Tracheo-oesophageal fistula in highly active antiretroviral therapy patient with AIDS

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Abstract

Introduction
To the best of our knowledge, other than tuberculosis, tracheo-oesophageal fistula complicates oesophageal ulcers and has been rarely reported. Tracheo-oesophageal fistula, secondary to AIDS-defining infectious diseases and neoplasia, is regressing since the introduction of highly active antiretroviral therapy. It occurs as a complication of tuberculosis of the digestive tract and airways. Other infections causing deep oesophageal ulcers include cytomegalovirus, herpes simplex virus type 2 and HIV. Several studies have reported resistances of HIV1 to many antiretroviral drugs, making the occurrence of opportunistic gastrointestinal disease possible in patients treated with such drugs, particularly in the severely immunodepressed. The outcome is generally poor in the absence of treatment with an average survival rate of one to six weeks. This paper reports a case of tracheo-oesophageal fistula in a highly active antiretroviral therapy patient with AIDS.

Case report
We are reporting the case of a 43-year-old Cameroonian man, who was type 1 HIV-infected, classified CDC stage C3. He has been on treatment for about 10 years and on combination therapy for about six months. He presented with cough during swallowing, odynophagia and weight loss, complicating a medically treated tracheo-oesophageal fistula, whose outcome was poor.

Conclusion
Despite the various antiretroviral regimens available in developing countries, HIV-related digestive diseases remain a challenge with poor prognosis due to other limited therapeutic options. The diagnosis of tracheo-oesophageal fistula should be considered in the context of persistent cough during swallowing in patients with AIDS.

Introduction
Tracheo-oesophageal fistula, secondary to AIDS-defining infectious diseases and neoplasia, is regressing since the advent of highly active antiretroviral therapy (HAART). In some rare instances, it occurs as a complication of tuberculosis (TB) of the digestive tract and airways. Other infections causing deep oesophageal ulcers include cytomegalovirus (CMV), herpes simplex virus (HSV) type 2 and HIV. The diagnosis of tracheo-oesophageal fistula is generally considered in the setting of persistent cough during swallowing. Endoscopy and histopathologic analysis in the majority of cases enable precise diagnosis and permit the initiation of treatment. The outcome is generally poor in the absence of treatment with an average survival rate of one to six weeks.

We are reporting the case of a 43-year-old type 1 HIV-infected patient, classified CDC stage C3, who has been on treatment for about 10 years and on HAART for about six months.

Case report
A 43-year-old HIV-1 infected patient, at stage C3 following the CDC classification, diagnosed 10 years before consultation, was on Aluvia® (Lopinavir 200 mg + Ritonavir 100 mg) two tablets bid and Duovir® (Lamivudine 150 mg + Zidovudine 150 mg) one tablet bid for six months, presented at the Yaounde University Teaching Hospital with persistent cough and odynophagia. A month before, he had reported unintentional weight loss, unremitting cough and persistent fever. Following a chest X-ray, which revealed diffuse right lung infiltrates, he was placed on amoxicillin 500 mg/clavulanic acid 125 mg (Augmentin®) one bid for 10 days. The persistence of the symptoms prompted the institution of co-trimoxazole (trimethoprim 160 mg + sulfamethoxazole 800 mg) two tablets bid for another 10 days in view of treating an eventual Pneumocystis jiroveci infection still with no avail. Instead, the symptoms grew worse with cough exacerbated by swallowing, purulent sputum and retrosternal pain. A second chest X-ray revealed a right lower lobar pneumonia. The diagnosis of pulmonary TB was considered and the patient was treated as a smear negative pulmonary TB. He was given rifampicin (R) 150 mg,isoniazide (H) 75 mg, ethambutol (E) 75 mg and pirazinamide (Z), that is, four tablets of the locally available fixed dose regimen corresponding to his weight of 60 kg. A slight improvement was noticed during the first two weeks of treatment after which he complained of progressive odynophagia.

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This prompted an upper endoscopy, which revealed an oesophageal ulcer at 24 cm from the dental arcade associated with a tracheo-oesophageal fistula (Figure 1). The tract of the fistula could not be analysed due to the lack of a water-soluble contrast agent, in order to avoid chemical pneumonia. Histopathologic analysis using routine staining techniques of the biopsied specimen concluded in a benign idiopathic ulcer.

The patient was placed on the antisycretory drug esomeprazol (Inexium™ 40 mg), one tablet bid. No other therapeutic option was offered to the patient, given the limited therapeutic options available. The patient’s clinical state rapidly depreciated and he died a week later.

Discussion

The occurrence of opportunistic gastrointestinal diseases is regressing since the introduction of combination antiretroviral therapy. However, several studies have reported resistances of HIV-1 to many antiretroviral drugs, making the occurrence of HIV-related oesophageal diseases possible in patients treated with such drugs, particularly in the severely immunodepressed. Among these diseases, tracheo-oesophageal fistula appears as one of the rare lesions in patients with pulmonary TB or as a complication of a TB-induced oesophageal ulcer. The particularity of our case report lies in the fact that, to the best of our knowledge, tracheo-oesophageal fistula complicating oesophageal ulcers, other than due to TB (namely, CMV, HSV or HIV), have been rarely reported. Our patient had risk factors for the development of HIV-related oesophageal diseases, namely HIV-1, CD4 count less than 200 cell/mm³ (the CD4 count was 40 cell/mm³).

There is no association between oesophageal symptoms and the various oesophageal diseases during HIV infection. Nevertheless, cough during swallowing brings up the problem of an oesophagotracheal leakage of foods and liquids due to the presence of a fistula. Other possible symptoms include odynophagia, retrosternal pain and dysphagia. The diagnosis of a tracheo-oesophageal fistula is suspected in case of cough during swallowing and confirmed at bronchoscopy or endoscopy. Endoscopy presents as an advantage to the possibility of realising multiple biopsies for histopathologic analysis, which is necessary for the etiologic diagnosis of TB. Routine staining techniques are sufficient enough for the diagnosis of most opportunistic infections in the HIV-immunodepressed patients. In case of TB, histopathologic analysis rarely shows acid fast bacilli and caseous granulomas. For the diagnosis of HIV-related idiopathic ulcers, an electron microscopy is needed to confirm the presence of the virus. Despite its diagnostic yield, the routine utilisation of polymerase chain reaction (PCR) has not been advocated by many authors. It will be ideal to combine several diagnostic methods. For our patient, we made use of routine staining techniques. Despite the absence of confirmation by electron microscopy, his fistula appeared to be a complication of an HIV-induced oesophageal ulcer.

Once the diagnosis of a tracheo-oesophageal fistula is established, the appropriate initial management is protecting the airways by either a tracheostomy, distally to the fistula or feeding the patient through a jejunol tube placed through a percutaneous gastrostomy and preventing gastro-oesophageal reflux. Surgery is performed later when the ventilatory and nutritional status are adequate. For our patient, we did not have the tube for percutaneous gastrostomy to allow enteral nutrition and tracheostomy was not judged necessary. It has been established that medical treatment is seldom necessary. Prednisone and thalidomide are well-established therapeutic options for an idiopathic ulcer. Thalidomide is not available in Cameroon. As for prednisone, though available, administering corticosteroids to our patient was not proper considering the fact that our working diagnosis was pulmonary tuberculosis. Survival varies from a week to six in the absence of an appropriate treatment. Death usually occurs as a consequence of malnutrition and lower respiratory tract infections. Our patient lived for less than eight weeks.

Figure 1: Tracheo fistula. (A: The arrow shows the bronchial secretions from the tracheo-oesophageal fistula; B: The arrow shows the esophageal lumen; C: The arrow shows the orifice of the tracheo-oesophageal fistula.)

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Conclusion
Despite the various antiretroviral regimens available in developing countries, HIV-related digestive diseases remain a challenge with poor prognosis due to limited therapeutic options. The diagnosis of tracheo-oesophageal fistula should be considered in the context of a persistent cough during swallowing in patients with AIDS.

Consent
Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Abbreviations list
CMV, cytomegalovirus; HAART, highly active antiretroviral therapy; HSV, herpes simplex virus; PCR, polymerase chain reaction; TB, tuberculosis.

References