

for 14 days with a diagnosis of pneumonia, and received ertapenem, clindamycin, corticosteroids and bronchodilators. Seven days after discharge, he was brought to our hospital due to progressive deterioration. Physical examination revealed cachexia, crackles in the left hemithorax, and slightly tender abdomen. Clinical laboratory testing showed increased acute phase reactants, without eosinophilia. Computed axial tomography showed consolidation in the left upper lobe, areas of cavitation with irregular walls (Fig. 1A), and marked dilation of the loops of the small intestine. The patient was treated with piperacillin-tazobactam and amikacin, without improvement. Fiberoptic bronchoscopy was performed, showing a greenish liquid secretion, shown on bronchial aspirate cytology to contain *Strongyloides stercoralis* filariform larvae (Fig. 1B). In view of the diagnosis of pulmonary infestation, albendazole and ivermectin were added to the therapeutic regimen, but the patient progressively worsened and died 26 days later.

Strongyloides stercoralis filariform larvae penetrate through the skin and travel through the venous system to the right heart cavities, and from there to the lungs. They pass through the glottis to the digestive system where they lay their eggs, releasing non-infectious rhabditiform larvae. These can transform to invasive filariforms during the autoinfection cycles, penetrating the intestinal mucosa to complete the cycle,⁵ producing ulcerations making the patient susceptible to bacteremia. In pulmonary infestation, larvae infiltrate the alveolar and vascular spaces, progressing to diffuse hemorrhagic interstitial pneumonitis, acute respiratory distress syndrome, or lobar pneumonia, as in the case discussed here.^{2,3}

Our patient worked barefoot in the fields, which may explain the port of entry, while various factors may have precipitated the infestation: gastrectomy with the consequent achlorhydria, absence of spleen, malnutrition, and corticosteroid treatment.¹⁻⁴ It is interesting to note that *Strongyloides stercoralis* parasitosis is

not a strictly exotic disease: it is also endemic on the Mediterranean coast,² with the highest prevalence occurring in farmers in certain regions. It seems a reasonable approach to detect larvae in risk situations, particularly before initiating immunosuppressive treatments, to prevent disseminated disease and death.^{1,3,4}

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Three Cases of Odontological Foreign Body Bronchoaspiration[☆]



Broncoaspiración de cuerpo extraño odontológico. A propósito de tres casos

To the Editor:

We read with interest the article recently published by Gómez López et al.,¹ reporting a case of bronchoaspiration of a metal odontological foreign body. The authors emphasize the unusual nature of the aspirated material, since only 2 cases have been published, 1 of which was reported by the same authors.

In our hospital, we have had the opportunity to extract foreign bodies similar to that reported by the authors from 3 patients. In the first of these cases, on March 14, 2007, a foreign body was located in a 71-year-old man and subsequently extracted in the operating room under general anesthesia with rigid bronchoscopy and basket. On July 9, 2009, odontological material was extracted from a 63-year-old woman in the bronchoscopy room, with flexible 2.2 mm forceps under topical anesthesia only. On June 10, 2014, we attempted to extract material from the third patient, a 75-year-old woman, in the bronchoscopy room with flexible bronchoscopy and sedation with midazolam. The attempt failed, so the following day

the silicon prosthesis was removed with rigid bronchoscopy and rigid forceps under general anesthesia in the operating room.

In all cases, the patients were undergoing dental surgery with osseointegrated implants at the time of aspiration of the foreign body. The objects were lodged in the right bronchial tree (basal pyramid and intermediate bronchi), the effect of gravity causing the thickest section to settle in the distal position, with the point facing upwards, facilitating removal by the endoscopist, as described by Leuzzi et al.² The bronchoaspirated material to which we refer³ is the surgical steel tip of a manual torque wrench, 20 mm in length (several different sizes are marketed), with a 1.31 mm hexagonal

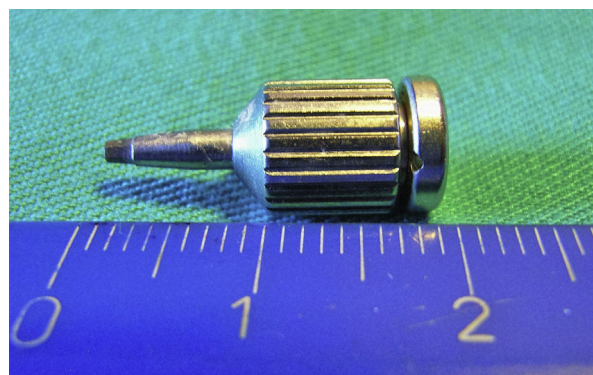


Fig. 1. Tip of manual torque key.

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tip. This device is attached to the torque wrench and used to screw in the titanium abutment screw, a step phase in this type of surgery. For safety purposes, these tips have a rotating crown which allows the screw to be turned, and is furnished with a small hole, as can be seen in the image, into which dental floss is introduced to prevent it falling and being aspirated (Fig. 1).

Our experience prompts us to make some comments. The methods of extraction in each of our 3 cases were very different, and the procedure depended basically on the degree of collaboration of the patient, and either flexible or rigid bronchoscopy was used to the same effect. We would advise an initial attempt with the former, as it is much more accessible in our hospital setting. However, any difficulty encountered during this procedure can be easily overcome with the use of rigid bronchoscopy. We were asked to provide 2 of the extracted pieces, which were presented as evidence in subsequent legal claims. Thus, we highlight the need for prevention of accidents of this kind, in view of their frequent occurrence, potential severity, and medical and legal implications.

Founder Mutation C.3344C>t(p.Pro1115Leu) in the EIF2KA4 Gene in Iberian Romani Patients With Pulmonary Venous-Occlusive Disease: A Warning for our Daily Practice*



Hallazgo de la mutación fundadora C.3344C>t(p.Pro1115Leu) en el gen EIF2KA4 en pacientes ibéricos de etnia gitana con enfermedad veno-oclusiva pulmonar: una llamada de atención a nuestra práctica diaria

To the Editor:

Pulmonary venous-occlusive disease (PVOD) is a rare form of pulmonary arterial hypertension. The incidence of this entity is unknown, partly due to underdiagnosis and mistaken classification as idiopathic pulmonary arterial hypertension (IPAH).

PVOD is distinguished by a marked reduction in carbon monoxide diffusing capacity (DLCO) and a typical radiological pattern. It occurs more often in men, and has a more aggressive course than IPAH.¹ Multiple causes, including genetic alterations, have been associated with its development. Recently, homozygous or compound heterozygous mutation of the *EIF2AK4* gene was described as the cause of PVOD. This mutation appears to occur in 25% of sporadic cases and 100% of familial cases, showing an autosomal recessive inheritance pattern and high penetrance.¹

Our group has described a homozygous founder mutation C.3344C>T(p.Pro1115Leu) in *EIF2AK4* in 18 patients from 10 highly consanguineous Romani families with several affected members (Table 1).²

All patients developed the disease as young adults (mean: 27.43±7.3 years), and most progressed rapidly to a fatal outcome (death or double-lung transplantation) in the first year after diagnosis.

Although the clinical characteristics of the patients varied on diagnosis, they all had a common trait: severely reduced DLCO.

It is interesting to note that the study of family members revealed a high incidence of death among relatives with no genetic studies but with a history suggestive of PVOD. Moreover, we

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found an alarming number of family members (59.7%) who were heterozygous carriers of the mutation, generating a risk of new homozygous cases in future generations (Table 1).

At the current time, the Romani population in Spain, a community characterized by a high level of consanguinity, is estimated to be around 750 000 individuals distributed around the whole country.^{3,4} Since this *EIF2AK4* mutation appears to be typical of the Romani race, and in view of the severity of the disease, we are facing a potentially serious public health problem among this population, which could be partially prevented by early genetic diagnosis and appropriate genetic counseling aimed at reducing the number of new cases.

Therefore, we believe that maintaining a high level of suspicion is essential for Spanish physicians: and that PVOD must be ruled out and a genetic study for *EIF2AK4* should be performed (as lung biopsy is contraindicated) in those Romani patients presenting with dyspnea and a family history of PAH or severely diminished DLCO. If *EIF2KA4* homozygous mutations are found, the patient must be rapidly referred to a hospital with an available lung transplantation program being the initiation of pulmonary vasodilators contraindicated due to the high risk of triggering severe pulmonary edema. Moreover, family members of carriers of this mutation must be screened and given appropriate genetic counseling, in order to avoid new cases in future generations and to prevent the propagation of this devastating disease.

Table 1
Summary of Families Studied.

| Family | No. of Index Cases | No. of Family Members Studied | No. of Healthy Heterozygous Family Members | No. of Homozygous Family Members Without PVOD |
|-----------|--------------------|-------------------------------|--|---|
| Family 1 | 3 | 12 | 7 | 0 |
| Family 2 | 1 | 0 | 0 | 0 |
| Family 3 | 4 | 13 | 8 | 1 |
| Family 4 | 1 | 1 | 1 | 0 |
| Family 5 | 1 | 0 | 0 | 0 |
| Family 6 | 3 | 28 | 21 | 1 |
| Family 7 | 1 | 1 | 0 | 0 |
| Family 8 | 0 | 8 | 3 | 0 |
| Family 9 | 2 | 5 | 1 | 0 |
| Family 10 | 2 | 1 | 0 | 0 |
| Total | 18 | 67 | 40 | 2 |

PVOD: pulmonary venous-occlusive disease.