breast, and colon/rectum) promotes angiogenesis and consequently leads to increased blood supply and faster tumor growth.[2,6] Bevacizumab is a monoclonal antibody that targets VEGF-A, preventing it from binding to its receptor, and thereby inhibiting angiogenesis. Nonetheless, the potential for recurrence and the short disease-free periods observed suggest that there are other factors at play and that VEGF inhibition alone is not sufficient to stop the tumor from growing.[7]

Five cases of cutaneous angiosarcoma treated with bevacizumab, alone or in combination, have been reported to date. A favorable response was reported in all cases, with good tolerance and longer survival than that seen with other treatments (Table 1).

The results of the first phase II clinical trial on the use of bevacizumab alone to treat metastatic or locally advanced angiosarcoma were recently published by Agulnik et al.[2] The results are promising, as half of the patients showed stable disease with a mean time to disease progression of 26 weeks.

Considering the scarcity of effective treatments and the results reported to date for bevacizumab in isolated cases and clinical trials, we believe that this drug could be considered in cases similar to the one we have presented. There are also clear indications that bevacizumab used in combination with other drugs could improve the prognosis in patients with angiosarcoma.

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Autochthonous Cutaneous Myiasis Due to *Chrysomya bezziana*☆

Miasis cutánea no importada por *Chrysomya bezziana*

Myiasis is a parasitic infestation of the tissues or organs of vertebrates (including humans) produced by the larvae of different species of fly (Diptera).[1–3] It is classified according to the association between the parasite and the host and is described as obligatory, facultative, or accidental. It can also be classified according to the anatomic site of infestation as cutaneous, intestinal, or cavitary myiasis.[1–3] Clinically, cutaneous myiasis is divided into furuncular, migratory, and wound forms.[1–3] Incidence is higher in tropical countries with a humid climate and a low socioeconomic level.[1–3]

We present a case of autochthonous cutaneous myiasis caused by *Chrysomya bezziana*, a species that is exceptional in Europe.

The patient was a 56-year-old man with a personal history of obesity, occasional alcohol consumption, arterial hypertension, sleep apnea-hypopnea syndrome, and extrinsic asthma. He was a heterozygous carrier of the PT20210A mutation, although he had no history of thrombosis. He had a vascular ulcer on the left leg that first appeared 10 months

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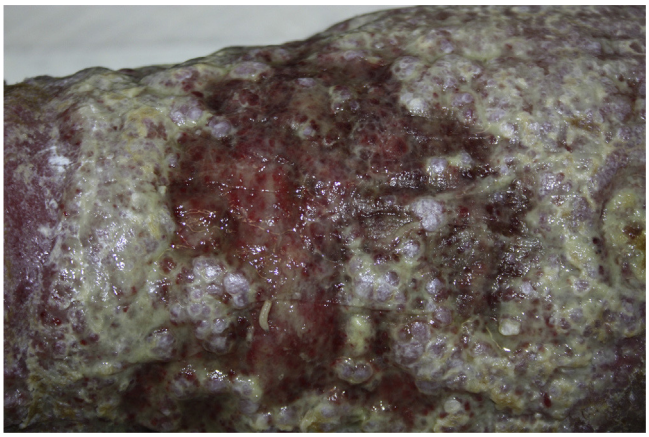

Figure 1 Ulcer on the left calf. Note the erythematous surface, which is papilliform in appearance, and abundant exudate. Several larvae can be seen on the surface of the lesion.

previously. Live larvae were detected during local care of the ulcer at his health center, from where he was referred to the emergency department. The patient had a good general status and denied having traveled abroad, recent contact with animals, or trips to the countryside. Physical examination revealed signs of chronic lymphedema in both legs. An ulcer was observed on his left calf. It measured 15 × 8 cm, had an erythematous surface, and was papilliform in appearance. It was also friable, with malodorous exudate (Fig. 1). Several mobile Diptera larvae were identified on the lesion. These were whitish in color and in 1 cm in length. They were eliminated mechanically and sent to the microbiology service for identification. The lesion was washed with saline solution and treated with an occlusive dressing of unscented pure petroleum jelly, and the patient was referred to the vascular surgery service. Macroscopically, the larvae extracted were identified as *C bezziana*; microscopically, subtle spine bands were visible around the body and the peritreme of the open posterior spiracle.

In developed countries, cutaneous wound myiasis is associated with poor hygiene, advanced age, psychiatric conditions, alcoholism, diabetes mellitus, and occlusive vascular disease.[1–3]

The 3 species that produce this form of myiasis throughout the world are *Cochliomyia hominivorax* (New World screw-worm), and *C bezziana* (Old World screw-worm)—both from the Calliphoridae family—and *Wohlfahrtia magnifica*, which belongs to the Sarcophagidae family.[1–3] Most cases of autochthonous myiasis reported in Spain involve various species of the Sarcophagidae family (flesh fly).[4]

The parasite *C bezziana* is distributed in tropical and subtropical areas of Africa, southeast Asia, India, and the Middle East.[1–4] It produces an obligatory form of myiasis that affects various domestic animals (cattle, horses, and pets) and, occasionally, humans.[1,4] An adult female can deposit hundreds of eggs on the margin of wounds. The eggs hatch in 16 hours, and the resultant numerous larvae feed for a week on the host's tissue. They then fall to the ground and complete their biological cycle to become adults.[1,4] Under optimal conditions, the biological cycle lasts approximately 20 days.[1,4] In humans, *C bezziana* produces wound infestations,[4] skin tumors[5], and cavities (orbit,[6] mouth,[7] and ear[8]), all of which have high morbidity. In severe cases, the patient can present with fever, a sensation of poor temperature regulation, pain, bacterial superinfection, leukocytosis with neutrophilia, or hypereosinophilia.[2]

In a review from the year 2000, the 47 species of Diptera responsible for myiasis in Spain did not include *C bezziana*.[9] In the Spanish scientific literature, we found only 3 cases of human myiasis caused by this species: the first involved a 41-year-old Spanish man with a personal history of chronic alcoholism who developed cutaneous myiasis on chronic ulcers on the left leg that were secondary to rhabdomyolysis[4]; the second involved a 65-year-old Spanish woman with myiasis of the ear who reported no recent trips abroad[8]; and the third involved a 54-year-old Spanish man with myiasis on a supraglottic squamous cell carcinoma of the larynx that manifested as a tumor mass on the anterior aspect of the neck.[6] International migration and climate change can explain, at least in part, why this species of fly is identified in countries that are not its natural habitat.[10]

In conclusion, we present a case of cutaneous myiasis on an ulcer in which *C bezziana* was identified as the causal agent. To our knowledge, this is the second case of autochthonous cutaneous myiasis caused by this species of fly. We wish to draw attention to this uncommon entity and to the importance of correct conservation and identification of larvae as part of routine clinical practice, since this condition is probably under-reported.

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Type 2 Mosaicism in Familial Glomangiomatosis☆

Mosaicismo tipo 2 en glomangiomatosis familiar

Autosomal dominant skin diseases can sometimes present as linear mosaicism or as segmental mosaicism that follows the Blaschko lines. Type 1 mosaicism is characterized by the presence of a body segment affected by the disease in a healthy individual as a consequence of a postzygotic germline mutation in the segment. Type 2 mosaicism is characterized by the diffuse presentation of the disease in association with superimposition of a more involved body segment and is caused by the loss of heterozygosity in this segment during embryonic development in an individual who is heterozygous for the disease.[1]

A 6-year-old girl with no personal history of interest presented with an asymptomatic segmentally distributed congenital bluish lesion that extended from the right groin along the medial aspect of the right thigh and leg to the right foot. The lesion had grown in proportion to the patient's own physical development. She also presented solitary nodular lesions that had begun to appear when she was 3 years old. These were painful to pressure and to changes in temperature and were scattered on all 4 limbs. Physical examination revealed a bluish plaque with palpable elastic nodules on the medial and posterior aspects of the right thigh and medial aspect of the right foot following a segmental course (Fig. 1). In addition, papules and nodules that were painful to pressure were observed on the contralateral foot and at distal sites on the other extremities. The patient's family history was remarkable in that her mother had scattered bluish nodular lesions (approximately 1 cm in diameter) on the limbs and trunk that first began to appear during adolescence and were similar to those of the patient (Fig. 2). These lesions had not been examined previously. Biopsy specimens of the mother's lesions were taken, and histopathology revealed a new vascular formation in the deep dermis with wide vascular lumens filled with red cells and lined by glomus cells. The picture was consistent with glomuvenous malformations (Fig. 3). The congenital bluish plaque, the subsequent appearance of scattered lesions, and the mother's history of findings that were consistent with glomangiomatosis led us to make a diagnosis of type 2 mosaicism in familial glomangiomatosis. Given that the patient's quality of life is not affected, a wait-and-see approach with regular follow-up was adopted.

Glomangioma, or glomuvenous malformation, is a hamartoma of the dermal glomus bodies, which are responsible for temperature regulation. The 2 clinical forms described are solitary glomus tumor and glomuvenous malformations, or multiple glomangiomas. The first presentation is the more common and manifests typically in adults as painful isolated bluish nodules at acral sites. Glomuvenous malformations, or multiple glomangiomas, are characterized by the pro-

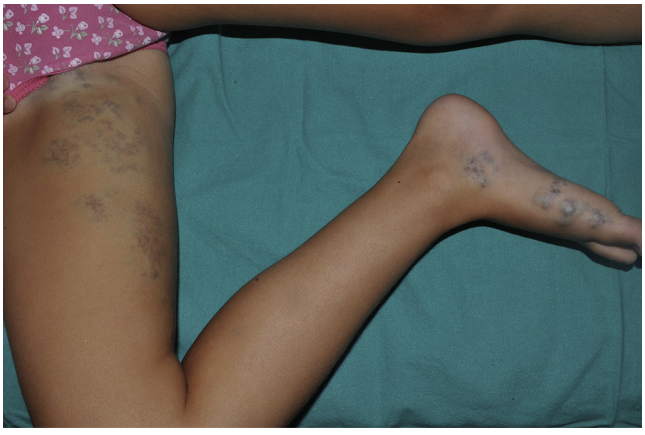

Figure 1 Segmentally distributed bluish plaque extending from the right groin along the medial aspect of the right thigh and leg to the right foot.

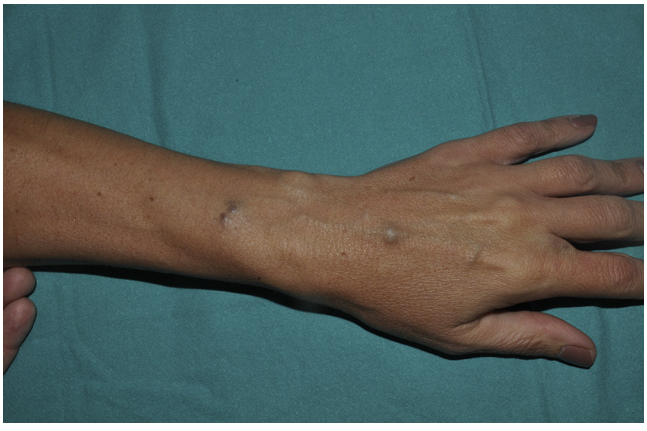

Figure 2 Bluish nodule on the mother's right forearm.

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