



Letter to the Editor



COVID-19 and social responsiveness: A comparison between children with Sotos syndrome and autism

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The outbreak of COVID-19 severely impaired health worldwide, and the subsequent emergency measures established by governments in relation to home confinement implicated indirect consequences on wellbeing and everyday life scheduling for children and adults.

Specifically, the Italian Government imposed home-confinement for all residents from March 9th 2020 to May 4th 2020. Most of the work activities were compulsorily interrupted except for those considered as essential (i.e. doctors, nurses, employed in supermarkets). Home exiting was permitted only for indispensable and critical requirements.

Children with special care needs and their families, represented a fragile population on which the extreme circumstances due to COVID-19 could have doubly impaired (Asbury et al., 2020). Interruption of school activities and therapeutical interventions delivered in-person, in addition to the reduced possibility to leave homes and subsequent social isolation, constituted the main challenges for children with neurodevelopmental disorders including Autism Spectrum Disorder (ASD) and genetic conditions such as Sotos Syndrome.

ASD is an early-onset and lifelong neurodevelopmental condition characterized by social communication and social skill deficits, restricted interests and repetitive behaviors (Diagnostic and Statistical

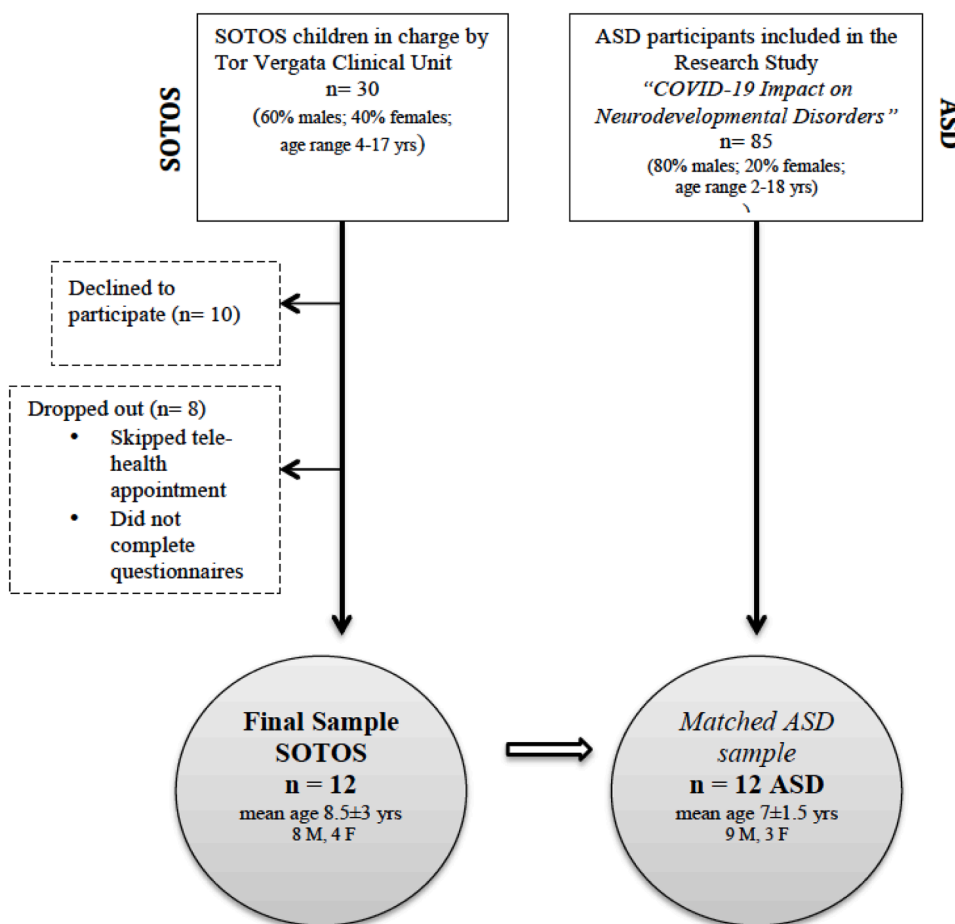


Fig. 1. Flowchart of the study: sample recruitment.

Illustrated in the Figure the main procedure of participants' recruitment.

Sotos Syndrome sample was constituted by children coming from the clinical database of the ChildPsychiatry Unit of the Tor Vergata Hospital. A multidisciplinary team (child psychiatrist, psychologist) of our Unit contacted the families by phone, described the study and invited them to participate, planning a tele-health appointment (10 declined to participate). 8 dropped out of the study (they skipped the tele-health appointment or did not complete the questionnaires required). Therefore the final sample consisted of 12 Sotos participants.

ASD sample comprised participants included in the research study, named "COVID-19 Impact on Neurodevelopmental Disorders", active at the Child Psychiatry Tor Vergata Unit. ASD participants were matched for age and sex with the SOTOS children included in the study.

Manual of Mental Disorders-Fifth Edition, DSM-5).

Sotos syndrome is a congenital overgrowth syndrome caused by abnormality of the NSD1 (nuclear receptor binding SET domain protein 1) gene located on chromosome 5 (Kurotaki et al., 2002) associated with cognitive impairment (mild intellectual disability or borderline functioning) and characterized by a clinical profile, including externalizing behaviors, anxiety and autistic symptoms, that has not been clearly defined yet (Lane et al., 2016).

Social deficit and behavioral inflexibility distinctive of ASD, cognitive impairment and behavioral difficulties characterizing Sotos individuals could have represented additional vulnerabilities in an extreme condition such as COVID-19 pandemic (Eshraghi et al., 2020).

Up to date, little is known regarding the possible consequences of COVID-19 isolation measures on children with special needs. Thus, we aimed to evaluate the short-term effects of pandemic within two fragile paediatric population Sotos Syndrome and ASD.

The study was approved by the local institutional review board (IRB) of the Rome Tor Vergata University Hospital (Register Number # 216.20). Parents of all participants gave written informed consent. Our sample was constituted by children with Sotos Syndrome coming from the clinical database of the Child Psychiatry Unit of the Tor Vergata Hospital. ASD sample comprised participants - matched for age and sex with SOTOS children - included in an active research study at the Child Psychiatry Tor Vergata Unit (Fig. 1).

We investigated the impact of COVID-19 consequences on the adaptive functioning, social skills and problematic behaviors of 12 children affected by Sotos Syndrome, in comparison to a group of 12 children with idiopathic Autism Spectrum Disorder (ASD) by comparing evaluations performed in 2019, before the COVID-19 outbreak (T0), with data collected during the pandemic (from May to September 2020) after the home confinement (T1, re-opening phase) (Table 1).

Specifically, we explored whether after the compulsory home confinement (T1), any behavioral and functioning change was reported by parents using standardized instruments that did not require the in-person administration: Adaptive Behavior Assessment System, Second Edition ABAS-II, Social Responsiveness Scale-SRS, Achenbach Child Behavior Checklist-CBCL.

Changes in standardized instruments were evaluated with the Independent-Samples t-test and the Paired-Samples t-test using SPSS v.23.0 (IBM Corp., Armonk, NY, USA) and results, if not otherwise specified, are given as Means \pm SD.

At baseline, before the Covid-19 outbreak (T0), the two groups of children (Sotos Syndrome and ASD) did not differ in adaptive functioning (ABAS General Adaptive Composite) and behavioral problems (CBCL Total), but a significant difference emerged in terms of social skills measured by the SRS questionnaire (Social Awareness; Social Cognition; Social Communication) (Constantino and Gruber, 2005) (Table 1). Not surprisingly, parents of participants with autism reported more social impairment in their children in comparison to caregivers of participants affected by Sotos Syndrome.

When comparing data collected during the pandemic, from May to September 2020 (T1), with evaluations performed before the COVID-19 outbreak (T0), no significant differences emerged in adaptive skills (ABAS-II), behavioral problems (CBCL), social responsiveness (SRS) of both Sotos and ASD children (Table 1). However, regarding social skills measured by SRS, interestingly, a tendency of worsening in social responsiveness emerged in Sotos participants during the pandemic (Table 1). Worthy of attention, Sotos children with Intellectual Disability presented higher scores on SRS at T1, suggesting a tendency of worse social functioning in this subgroup (M \pm SD Awareness: T0 55.7 \pm 29.5 T1 68.7 \pm 6.2 $p = .46$; Cognition T0 61 \pm 26.8 T1 83.2 \pm 5.4 $p = .19$; Communication T0 61.5 \pm 23.2 T1 84.7 \pm 8.8 $p = .05$; Motivation T0 62.2 \pm 36.8 T1 72.7 \pm 14.6 $p = .50$; Mannerism T0 70.7 \pm 29.6 T1 92.7 \pm 9.5 $p = .32$), in comparison to Sotos individuals without Intellectual Disability (M \pm SD Awareness: T0 54 \pm 5.7 T1 61.7 \pm 13 $p = .24$; Cognition T0 67 \pm 12.5 T1 69 \pm 15 $p = .55$; Communication 68.6 T0 \pm

16.7 T1 67 \pm 20.2 $p = .87$; Motivation T0 66.3 \pm 20.4 T1 66.3 \pm 17.5 $p = 1$; Mannerism T0 75.5 \pm 19 T1 78.5 \pm 26.7 $p = .70$).

Our findings suggest that, within our ASD and Sotos sample, parents did not perceive a significant worsening in the adaptive functioning, problematic behaviors and social skills of their children during the pandemic. However, within Sotos children, the social responsiveness - meant as cognition and communication skills, in addition to the competence of gathering social signals- emerged as the only behavioral feature reported by parents to be slightly impaired after the COVID-19 home confinement, impairment that was influenced by the level of Intellectual Quotient.

Therefore, we may speculate that Sotos children, who are generally less prone to withdrawn in comparison to ASD peers, may have majorly suffered the social isolation due to the compulsory measures for COVID-19 pandemic.

However, our results reflect a parental perception rather than an objective evaluation by clinicians, moreover our findings represent a trend (rather than a significant result) which should be considered with caution and replicated with other studies. Future research on the

Table 1
Characteristics of the sample and main results.

	SOTOS	ASD
N	12	12
Age (M \pm SD)	8.5 \pm 3 yrs	7 \pm 1.5 yrs
M/F	8/4	9/3
IQ (M \pm SD)	71 \pm 27.8	71 \pm 23.9
ID/ NO ID	5/7	7/5
SRS_Social Awareness (M \pm SD)		
• T0	54.6 \pm 17.6	70.4 \pm 9.6
• T1	63.9 \pm 11.2	71.3 \pm 11.3
• Paired Differences T0-T1 (M \pm SD; t_{p})	-9.3 \pm 20.5; -1.4; .18	-.91 \pm 10.04; -.30; .77
SRS_Social Cognition (M \pm SD)		
• T0	64.5 \pm 18.3	81.1 \pm 8.1
• T1	74.6 \pm 13.7	80.5 \pm 8.3
• Paired Differences T0-T1 (M \pm SD; t_{p})	-10.1 \pm 19.2; -1.66; .13	.54 \pm 9.48; .19; .85
SRS_Communication (M \pm SD)		
• T0	65.8 \pm 18.6	82.8 \pm 9.7
• T1	74 \pm 18.4	81.9 \pm 14.6
• Paired Differences T0-T1 (M \pm SD; t_{p})	-8.2 \pm 25.04; -1.04; .33	1 \pm 8.2; .36; .72
SRS_Social Motivation (M \pm SD)		
• T0	64.7 \pm 26.2	80.4 \pm 12.1
• T1	68.9 \pm 15.9	78.6 \pm 13.9
• Paired Differences T0-T1 (M \pm SD; t_{p})	-4.2 \pm 27.6; -.48; .64	1.81 \pm 13.06; .46; .65
SRS_Autistic Mannerisms (M \pm SD)		
• T0	73.6 \pm 22.2	82.4 \pm 12.5
• T1	84.2 \pm 21.9	88 \pm 15.9
• Paired Differences T0-T1 (M \pm SD; t_{p})	-10.6 \pm 27; -1.24; .24	-5.5 \pm 13.9; -1.31; .22
SRS_Tot (M \pm SD)		
• T0	81.4 \pm 15.8	83.9 \pm 9.4
• T1	78.3 \pm 18.1	85.7 \pm 13.1
• Paired Differences T0-T1 (M \pm SD; t_{p})	3.1 \pm 17.6; .56; .59	-1.8 \pm 11.4; -.53; .60
ABAS-II_GAC (M \pm SD)		
• T0	54.7 \pm 18.6	50.7 \pm 11.3
• T1	56.9 \pm 14.4	52 \pm 17.6
• Paired Differences T0-T1 (M \pm SD; t_{p})	-2.2 \pm 17.8; -.37; .71	-1.3 \pm 9.8; -.47; .65
CBCL_TOT (M \pm SD)		
• T0	64.8 \pm 8.1	62.8 \pm 9.5
• T1	62.3 \pm 8.8	64.2 \pm 9.1
• Paired Differences T0-T1 (M \pm SD; t_{p})	2.5 \pm 7.7; 1.12; .29	-1.33 \pm 6.08; -.76; .46

Legend: ASD = Autism Spectrume Disorder; IQ = Intellectual Quotient; M \pm SD = Mean \pm Standard Deviation; yrs = years; ABAS-II = Adaptive Behavior Assessment System, Second Edition; GAC = General Adaptive Composite score; CBCL = Child Behavior Checklist; SRS = Social Responsiveness Scale.

possible impact of COVID-19 on wider sample of children with special needs is necessary in order to correctly define the short and long term effects the pandemic may have determined on this fragile population.

Declaration of Competing Interest

The authors declare no potential conflict of interest.

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