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SCIENTIFIC LETTER TO THE EDITOR

Overlap Syndrome: A Child Cohort

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Sir,

Overlap syndrome is defined by the presence of two or more rheumatic diseases either on the basis of clinical involvement and/or presence of autoantibodies [1]. These disorders are ill defined and difficult to categorize. The most defined entity amongst them is mixed connective tissue disease (MCTD). The studies of child cohort with overlap syndrome are lacking in literature.

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A prospective observational study was done at the pediatric rheumatology clinic over a period of 8 y (2004–2011) to detect the incidence and delineate the pattern of overlap syndrome. ACR criteria were used for diagnosis of all rheumatological cases. Overlap was diagnosed, either at presentation or at follow up, by clinical features of two major rheumatic diseases along with autoantibodies or positive biopsy findings as applicable.

Out of the total 1,544 children with rheumatological diseases, 6 had overlap syndrome (Table 1). Four had overlap at presentation while two developed it 6–7 mo later. The overall incidence was 3.89 per 1,000 (95 % CI 0.03–7.74 per 1,000) with female preponderance (F: M= 5:1). MCTD was seen both in boy and girl, one each. Out of 210 Juvenile Idiopathic Arthritis (JIA) patients, 2 developed overlap syndrome with features of cutaneous scleroderma {2/210 approx (1 %), M: F=1:1[2]. Two girls out of 44 childhood lupus developed overlap. One had juvenile dermatomyositis whereas other had cutaneous scleroderma {2/44 (4.2 %)} [3]. Both patients had renal involvement.

Incidence of overlap in adults was reported to be 25 % from UK and 20 % involving mixed population from Poland [4, 5]. Our study, first of its kind from India, demonstrated much lower incidence in children.

Study of overlap syndrome in children regarding diagnostic criteria, autoantibody profile and prospective cohort is not documented in literature except sporadic case reports [1]. So the present study of child cohort with overlap syndromes may give some insight till the larger series in child population is available. Pediatricians must be careful and vigilant in searching overlap in rheumatological cases either at presentation or at follow up.

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Table 1 Summary of childhood cohort of overlap

No.	Age (y), sex	Onset (y)	Presentation	Diagnostic data	Final diagnosis
1.	3, F	1	Overlap JIA and CS	ANA, anti-dsDNA-negative Anti-U1snRNP-negative RF, Anti-Scl70-positive Skin biopsy-CS	Overlap JIA and CS
2.	2, F	1	Overlap JIA and CS	RF,ANA, anti-dsDNA and Anti-U1snRNP-negative Anti-Scl70-positive Skin biopsy-CS	Overlap JIA and CS
3.	8, F	7	MCTD	RF-positive ANA, Anti-Scl70-negative Anti-U1snRNP-positive	MCTD
4.	12, M	9	MCTD	RF, ANA, anti-dsDNA-negative Anti-U1snRNP-positive	MCTD
5.	3, F	2.5	JDM	ANA, Anti-dsDNA-negative Anti PM-Scl-positive Skin biopsy-JDM	Overlap SLE and JDM (after 6 mo)
6.	9, F	8	SLE	ANA, Anti-dsDNA, Anti- SCL70-positive Skin biopsy-CS	Overlap SLE and CS (after 7 mo)

JIA Juvenile idiopathic arthritis; *CS* Cutaneous scleroderma; *MCTD* Mixed connective tissue disorder; *JDM* Juvenile dermatomyositis; *SLE* Systemic lupus erythematosus; *ANA* Antinuclear antibody; *RF* Rheumatoid factor

References

1. http://www.wsahs.nsw.gov.au/services/rheumatology/documents/OVERLAP.pdf accessed on 8/9/2011
2. Nandi M, Ganguli SK, Mondal R, Ghosh A. Clinico-serological profile of juvenile idiopathic arthritis. Indian Pediatr. 2009;46:640–1.
3. Mondal R, Nandi M, Ganguli S, Ghosh A, Hazra A. Childhood lupus: experience from Eastern India. Indian J Pediatr. 2010;77:889–91.
4. Maddison PJ. Mixed connective tissue disease: overlap syndromes. Baillieres Best Pract Res Clin Rheumatol. 2000;14:111–24.
5. Sharp GC, Irvin WS, Tan EM, et al. Mixed connective tissue disease: an apparently distinct rheumatic syndrome associated with a specific antibody to an extractable nuclear antigen. Am J Med. 1972;52:148–59.