

Cognitive and executive functions, social cognition and sense of coherence in adults with fetal alcohol syndrome

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Background: Primary disabilities in children with fetal alcohol syndrome (FAS) are the results of alcohol's teratogen effect on the fetal brain. Reduced cognitive and executive functions and social cognition are examples of such disabilities. Little is known about primary disabilities in adults with FAS as well as their sense of coherence (SoC). There is thus a need for knowledge about FAS in adulthood. *Aims:* To investigate cognitive and executive functions, social cognition and SoC in adults with FAS. *Methods:* Twenty adults with FAS (mean age: 30 years) were compared with 20 individuals matched on gender and age. Berg's Card-sorting Test-64, the Tower of Hanoi, Raven's Coloured Progressive Matrices, Digit Span, Faux Pas and the Swedish version of Antonovsky's Sense of Coherence Scale (SoC-29) were used. *Results:* The FAS group had a weak SoC and displayed deficits in the neuropsychological tests sensitive to cognitive and executive functions and social cognition. The FAS group's median SoC score was 112, lower than the comparison group's median of 133 ($P < 0.001$). The FAS group had median scores of 29.0 on Raven's Matrices. The median for Digit Span was 5 forwards and 3 backwards, lower than in the comparison group ($P < 0.001$). *Conclusions:* Reduced cognitive and executive functions and impaired social cognition are assumed to have a major impact on life for adults with FAS. We suggest that the findings showing that adults with FAS had a weak SoC, with particularly low scores on the manageability scale, reflect their experiences of living with those primary disabilities. *Clinical implications:* This study may enhance healthcare for individuals prenatally exposed to alcohol. In general, it contributes with knowledge about this group of individuals who need to be more visible in healthcare, and particularly, it demonstrates some of the neuropsychological disabilities they might have.

• *Adulthood, Cognitive and executive functions, Fetal alcohol syndrome, Sense of coherence, Social cognition.*

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Fetal alcohol syndrome (FAS) was first described in 1973 (1) and since then research has demonstrated that alcohol is a potent physical and behavioural teratogen (2). Since 2005 (3), FAS is regarded as the endpoint of the fetal alcohol spectrum disorders (FASD), the umbrella term used today comprising the range of possible physical, behavioural and cognitive effects resulting from prenatal alcohol exposure (4). Disabilities due to the alcohol's teratogen effect on the developing fetal brain (i.e. primary disabilities) are, for instance, reduced cognitive and executive functions (5) and impaired social cognition in children (6). Examples of impaired cognitive functions are overall intellectual performance, learn-

ing and memory (7). The deficits have been linked to damage to the corpus callosum (8), with variance in volume ranging from thinning to complete agenesis, in children with FASD (9). Concerning reduced executive functions, impairments in planning ability, cognitive flexibility and selective inhibition are commonly found in children with FASD (10). Some of these deficits may be explained by decreased growth of the orbitofrontal brain lobe due to the alcohol's teratogen effect (11). Social cognition is related both to executive and cognitive functions as well as to perceptual and emotional processing. Theory of Mind (ToM) specifically involves reasoning about the beliefs and intentions of others (12) and is

considered one part of the social cognition. In a study by Rasmussen and colleagues (13), it was found that children with FASD had difficulties with ToM tasks. Deficits in these processes have been linked to damage in both the frontal lobes and the corpus callosum due to the teratogen effect of alcohol (14).

Being born with FASD may thus mean living with primary disabilities such as reduced cognitive and executive functions and impaired social cognition. These disabilities can explain the commonly occurring difficulties in adaptive behaviours, which require a cognitive capacity that enables integrating information from the environment, selecting appropriate responses and exhibiting adequate behaviour (15). Adaptive behaviours are essential for an individual in order to live an independent life, maintain social relationships, be sensitive to others and become integrated in society (16). The extent to which adaptive behaviours are functioning can be described with what is called the individual's sense of coherence (SoC). This term is meant to consist of three components: meaningfulness, manageability and comprehensibility (17). SoC is an expression of the extent to which: 1) the individual's confidence in life is structured and predictable, 2) the individual's resources are sufficient, and 3) the individual's life is worth the engagement needed (17). In other words, the individual must rely on his/her own ability to plan, solve problems and adapt to changing conditions. These are abilities that require cognitive and executive functions and a functioning social cognition. On the meta-level, it is reasonable to assume that for individuals with FASD, the primary disabilities affect their SoC, i.e. the extent to which they perceive their lives as meaningful, manageable and comprehensible.

Aims

Only a few studies have addressed the long-term outcome of FASD (e.g. 5, 18–23), especially neuropsychological effects. There is thus a need for more knowledge about how life develops for adults who have been prenatally exposed to alcohol. Handling social situations is an example of the demands that increase in adulthood, but social abilities in children with FAS do not seem to improve with age (24). The aim of this study was therefore to investigate cognitive and executive functions, social cognition and SoC in adults with FAS. Based on what is known about children with FASD from previous research (e.g. 5, 18–23), we hypothesized that even in adulthood, individuals with FAS would have reduced cognitive and executive functions and impaired social cognition. As many situations in adulthood require these functions, we assumed that the disabilities would have a major impact on well-being and adaptation to everyday life. Based on this assumption we also hypothesized that the adults with FAS in this study would have a weak SoC.

Methods and materials

Study group with FAS

The study group consisted of 20 individuals, 13 women (65%) and seven men (35%), aged between 18 and 41 years (mean age: 30 years). Inclusion criteria were age at least 18 and having a medical record confirming the FAS diagnosis. For further details on the FAS group, see Table 1.

Almost half the group ($n = 8$) was recruited from, and presented in detail in, a follow-up study on psychosocial outcomes in adults with FAS (25). Remaining participants

Table 1. Demographics on the fetal alcohol syndrome (FAS) group ($n=20$) and the comparison group ($n=20$).

	FAS group	Comparison group
Age, mean \pm standard deviation, years	30 \pm 8	30 \pm 8
Diagnosis		
FAS	100% (20)	0% (0)
ADHD	10% (2)	0% (0)
Highest completed education		
Primary school, 9 years (including special education)	15% (3)	0% (0)
Secondary education (including special education)	85% (17)	5% (1)
Post-secondary education	0% (0)	95% (19)
Income		
Employed	30% (6)	25% (5)
Educational allowance	0% (0)	45% (9)
Social welfare	0% (0)	5% (1)
Disability pension	45% (9)	10% (2)
Supplied by closely related	5% (1)	10% (2)
Other	20% (4)	5% (1)

Data are presented as percentages (%) and numbers of individuals (in brackets). ADHD, attention-deficit hyperactivity disorder.

($n = 12$) were recruited from the Swedish National FAS Organization. The eight participants recruited from the follow-up study on FAS had been diagnosed as children. The diagnostic criteria for FAS (1, 26) required: 1) confirmed alcohol consumption during pregnancy, 2) characteristic pattern of facial anomalies, 3) growth retardation, and 4) neurodevelopmental abnormalities in the central nervous system. For remaining 12 participants, they and/or their parents affirmed that they had medical records confirming their FAS diagnosis.

Comparison group

Participants in the comparison group ($n = 20$) were individually matched with the participants in the study group by age and gender. A research assistant distributed information about the project in public places, such as libraries in the centre of Gothenburg, in order to recruit the comparison group. After reading the information, people willing to participate were asked to provide their contact information. Individuals who matched someone in the study group in terms of gender and age, and who did not have any self-estimated physical or psychological disabilities, were invited to participate. Inclusion criteria were matching someone in the study group by gender and age, being healthy, and voluntarily accepting participation. For further details on the comparison group, see Table 1. The Regional Ethical Review Board in Gothenburg approved the study.

Procedure and measures

All participants gave verbal informed consent and were invited to data collection. Travel expenses, accommodation costs and food were paid for by the research project, and the participants received compensation (two cinema tickets worth approximately €22). An experienced test leader conducted all tests in the order described below. The data collection lasted between 1 and 2 h (1-h lunch break and 15-min coffee break excluded).

To gain a measure of intellectual level in the FAS group, Raven's Coloured Progressive Matrices (RCPM) (27) was used. RCPM assesses cognitive functions in terms of logical thinking with visually presented material. This coloured version of Raven's matrices is designed for children, elderly persons and individuals with mild to severe learning difficulties (27). To obtain the participants' IQ an extensive test had been needed, and to avoid fatigue RCPM was used. This test has previously been used when studying adults with learning disabilities (28). The score range is 0–36 (for further details on RCPM, see Supplementary Appendix available online at <http://informahealthcare.com/doi/abs/10.3109/08039488.2015.1009487>). Due to the target group for the test and as ceiling effects were expected, the comparison group did not conduct it.

The Digit Span Task from the Wechsler Intelligence Scale for adults, fourth edition (29), was used for assessing short-term memory and working memory. Digit Span has previously been used when studying children with FASD (30). Digit Span task is divided into two variants, forwards and backwards, measuring short-term memory and working memory, respectively. The dependent measure was the longest series correctly recalled for each variant. For further details on Digit Span Task, see Supplementary Appendix available online at <http://informahealthcare.com/doi/abs/10.3109/08039488.2015.1009487>.

The computerized Psychology Experiment Building Language (PEBL) version of the Wisconsin Card-Sorting Test (WCST, 31), Berg's Card-Sorting Test-64 (BCST-64, 32), was applied. WCST has previously been used when studying children with FASD (33). It is a neuropsychological test of executive functions, focusing on cognitive flexibility and set-shifting, inhibitory control and inhibition of perseverative responses. The dependent measures were total number of correct responses (of the maximum), the total number of errors, the total number of non-perseverative errors and the total number of perseverative errors. A perseverative error was defined as an incorrect response to a changed or new category that would have been correct for the immediately preceding category. For further details on BCST-64, see Supplementary Appendix available online at <http://informahealthcare.com/doi/abs/10.3109/08039488.2015.1009487>.

The computerized (PEBL) version of the Tower of Hanoi (32) was used. A modified version of Tower of Hanoi has previously been used when studying children with FASD (34). This is a neuropsychological test of executive functions such as cognitive and spatial planning, inhibitory control and inhibition of perseverative responses. Dependent measure was the difference between the number of steps used by the participant and the lowest possible number of steps needed to solve the task. For further details on Tower of Hanoi, see Supplementary Appendix available online at <http://informahealthcare.com/doi/abs/10.3109/08039488.2015.1009487>.

To assess ToM, the Swedish adult version of Faux Pas (35, 36) was applied. To avoid fatigue, a shortened version with eight of the original 20 stories was used. Four stories describing a faux pas, i.e. someone saying something that unintentionally causes embarrassment to another person, alternating with four stories without faux pas, were read to the participant. After each story had been read, the participant was asked questions to determine whether he/she had identified the faux pas. Additionally, two comprehension control questions were asked after each example, to ascertain that the participant had understood the story. The score range was 0–6 for stories with faux pas, 0–2 for stories without faux pas and 0–2 for comprehension-control questions. The maximum

score for the identification of faux pas was 32, and for comprehension-control questions 16. For further details on the Faux Pas Test, see Supplementary Appendix available online at <http://informahealthcare.com/doi/abs/10.3109/08039488.2015.1009487>.

Finally, the Swedish version of Antonovsky's Sense of Coherence Scale (SoC-29) (17) was used to measure SoC. This self-rating questionnaire uses a 7-point scale anchored by two statements, on which the participants respond to 29 items. The general SoC is measured based on three domains: manageability, comprehensibility and meaningfulness. The SoC-29 version has 10 items linked to the manageability subscale (maximum score 70), 11 items linked to the comprehensibility subscale (maximum score 77) and eight items linked to the meaningfulness subscale (maximum score 56). Total SoC-29 scores thus range from 29 to 203, with higher scores reflecting stronger SoC. This self-rating questionnaire SoC-29 was turned into an interview to avoid outcome being dependent on reading ability and to gain the opportunity to explain wordings if necessary.

Data analyses

AS data was not normally distributed, median value differences between the FAS group and the comparison group were tested using the non-parametric Mann-Whitney *U*-test. Tests were two-tailed, a $P < 0.05$ was regarded as significant and effect sizes were calculated. Statistical analyses were performed with the SPSS software package, version 22.

Results

Table 2 shows the results in detail. In Raven's Coloured Matrices, testing level of intellectual functioning, the median in the FAS group, was 29 of a maximum of 36. On Digit Span forwards, the median in the FAS group was 5, significantly lower than the comparison group's median of 7 ($U = 35.5$, $z = -4.47$, $P < 0.001$, $r = -0.72$). In Digit Span backwards, the median in the FAS group was 3, which was significantly lower than 5 in the comparison group ($U = 20.5$, $z = -4.93$, $P < 0.001$, $r = -0.79$).

In the first test of executive functions, Berg's Card-Sorting Test, the median number of total correct responses in the FAS group was 33.0 (maximum 64). In the comparison group, the median was higher at 51.5 ($U = 21.5$, $z = -4.74$, $P < 0.001$, $r = -0.76$). In the FAS group, 64.5% of the errors were non-perseverative with a median of 20, significantly higher than in the comparison group, in which the median number of non-perseverative errors was 5.5 ($U = 46.5$, $z = -4.05$, $P < 0.001$, $r = -0.65$). The median number of perseverative errors, 7 in the FAS group and 6 in the comparison group, did not differ ($p = \text{n.s.}$). See Table 1 for further details. Tower of Hanoi showed that the FAS group required significantly more steps to solve the task than the comparison group ($U = 102.0$, $z = -2.65$, $P = 0.007$, $r = -0.42$).

The Faux Pas median score for identification of faux pas in the FAS group was 25 (of a maximum of 32). The median in the comparison group, 31, was significantly higher ($U = 62.0$, $z = -3.63$, $P < 0.001$, $r = -0.58$).

Table 2. Results of Raven's Coloured Progressive Matrices, Digit Span, Berg's Card-Sorting Test, Tower of Hanoi, Faux Pas and sense of coherence tests in the fetal alcohol syndrome (FAS) group and the comparison group.

	FAS group ($n = 20$)		Comparison group ($n = 20$)
	Median	Median	
Raven's Coloured Progressive Matrices (Max 36)	29.0		
Digit Span			
Forwards	5	7	$U = 35.5$, $z = -4.47$, $P < 0.001$, $r = -0.72$
Backwards	3	5	$U = 20.5$, $z = -4.93$, $P < 0.001$, $r = -0.79$
Berg's Card-Sorting Test-64			
Total correct responses (Max 64)	33.0	51.5	$U = 21.5$, $z = -4.74$, $P < 0.001$, $r = -0.76$
Total errors	31.0 (mean 30.4)	12.5 (mean 12.6)	$U = 21.5$, $z = -4.74$, $P < 0.001$, $r = -0.76$
Non-perseverative errors	20.0 (mean 23.0)	5.5 (mean 5.8)	$U = 46.5$, $z = -4.05$, $P < 0.001$, $r = -0.65$
Perseverative errors	7.0 (mean 7.4)	6.0 (mean 6.8)	$U = 186.5$, $z = -0.099$, $P = 0.923$, $r = -0.02$
Tower of Hanoi			
Difference (participant vs. shortest possible; the lower the better)	28.0	13.5	$U = 102.0$, $z = -2.65$, $P = 0.007$, $r = -0.42$
Faux Pas			
Total (maximum score = 32)	25.0	31.0	$U = 62.0$, $z = -3.63$, $P < 0.001$, $r = -0.58$
Comprehension-control questions (maximum score = 16)	16.0	16.0	$U = 160.0$, $z = -2.08$, $P = 0.29$, $r = -0.33$
Sense of coherence			
Total (maximum score = 203)	112.0	133.0	$U = 91.0$, $z = -2.78$, $P = 0.005$, $r = -0.45$
Manageability (maximum score = 70)	36.0	50.0	$U = 60.0$, $z = -3.66$, $P < 0.001$, $r = -0.59$
Comprehensibility (maximum score = 77)	34.0	40.0	$U = 141.5$, $z = -1.36$, $P = 0.18$, $r = -0.22$
Meaningfulness (maximum score = 56)	37.5	41.0	$U = 129.0$, $z = -1.72$, $P = 0.09$, $r = -0.28$

The comprehension-control scores ranged from 14 to 16 (maximum = 16) in the FAS group and the median score in both groups was 16. See Table 1 for further details.

The total score reflecting global SoC differed between the groups: $U=91.0$, $z=-2.78$, $P=0.005$, $r=-0.45$. The median manageability subscale score in the FAS group was 36 (of a maximum of 70), lower than in the comparison group where the median was 50 ($U=60.0$, $z=-3.66$, $P<0.001$, $r=-0.59$). The median comprehensibility and meaningfulness subscale scores did not differ between the groups. See Table 1 for further details.

Discussion

This study showed that individuals with FAS, even in adulthood, may have reduced cognitive and executive functions and impaired social cognition, which also supported our hypothesis. Furthermore, our findings supported the hypothesis that they also would have a weak SoC.

Previous studies (e.g. 7) have shown that children with FASD have cognitive disabilities, such as impairments in intellectual performance, learning and memory. The median IQ is 86, range 29–126 in individuals with FASD aged 6–51 (21). In line with these previous studies (7, 21), the FAS group in this study scored low on RCPM, which was interpreted as indicating a low level of intellectual function. Studies in young adults with FASD have reported memory deficits (23) and deficits in the ability to maintain and manipulate information in the working memory (22). In this study the Digit Span Task was used to assess aspects of memory: short-term and working memory. The adults with FAS performed more poorly than the comparison group on both measures. It was clear that Digit Span backwards, which requires more integration and coordination of cognitive functions, was more difficult than Digit Span forwards, which is not as complex. These findings, indicating memory deficits in adults with FAS, suggested that several everyday situations may be difficult to manage.

Previous studies in children with FASD (e.g. 10) have shown that reduced executive functions are common. Both tests of executive functions used in this study (i.e. BCST-64 and Tower of Hanoi) revealed deficits in the FAS group. The adults with FAS had more difficulties solving tasks requiring cognitive and spatial planning, flexibility, set-shifting and inhibitory control than the comparison group. The most common comorbidity with FASD is attention deficit-hyperactivity disorder (ADHD) occurring in 48% of individuals with FASD (37). A previous study on children with ADHD (38) as well as a study on children with FASD (39) showed an increase in perseverative responses in tests of executive functions. Based on these results, we expected perseverative

responses to be common in the FAS group. Perseverative responses indicate difficulties in set-shifting and inhibitory control, i.e. executive function disabilities. However, non-perseverative errors (i.e. an unspecified failure to solve the task) were the most frequent in this FAS group. This might be because solving the task was too difficult for this group, requiring a higher level of cognitive function, a suggestion that may be supported by their low number of correct responses. Despite the FAS group appearing to have understood the instructions, they seemed to have difficulties grasping the rules and consequently failed to reach even the level of making perseverative errors; their errors were random. In contrast, the comparison group made a low total number of errors, with non-perseverative and perseverative errors equally distributed. Importantly, two (10%) of the adults in the FAS group had a diagnosed ADHD and there could be more in the group, undiagnosed. The results from the tests of executive functions in this study may be related to FAS specifically, and to comorbid attention deficits and impaired inhibitory control.

A previous review of social skills deficits (4) reported that children with FASD exhibited social difficulties, particularly impairments in socially appropriate interactions, poor social judgment and difficulties understanding social cues. In this study, the adults' social cognition, defined as their ToM, was investigated. ToM is considered the part of social cognition dealing with reasoning about the beliefs and intentions of others (12). Data from Faux Pas showed clear differences between the groups. The FAS group had difficulties identifying situations that unintentionally caused embarrassment to another person, indicating their uncertainty about social codes and what is considered appropriate in social interactions. This indicates problems in real life, as ToM is closely related to community functioning. It should be emphasized that there is a need for psychosocial interventions in this area. In addition, based on our results, we suggest that future studies are needed to show how social cognition functions are affected in adulthood by prenatal alcohol exposure.

SoC is a way of describing a sense of meaningfulness, manageability and comprehensibility in life (17). A strong SoC means being confident that life is structured and predictable and that one's own resources are sufficient (17). In other words, the individual must rely on the own abilities for planning, problem solving and adaptation to changing conditions, abilities requiring a certain level of cognitive and executive functions, as well as of social cognition. However, the FAS group scored significantly lower on the SoC-29 than the comparison group. On the other hand, although there were significant differences between the groups on the manageability subscale, there were no differences between the groups on the comprehensibility and meaningfulness

subscales. This may be interpreted as although the adults with FAS cannot always manage their lives; they nonetheless find life quite comprehensible and meaningful.

Method discussion and limitations

Data in this study were collected with modified tests—shortened versions, tests for individuals with learning difficulties etc.—therefore, a comparison group was required. A limitation is that we chose to use healthy individuals matched on gender and age. Instead, individuals matched on IQ or differences in environmental influences or with comparable disabilities as the FAS group could have been chosen. On the other hand, this might have led to other design considerations and counterintuitive findings about neurocognitive function (40). Data from the comparison group might be regarded as reference values. Another limitation in this study is that the measurements used do not distinguish specifically what dysfunctions are FAS specific. Impaired executive functions and intellectual level may influence measurements that are primarily intended to measure something else, such as ToM. However, the results in this study provide a picture of how primary disabilities may appear in individuals with FAS in adulthood.

All in the FAS group were well aware of their FAS diagnosis and had foster parents who also were familiar with the problems of FAS. An inclusion criterion was having a medical record certifying the FAS diagnosis, which all subjects in the FAS group had. A limitation in the study is that it was not required to present the medical records. However, the adults with FAS themselves and their foster parents affirmed having medical record certifying the FAS diagnosis, which they could order on request.

Another limitation in this study is that measures used were not validated for studies on adults with FASD. As measures for individuals with disabilities are lacking, there is a need for development of such measures. The IQ is unknown in the FAS group, but level of intellectual function was obtained by use of RCPM (27), a test previously used when studying adults with Down syndrome, who like many individuals with FASD have learning disabilities (28). As adults with FASD may have a low level of intellectual function (21), from an ethical perspective and out of respect for the FAS group, this was considered a suitable choice of test. Digit Span, BCST-64 and Tower of Hanoi were used and have been used in previous studies on children with FASD (30, 33, 34). In Faux Pas, the participants' high scores on the comprehension-control questions indicated an appropriate difficulty level. There were no floor or ceiling effects, and the participants understood the instructions, implying that measures used were appropriate. In addition, SoC-29 was made as an interview to give the adults with FAS opportunity to ask if there was something they had not understood.

In conclusion, it is previously known that children with FASD have reduced cognitive and executive functions (5) and impaired social cognition (6), but further research is needed on how neuropsychological disabilities develop from childhood and how they affect life in adults prenatally exposed to alcohol. The applied measurements in this study showed that individuals with FAS even in adulthood may have reduced cognitive and executive functions and impaired social cognition. As mentioned above, these disabilities can be assumed to have a major impact on life, and we suggest that the weak SoC found in this study, with particularly low scores on the manageability scale, may be an expression of an adult's experiences of living with these disabilities.

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Supplementary material available online

Supplementary Appendix