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Refractory Visceral Leishmaniasis Responding to Second-Line Therapy

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Abstract : Introduction : In India, Leishmania donovani is the only parasite causing Leishmaniasis. The parasite infects the reticuloendothelial system and is found in the bone marrow, spleen and liver. Treatment of choice is amphotericin-B with sodium stibogluconate being an alternative. Miltefosine is useful in refractory cases. In our case, Leishmaniasis occurred in a person residing in western India (which is quite rare) and it failed to respond to two different drugs (again an uncommon feature) before it finally responded to a third one. Description: A 50 year old lady, a resident of western India, with no history of recent travel, presented with an ulcer on the left side of the nose since 8 months. She was apparently alright 8 months back, when she noticed a small ulcerated lesion on the left ala of the nose which was immediately biopsied. The biopsy revealed amastigotes of Leishmania for which she was administered intra-lesional sodium stibogluconate for 1 month (4 doses every 8 days).Despite this, there was no regression of the ulcer and hence she presented to us for further management. On examination, her vital parameters were normal. Barring an ulcer on the left side of the nose, rest of the examination findings were unremarkable. Complete blood count was normal. Ultrasound abdomen showed hepatomegaly. PET-CT scan showed increased metabolic activity in left ala of nose, hepatosplenomegaly and increased metabolic activity in spleen and bone marrow. Bone marrow biopsy was done which showed hypercellular marrow with erythroid preponderance. Considering a diagnosis of leishmaniasis which had so far been unresponsive to sodium stibogluconate, she was started on liposomal amphotericin-B. At the time of admission, her creatinine level was normal, but it started rising with the administration of liposomal amphotericin-B, hence the dose was reduced. Despite this, creatinine levels did not improve, and she started developing hypokalemia and hypomagnesemia as side effects of the drug, hence further reductions in the dosage were made. Despite a total of 3 weeks of liposomal amphotericin-B, there was no improvement in the ulcer. As had so far failed to respond to sodium stibogluconate and liposomal amphotericin-B, it was decided to start her on miltefosine. She received the miltefosine for a total of 12 weeks. At the end of this duration, there was a marked regression of the cutaneous lesion. Conclusion: Refractoriness to amphotericin-B in leishmaniasis may be seen in up to 5 % cases. Here, an alternative drug such as miltefosine is useful and hence we decided to use it, to which she responded adequately. Furthermore, although leishmaniasis is common in the eastern part of India, it is a relatively unknown entity in the western part of the country with the occurrence being very rare. Because of these 2 reasons, we consider our case to be a unique one.

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