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Outcomes of Pregnancy in Women With Idiopathic Inflammatory Myopathies in Africa



Dear Editor,

Idiopathic inflammatory myopathies (IIMs) are associated with adverse pregnancy outcomes in Caucasian and Asian populations,^{1,2} but this issue is unclear in Africa. For the purpose of this paper, we conducted a systematic review of the literature to identify studies on IIMs and pregnancy in Africa from electronic and hand searches up to March 4, 2021, using key search terms referring to IIMs, pregnancy and African countries as per the United Nations Classification.³

Of 118 records retrieved from PubMed, Embase, Africa Journals Online and hand searches, we included 4 relevant case reports and 2 case-series^{4–9} from Gabon, Mali, Morocco, Senegal and Tunisia. The search strategy in PubMed and Embase as well as the study selection process are summarized in the [Supplementary Table and the Supplementary Figure](#). Included records report a total of 18 singleton post-IIM pregnancies and 10 singleton pre-IIM pregnancies in 12 women aged 26–42 years at conception. Among women with ethnicity data, 6 were Black Africans, 1 Black Caribbean and 1 North African. Specified IIM subtypes were overlap myositis ($n=4$), dermatomyositis ($n=4$) and immune-mediated necrotizing myopathy ($n=2$). Regarding pre-IIM pregnancies, there were only 2 adverse pregnancy outcomes: medical termination of a pregnancy (for unspecified cause) and one stillbirth. In women with post-IIM pregnancy data, 8 of 18 pregnancies were successful. Adverse maternal outcomes recorded in post-IIM pregnancies

were premature delivery ($n=4$), cesarean section ($n=3$), medical termination for unspecified causes ($n=3$) and pulmonary infection ($n=1$). Adverse fetal/neonatal outcomes were pre-term birth ($n=4$), neonatal death ($n=2$), small for gestational age ($n=2$), stillbirth ($n=1$) and neonatal lupus ($n=1$) ([Table 1](#)).

Maternal and offspring outcomes of pre- and post-IIM pregnancies are poorly characterized in Africa. It remains unknown whether the observed adverse outcomes were coincidental or connected with IIMs, although this small pooled sample likely suggests together with studies from other regions^{1,2} that, increased rates of adverse outcomes may be observed in women (and their infants) with IIMs in Africa as well. There is a need for a prospective multicenter African registry to better assess the link between IIMs and adverse pregnancy outcomes, as well as the impact of pregnancy on IIM activity in Africa.

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Conflicts of interest

The authors declare no conflict of interest.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at [doi:10.1016/j.reuma.2021.04.005](https://doi.org/10.1016/j.reuma.2021.04.005).

Table 1
Characteristics and outcomes of pregnancy in women with idiopathic inflammatory myopathies in Africa.

Study	Period of recruitment	Country	Patient number	Ethnicity/race	Age, years	Number of pre-IIM pregnancy(ies)	Outcomes of pre-IIM pregnancy	Year of diagnosis of IIM	Subtype of IIM	Post-IIM pregnancy		Treatment before pregnancy	Disease activity at conception and throughout pregnancy	treatment of IIMs through pregnancy	Outcomes of post-IIM pregnancy	Post-partum outcome of IIMs													
										Number	Year																		
Iba-Ba, 2009 [4]	2004–2005	Gabon	1	Black African	30	0	–	2004	Myositis overlapping with SLE	1	2005	-GCs -AM	Inactive	None	-Premature delivery -Caesarean section at 32 weeks, due to pneumonia -Neonatal death at day 12 of life	NR													
Kaddour, 2009 [5]	1979–2007	Tunisia	2	NR	21	0	–	1993	Myositis overlapping with RA	0	–	–	–	–	–	–	–												
					27					1	1999	GCs	Inactive	GCs	Successful	NR													
					28					2	2000	-GCs -MTX	Inactive	-GCs -MTX	Medical termination for unspecified cause	NR													
			3		30	2	Successful	2000	Myositis overlapping with APS	3	2002	GCs	Inactive	GCs	Inactive	GCs	Successful	NR											
					32					4									2004	GCs	Inactive	GCs	Successful	NR					
					22					1									2004	GCs	Inactive	GCs	Stillbirth at 28 weeks gestation	NR					
					26					2									2006	GCs	Inactive	-GCs -LDA -LMWH	-Premature delivery at 32 weeks gestation -Neonatal death at day 3	NR					
			4		41	3	Successful	2002	Myositis overlapping with SLE	0	–	–	–	–	–	–	–	–	–										
					42					1										2003	-GCs -AM	Inactive	-GCs -AM	Medical termination for unspecified cause	NR				
					25					0										2003	DM	0	–	–	–	–	–	–	–
28	1	2006	-GCs -AM -MTX	Inactive	-GCs -AM -MTX	Medical termination for unspecified cause	NR																						
Ousmane, 2016 [6]	2012	Senegal	6	Black Caribbean	26	1	Successful	2008	DM	1	2008	–	–	–	–	Successful	NR												
			7		1	Successful	2012	IMNM	0	–	–	–	–	–	–	–	Flare												
Awatef, 2016 [7]	NR	Morocco	8	Black African	35	1	Successful	2012	IMNM	0	–	–	–	–	–	–	Flare												
			9		0	–	NR	DM	1	NR	NR	NR	Active	-GCs -AM	Successful	Improvement													
Cisse, 2018 [8]	NR	Mali	10	Black African	NR	NR	NR	NR	DM	1	NR	NR	NR	NR	Neonatal cutaneous lupus	NR													
Iba-Ba, 2019 [9]	2008–2018	Gabon	11	Black Africans	20	NR	NR	NR	NR	NR	1	NR	NR	NR	Inactive	NR	-Caesarean section	NR											
					21												NR	NR	NR	NR	NR	NR	NR	-Term Live birth	NR				
					23												NR	NR	NR	NR	NR	NR	NR	-Premature delivery	NR				
					24												NR	NR	NR	NR	NR	NR	NR	-Caesarean section	NR				
					24												NR	NR	NR	NR	NR	NR	NR	-SGA	NR				
12	29	NR	NR	NR	NR	NR	NR	NR	NR	0	–	–	–	–	–	–	–												
	36.5																	NR	NR	NR	NR	NR	NR	NR	NR	Inactive	NR	Successful	NR
	39.5																	NR	NR	NR	NR	NR	NR	NR	NR	NR	Inactive	NR	Successful

IIM, idiopathic inflammatory myopathy; SLE, systemic lupus erythematosus; GCs, glucocorticoids; AM, antimalarials; NR, not reported; RA, rheumatoid arthritis; MTX, methotrexate; APS, antiphospholipid syndrome; LDA, low-dose aspirin; LMWH, low molecular weight heparin; DM, dermatomyositis; IMNM, immune-mediated necrotizing myopathy; SGA, small for gestational age.

References

- Munra S, Christophe-Stine L. Pregnancy in myositis and scleroderma. *Best Pract Res Clin Obstet Gynaecol.* 2019; <http://dx.doi.org/10.1016/j.bpobgyn.2019.10.004>.
- Che WG, Hellgren K, Septhansson O, Lundberg IE, Holmqvist M. Pregnancy outcomes in women with idiopathic inflammatory myopathy, before and after diagnosis—a population-based study. *Rheumatology.* 2020;59:2572–80.
- United Nations Statistics Division. Geographic regions. USA, New York; 2020. <https://unstats.un.org/unsd/methodology/m49/> [accessed 30.12.20].
- Iba-Ba J, Mayi-Tsonga S, Bignoumba IR, Diallo T, Kombila M, Coniquet S, et al. Dermatomyosite et grossesse: une observation au Gabon. *Med Trop.* 2009;69:603–5.
- Kaddour N, Marzouk S, Frigui M, Chaabouni Y, Maazoun F, Ben Salah R, et al. Grossesse au cours des dermatomyosites et polymyosites. *Rev Méd Intern.* 2009;30:S97.
- Ousmane C, Makhtar BEH, Fatoumata B, Massi GD, Lémine DSM, Soda DSM, et al. Polymyositis and anti-SRP antibodies and pregnancy about 2 cases. *PAMJ.* 2016;24:192.
- Awatef K, Salim G, Zahra MF. A rare case of dermatomyositis revealed during pregnancy with good outcome. *PAMJ.* 2016;23:117.
- Cisse L, Karabinta Y. Neonatal lupus in an infant of a mother followed up for dermatomyositis: medical images. *PAMJ.* 2018;31:117.
- Iba Ba J, Nseng N, Ntsame N, Igalá M, Kombila U, Malekou M, et al. Grossesses au cours de maladies auto-immunes en zone subsaharienne à travers l'expérience du service de médecine du CHU de Libreville. *Médecine Santé Trop.* 2019;29:206–12.

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The Value of a Negative Antinuclear Antibody (ANA) Test: An Often Forgotten Result



El valor de una prueba de anticuerpos antinucleares (ANA) negativa: un resultado a menudo olvidado

Dear Editor:

It is quite clear that medicine is biased towards positive results and the same applies to the practice of pathology.¹ One of the ubiquitous tests in autoimmunity, the antinuclear antibody (ANA) suffers from this very same fate. A number of guidelines report on the clinical utility of a positive ANA and dissuade clinicians from requesting this test in the setting of low pre-test probability

for an ANA-associated autoimmune disorder (AAD).² This is certainly sound advice and prevents unnecessary investigations and healthcare expenditure. Yet, it is important to realise the clinical importance and pitfalls of a negative ANA results which sometimes becomes forgotten.

The internationally-accepted “gold standard” to measure ANA is via indirect immunofluorescence on HEP-2 cells.³ A negative ANA test on HEP-2 substrate usually means that there is no significant detection of IgG ANA (in the nucleus) at a specified dilution of serum – usually 1:80 to 1:160. There is a move to also classify positive cytoplasmic and mitotic staining of the HEP-2 substrate as ANA positive.^{3,4} This may improve the sensitivity of detecting AADs and prompt appropriate further testing and follow-up (Fig. 1).³

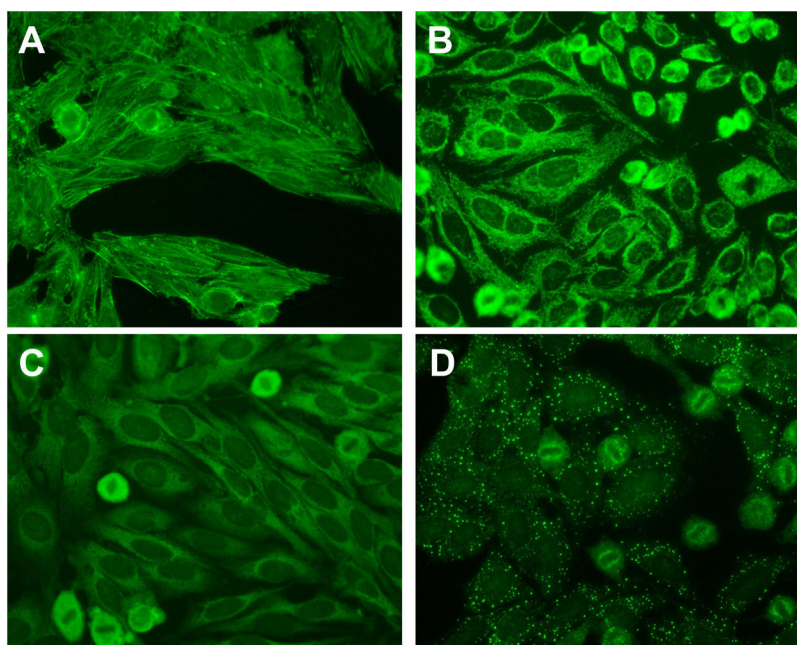


Fig. 1. Example cytoplasmic staining on the HEP-2 substrate. (A) F-actin staining suggesting the presence of smooth muscle antibodies found in autoimmune hepatitis and related disorders. (B) Coarse, granular cytoplasmic staining suggestive of anti-mitochondrial antibodies found in primary biliary cirrhosis. (C) Smooth, homogenous cytoplasmic staining suggestive of anti-ribosomal P antibodies found in systemic lupus erythematosus. (D) Large cytoplasmic dots staining suggestive of anti-GW bodies. All micrographs are taken at a magnification of 400×.