10 mg montelukast every day and subcutaneous mepolizumab every 4 weeks. To date, no side effects have been observed, except for slight local edema at the injection site in the hours following administration of the drug.

In conclusion, we report the case of a pediatric patient with an asthma phenotype more usually seen in adults. Poor response to escalation of his usual treatment led to the prescription of mepolizumab, and the patient has shown good clinical, laboratory and functional response 1 year after starting.

References

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Pulmonary Arteriovenous Malformation After Metastatic Gestational Trophoblastic Tumor

Malformación arteriovenosa pulmonar después de un tumor trofoblástico gestacional metastásico

Dear Editor:

Pulmonary arteriovenous malformations (PAVMs) are abnormal communications between an artery and a vein without an intervening capillary component.1,2 While most PAVMs are congenital, caused by hereditary hemorrhagic telangiectasia (also known as Osler-Weber-Rendu disease), approximately 20% are acquired and can be due to chronic liver disease, schistosomiasis, mitral stenosis, trauma, previous cardiac surgery, actinomycosis, Fanconi syndrome, tuberculosis or tumors.3,4,5 Tumors may be followed by the formation of PAVMs upon long-term complete remission with treatment, a phenomenon which therefore may especially occur in case of metastatic tumors that are amenable to curing using chemotherapy, i.e. choriocarcinoma. Here, we report a case of PAVM following metastatic gestational trophoblastic neoplasia (GTN).

We report a case of a 67-year-old woman, occasional cigar smoker, who during her first pregnancy in 1977, when she was 27, presented with a hydatidiform mole requiring several curet tages. After an uterine rupture, a hysterec tomoy was performed. In the postoperative period, pulmonary metastases were detected, visualized as a perihilar left opacity. Chemotherapy with methotrexate and vincristine was then performed from April to June 1977, with a complete remission.

In December 2016, after an upper digestive endoscopy, a respiratory infection was diagnosed. In order to control the response to therapeutics, she underwent a chest radiography revealing a pulmonary nodule. The CT-scan showed a nodule, 20 mm in diameter, located in the apical segment of the left lower lobe (Fig. 1). Upon contrast injection, on the arterial phase, a complete fill-in of this nodule was seen, which formed an aneurysmal sac, with a connection to a sub-segmental branch of the upper branch of the pulmonary artery, and to a pulmonary vein, at the lower end. No internal thrombosis was observed. The diagnosis of PAVM was made. The patient was asymptomatic and denied episodes of hemoptysis, trauma, thoracic surgery, and no personal nor family history of epistaxis. At examination, no telangiectasia was seen in the oral cavity, lips, face or fingertips. The pulmonary auscultation was normal, with no audible thora catic murmur. Peripheral oxygen saturation was normal, with 97% at resting and in room air. Human chorionic gonadotropin (hCG) and hCG beta chain were low (<1 UI/L and <0.04 ng/mL respectively). The echocardiography and spirometry were normal.

Based on the history of molar pregnancy, and on the imaging demonstrating a typical PAVM in a territory where a metastasis of trophoblastic tumor had regressed with treatment, we diagnosed isolated PAVM developed on pulmonary sequelae of a metastatic molar pregnancy. The patient underwent percutaneous coil vaso-occlusion, with no complication and a good immediate angiographic result.

Molar pregnancies are classified as gestational trophoblastic disease, which include hydatidiform moles, invasive moles, choriocarcinomas, placental site trophoblastic tumors and epithelioid trophoblastic tumors.6 They occur at a rate of approximately one in every 600 conceptions.8 Approximately 10% of complete hydatidiform moles and 0.5% of partial hydatidiform moles undergo malignant transformation called GTN.5,7,8 In the majority of GTNs, the disease is limited to the uterus where the abnormal trophoblast proliferation and localized hCG production may lead to
focal vascular changes, including the formation of AVMs. A recent systematic review of uterine AVMs following gestational trophoblastic neoplasia found 50 cases. Although the lung is the most frequent site of metastasis in GTN, the formation of PAVMs has been rarely described, and mostly in cases of choriocarcinoma.

Although most patients are asymptomatic, PAVM can lead to dyspnea and complications such as hemoptysis, hemothorax, brain abscess or embolic events. Even though the natural history of PAVMs is not well known, morbidity and mortality of untreated PAVM are thought to be higher than those associated with treatment. PAVM can be treated either by surgery or by embolization but the latter has been recommended as treatment of choice. The possibility of recurrence due to recanalization occurring in up to 25% of cases is the major concern with embolization therapy.

As far as we know, this is the second report of a PAVM after a gestational trophoblastic disease such as molar pregnancy. As GTN are highly vascularized tumors, due to high levels of HCG which stimulate angiogenesis, we hypothesize that a PAVM was formed within or next to the pulmonary metastasis of the GTN and remained at the metastatic site, even though the metastasis was successfully controlled by chemotherapy and no recurrence was found (proved by the low serum value of HCG). Such mechanism can only be hypothesized for metastasis from tumors which can be cured by chemotherapy. In fact, it has been described that AVM can persist after a successful eradication of the GTN by chemotherapy.

References
Nocardia otitidiscaviarum Infection in an Immunocompetent Patient

Infección pulmonar por Nocardia otitidiscaviarum en paciente inmunocompetente

To the Editor,

Nocardia is a strictly aerobic Gram-positive bacterium that is partially acid-alcohol-resistant and forms long branched filaments. Species of medical significance are: N. asteroides, N. brasiliensis, N. pseudobrasiliensis and N. otitidiscaviarum. Infection by the latter is rare compared with other species of Nocardia and rarely causes infection in humans, even in immunocompromised patients. All species are universally distributed and routinely found in soil, sand, dust, and stagnant water.

We report a case of pyopneumothorax caused by Nocardia otitidiscaviarum, in an 89-year-old woman with mild persistent asthma, bronchiectasis in the left upper and lower lobes, and obesity. She had had 2 episodes of community-acquired pneumonia approximately 5 and 10 years previously. She had never smoked and could perform activities of daily living independently. Her usual medication consisted of benzodiazepines, proton pump inhibitor, and long-acting β-2 adrenergic bronchodilators combined with corticosteroids (budesonide/formoterol 160/4.5 mcg, 1 inhalation every 12 h) and on-demand short-acting β-2 adrenergic receptors for several years. She presented in the emergency department in December 2015 with a 1-week history of cough without expectoration, progressive dyspnea on moderate exertion, febrile sensation, constipation, nausea, and vomiting. She denied chest pain, dysuria, and other symptoms. On physical examination, she was conscious, oriented, and collaborative; she had tachypnea 20 breaths/minute, SpO2 86% breathing room air, blood pressure 133/55 mmHg, heart rate 87 bpm, and axillary temperature 39.6°C. On pulmonary auscultation, breath sounds were normal, with abundant disperse rhonchi. The rest of the physical examination was normal. Chest X-ray was performed, showing right basal peribronchial thickening, cardiomegaly, and aortic elongation. ECG with no changes. Arterial blood gases: pH 7.46, pCO2 30 mmHg, and pO2 48 mmHg. Abnormal clinical laboratory results were sodium were 133 mmol/L, LDH 460 U/L, PCR 22 mg/l, leukocytosis 10.83×10⁹/l, and neutrophilia 9.35×10⁹/l. Urine analysis showed moderate pyuria.

Diagnoses of acute partial respiratory failure, respiratory infection, and urinary infection were given, and the patient was admitted for antibiotic treatment in the form of levofloxacin, in addition to aerosols and corticosteroids. Escherichia coli in urine was confirmed on day 3 of admission.

The patient’s progress was poor. Eight days later, an image was observed in a radiographic follow-up of consolidation in the right middle and left lower lobes, so nosocomial pneumonia was suspected and antibiotic treatment was switched to ceftazidime and ciprofloxacin. On day 9, urine was positive for pneumococcal antigen. On days 12 and 16 after admission, N. otitidiscaviarum was isolated in a sputum culture, and treatment was changed, in line with the sensitivity studies, to cotrimoxazole, amikacin, and imipenem. In view of the worsening symptoms, a chest computed tomography was then performed (Fig. 1) which revealed right hydropneumothorax and areas of consolidation, some of which were cavitary, predominantly in the right lung base. A pleural drainage tube was placed, from which purulent fluid was obtained, determined to be predominantly polymorphonuclear exudate with a pH <7.2, indicating, more specifically, a case of pyopneumothorax. Throughout the period of admission, the patient continued to require high oxygen levels to maintain SatO2 at around 90%, and after suffering paralytic ileus that could not be corrected despite placement of a nasogastric and rectal tube, she died 35 days after admission.

Nocardiosis is a suppurative infection that can occur acutely or chronically and tends to disseminate. It generally appears as an opportunistic infection in individuals with T cell-mediated immunosuppression. The most common risk factors are occult malignancies, diabetes, HIV, and COPD, although it can also occur in immunocompetent patients (up to one third of all cases). The most common routes of infection are inhalation and cutaneous inoculation. Pulmonary nocardiosis is the most common clinical presentation. This is a severe, sporadic disease that is more prevalent among men, and generally occurs in the fourth decade of life. Central nervous system and subcutaneous involvement may be present in up to 30% and 15% of cases, respectively.

Fig. 1. Transversal CT slices of the chest performed on day 23 of admission, showing right hydropneumothorax, right lower lobe atelectasis and areas of consolidation, some cavitary, mainly in the right lung base. Cavitary pulmonary nodules and ground-glass opacities adjacent to the area of consolidation can be seen in the left lower lobe.


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