admission could have suggested malabsorption by exudative enteropathy, for which strongyloidiasis is one of the main aetiologies. Disseminated strongyloidiasis leads to loss of function of the digestive and pulmonary barriers, explaining the multiple germs found, and their pulmonary (*Streptococcus pneumoniae*, *H. influenzae*) and digestive (*E coli*, *Enterococcus faecalis*) nature. Lastly, the atypical nature of the last germ identified by tracheal bronchoaspiration prompted the physicians to prescribe bronchoalveolar lavage, where the larvae were clearly visible, thus leading to the definitive diagnosis of disseminated strongyloidiasis infection (figure 1).⁵ Blood tests are also an integral component of the standard diagnostic workup but are no longer especially necessary once the parasite has been isolated and identified on direct examination.

Regarding the risk factors for strongyloidiasis, our patient had not travelled to the French Antilles for many years, which might have led us to think that a diagnosis of strongyloidiasis was unlikely. However, reinfection is always possible, even many years after initial contamination, and a transitory impairment of immune capacity can trigger the transition from isolated latent infection of the digestive tract to disseminated disease. Given that our patient lived alone as a recluse, the digestive symptoms had most likely begun several weeks before for him to have reached such a severe level of hypoproteidaemia. Apart from the patient's malnourished status and chronic alcoholism, we did not find any other immunodepressive factors prior to admission in critical care. Indeed, the patient was HIV-negative and did not have cancer or any immunosuppressive treatment. Our patient initially started to recover, but suffered sudden and drastic clinical deterioration a few days after the introduction of hydrocortisone hemisuccinate at 200 mg/day. The patient's status deteriorated rapidly, and he presented with concomitant bacterial infection and pneumonia due to *M. odoratimimus*, which is a naturally resistant microbe that is only susceptible to cefepime and carbapenems.⁶ Hydrocortisone hemisuccinate is recommended for the treatment of septic shock with high doses of catecholamines. However, systemic corticosteroid therapy is clearly identified as the main risk factor for disseminated strongyloidiasis. Another point to be considered is that septic shock itself can lead to an immunocompromised state, in particular because of the development of compensatory anti-inflammatory response syndrome. In this context, there are changes in the host innate and adaptive immunity consistent with immunosuppression.⁷

Mortality in patients presenting with disseminated strongyloidiasis is high, at around 70%.⁸ This excess mortality is most likely multifactorial in origin. First, the infection itself is serious, and the diffuse infiltration of the organs by the larvae can cause multiorgan failure. Furthermore, loss of function of the digestive and pulmonary barriers perpetuates the sepsis by continuously providing new microbes. In addition, delayed diagnosis is likely, especially in countries where the prevalence is low and infection with strongyloides is not systematically considered, and this delay may have deleterious consequences. Lastly, in the more severe forms with diffuse intestinal disease, oral administration of ivermectin can be inefficacious, since malabsorption precludes sufficient bioavailability of the drug to have any therapeutic effect.⁹ In critical care, administration by the intravenous route is preferable.¹⁰

Learning points

- We report a case of disseminated strongyloidiasis responsible for septic shock, with recurrent bacterial pneumonia and exudative enteropathy.
- This diagnosis, which is rare in western countries and rarely reported in critical care,¹¹ should be considered in patients from tropical countries presenting with severe sepsis and both respiratory and digestive disease, even long after any travel in at-risk areas.
- Suspicion of strongyloidiasis should be especially high if bacteriological tests reveal multiple germs, reflecting the loss of function of digestive and pulmonary barriers.
- In such cases, systemic corticosteroid therapy should be avoided until the diagnosis of strongyloidiasis is definitively ruled out.

Acknowledgements The authors thank Fiona Ecarnot (EA3920, University hospital Besancon, France) for translation and editorial assistance.

Contributors FM and GP drafted the manuscript. FG identified the disease by direct examination of bronchoalveolar lavage. FM, FG, GC and GP have reviewed the manuscript and consistently improved its content.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- Montes M, Sawhney C, Barros N. Strongyloides stercoralis: there but not seen. *Curr Opin Infect Dis* 2010;23:500–4.
- Basile A, Simzar S, Bentow J, et al. Disseminated Strongyloides stercoralis: hyperinfection during medical immunosuppression. *J Am Acad Dermatol* 2010;63:896–902.
- Cataño JC, Pinzon MA. Strongyloides pneumonia. *Am J Trop Med Hyg* 2012;87:195.
- Vandebosch S, Mana F, Goossens A, et al. Strongyloides Stercoralis infection associated with repetitive bacterial meningitis and SIADH: a case report. *Acta Gastroenterol Belg* 2008;71:413–17.
- Apewokin S, Steciuk M, Griffin S, et al. Strongyloides hyperinfection diagnosed by bronchoalveolar lavage in an immunocompromised host. *Cytopathology* 2010;21:345–7.
- Benedetti P, Rassu M, Pavan G, et al. Septic shock, pneumonia, and soft tissue infection due to Myroides odoratimimus: report of a case and review of Myroides infections. *Infection* 2011;39:161–5.
- Boomer JS, To K, Chang KC, et al. Immunosuppression in patients who die of sepsis in multiple organ failure. *JAMA* 2011;306:2594–605.
- Lim S, Katz K, Krajden S, et al. Complicated and fatal Strongyloides infection in Canadians: risk factors, diagnosis and management. *CMAJ* 2004;171:479–84.
- Segarra-Newnham M. Manifestations, diagnosis, and treatment of Strongyloides stercoralis infection. *Ann Pharmacother* 2007;41:1992–2001.
- Donadello K, Cristallini S, Taccone FS, et al. Strongyloides disseminated infection successfully treated with parenteral ivermectin: case report with drug concentration measurements and review of the literature. *Int J Antimicrob Agents* 2013;42:580–3.
- de Boer MP, Bethlem-Schaap C, de Jager S, et al. Fatal strongyloidiasis in an immunocompromised HTLV-I infected patient. *Intensive Care Med* 2013;39:339–40.

Copyright 2015 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- Submit as many cases as you like
- Enjoy fast sympathetic peer review and rapid publication of accepted articles
- Access all the published articles
- Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow