Research in Developmental Disabilities 63 (2017) 151–159

Contents lists available at ScienceDirect

Research in Developmental Disabilities

Substance use in individuals with mild to borderline intellectual disability: A comparison between self-report, collateral-report and biomarker analysis

Joanneke E.L. VanDerNagel[a,b,c,*], Marion Kiewik[a,c], Marike van Dijk[a,b], Robert Didden[d,e], Hubert P.L.M. Korzilius[f], Job van der Palen[g,h], Jan K. Buitelaar[i], Donald R.A. Uges[j], Remco A. Koster[j], Cor A.J. de Jong[a,d]

[a] Nijmegen Institute for Scientist-Practitioners in Addiction, Radboud University, P.O. Box 6909, 6503 GK Nijmegen, The Netherlands
[b] Tactus Addiction Treatment, Raiffeisenstraat 75, 7514 AM Enschede, The Netherlands
[c] Aveleijn, Grotestraat 260, 7622 GW Borne, The Netherlands
[d] Behavioural Science Institute, Radboud University, P.O. Box 9104, 6500 HE Nijmegen, The Netherlands
[e] Trajectum, P.O. Box 40012, 8004 DA Zwolle, The Netherlands
[f] Institute for Management Research, Radboud University, Thomas van Aquinostraat 5, 6525 GD Nijmegen, The Netherlands
[g] Department of Research Methodology, Measurement and Data Analysis, Faculty of Behavioral Sciences, University of Twente, P.O. Box 217, 7500 AE Enschede, The Netherlands
[h] Medical School Twente, Medisch Spectrum Twente, Ariënsplein 1, 7511 JX Enschede, The Netherlands
[i] Department of Cognitive Neuroscience, Radboud University Medical Centre, P.O. Box 9101 (204), 6500HB Nijmegen, The Netherlands
[j] University of Groningen, University Medical Center Groningen, Department of Clinical Pharmacy and Pharmacology, Clinical Pharmaceutical and Toxicological Laboratory, P.O. Box 30.001, 9700 RB Groningen, University of Groningen, The Netherlands

ARTICLE INFO

Article history:
Received 14 November 2015
Received in revised form 15 April 2016
Accepted 16 April 2016
Available online 25 April 2016

Keywords:
Substance use
Intellectual disability
Assessment
Questionnaire
Biomarker
Self-report
Collateral-report
Smoking
Alcohol
Illicit drugs

ABSTRACT

Background and aims: Individuals with mild or borderline intellectual disability (MBID) are at risk of substance use (SU). At present, it is unclear which strategy is the best for assessing SU in individuals with MBID. This study compares three strategies, namely self-report, collateral-report, and biomarker analysis.
Methods and procedures: In a sample of 112 participants with MBID from six Dutch facilities providing care to individuals with intellectual disabilities, willingness to participate, SU rates, and agreement between the three strategies were explored. The Substance use and misuse in Intellectual Disability – Questionnaire (SumID-Q; self-report) assesses lifetime use, use in the previous month, and recent use of tobacco, alcohol, cannabis, and stimulants. The Substance use and misuse in Intellectual Disability – Collateral-report questionnaire (SumID-CR; collateral-report) assesses staff members' report of participants' SU over the same reference periods as the SumID-Q. Biomarkers for SU, such as cotinine (metabolite of nicotine), ethanol, tetrahydrocannabinol (THC), and its metabolite THCCOOH, benzoylecgonine (metabolite of cocaine), and amphetamines were assessed in urine, hair, and sweat patches.
Results: Willingness to provide biomarker samples was significantly lower compared to willingness to complete the SumID-Q ($p < 0.001$). Most participants reported smoking, drinking alcohol, and using cannabis at least once in their lives, and about a fifth had ever used stimulants. Collateralreported lifetime use was significantly lower.

* Corresponding author.
E-mail address: j.vandernagel@tactus.nl (J.E.L. VanDerNagel).

http://dx.doi.org/10.1016/j.ridd.2016.04.006

However, self-reported past month and recent SU rates did not differ significantly from the rates from collateral-reports or biomarkers, with the exception of lower alcohol use rates found in biomarker analysis. The agreement between self-report and biomarker analysis was substantial (kappas 0.60–0.89), except for alcohol use (kappa 0.06). Disagreement between SumID-Q and biomarkers concerned mainly over-reporting of the SumID-Q. The agreement between SumID-CR and biomarker analysis was moderate to substantial (kappas 0.48 – 0.88), again with the exception of alcohol (kappa 0.02). In this study, the three strategies that were used to assess SU in individuals with MBID differed significantly in participation rates, but not in SU rates. Several explanations for the better-than-expected performance of self- and collateral-reports are presented. We conclude that for individuals with MBID, self-report combined with collateralreport can be used to assess current SU, and this combination may contribute to collaborative, early intervention efforts to reduce SU and its related harms in this vulnerable group.

What this paper adds?

This paper is the first to compare three strategies to assess substance use among individuals with mild to borderline intellectual disability: self-report with a questionnaire developed for this population (i.e., SumID-Q), collateral-report by staff members, and biomarker analysis of urine, hair and sweat patch samples. We found that biomarker analysis was of limited additional value compared to self-report or collateral-report in the assessment of substance use, especially given the additional costs of and lower willingness to participate in biomarker analysis.

1. Introduction

Individuals with mild to borderline intellectual disability (MBID) (IQ 50–85, American Psychiatric Association [APA], 2013) are at risk of substance use (SU) and substance use disorder (SUD; Carroll Chapman & Wu, 2012; van Duijvenbode et al., 2015). For epidemiological purposes as well as case identification in clinical settings, several strategies may be used to assess SU: self-report, collateral-report (report by professionals, family members, or peers on participant's use), and biomarker analysis of urine, hair, or sweat patches. At present, it is unclear which strategy is most suitable for the assessment of SU in individuals with MBID.

In individuals without MBID, self-report, collateral-report, and biomarker analysis have been compared in a range of studies (see e.g., Akinci, Tarter, & Kiriski, 2001; Connors & Maisto, 2003; de Beaurepaire et al., 2007; Fendrich, Johnson, Wislar, Hubbell, & Spiehler, 2004). In epidemiological studies, collateral-report has yielded similar or lower estimations of SU compared to self-report (e.g., Connors & Maisto, 2003; Stasiewicz et al., 2008). However, self-report has yielded lower SU rates compared to biomarker analysis (see e.g., Fendrich et al., 2004). Specifically, high rates of under-reported illicit drug use and alcohol use (i.e., no self-reported SU while biomarker analysis was positive) have been found, combined with lower rates of over-reported SU (i.e., self-reported SU while biomarker analysis was negative) (Akinci et al., 2001; de Beaurepaire et al., 2007). For instance, in patients in a psychiatric hospital, de Beaurepaire et al. (2007) found that 52% under-reported and 14% over-reported illicit drug use and 56% under-reported and 23% over-reported alcohol use compared to the biomarker analysis. For tobacco use, the rates of under-reporting were much lower (1–10%; Rebagliato, 2002).

In individuals with MBID, both collateral-report and self-report have been used to estimate the rates of SU (see Carroll Chapman & Wu, 2012; van Duijvenbode et al., 2015). However, both strategies have shortcomings. For instance, some evidence suggests that collateral-report is more sensitive to more severe cases of SU in MBID (VanDerNagel, Kiewik, Buitelaar, & De Jong, 2011). Additionally, self-reported SU may be even more biased in individuals with MBID, especially when questionnaires not adapted to the needs of this group are used (McGillicuddy, 2006; van Duijvenbode et al., 2015).

Given the potential for bias related to self-report and collateral-report, biomarker analysis seems appealing as a more objective measurement of SU in individuals with MBID. Nevertheless, its usability and validity depend on several factors. First, false positive testing can occur due to environmental contamination (e.g., second hand smoking, or accidental transfer of the substance to the sampling site), the use of prescribed medication, or the use of products such as baby wash (Brahm, Yeager, Fox, Farmer, & Palmer, 2010; Cotten, Duncan, Burch, Seashore, & Hammett-Stabler, 2012). Second, false negative testing can occur due to tampering with the sample ('cheating the drug test') or dilution of the substance in incidental use (Fendrich et al., 2004; Hoiseth et al., 2008). Third, both the window of detection of SU and the threshold of detectable use vary across different types of biomarker analysis. For instance, hair analysis is suitable to detect SU over long periods, depending on hair length (Cooper, Kronstrand, & Kintz, 2012; Koster, Alffenaar, Greijdanus, VanDerNagel, & Uges, 2014a). Drug patches absorb traces of substances and their metabolites through the skin during the time they are worn, which can be up to one week (Koster, Alffenaar, Greijdanus, VanDerNagel, & Uges, 2014b). Urine analysis provides information about more recent use based on the pharmacokinetic properties of the substance of interest from days or even hours (cocaine, alcohol) to weeks (cannabis, nicotine) before sampling (Moeller, Lee, & Kissack, 2008; Wojcik & Hawthorne, 2007). Finally,

Table 1
Participant characteristics ($n = 112$).

	M(*SD*)	*n* (%)
Gender		
Male		75 (66.4)
Age (in years)	39.2 (15.9)	
Level of ID[a]		
Mild (IQ 50–70)		73 (65.2)
Borderline (IQ 71–85)		39 (34.8)
Mean IQ ($n = 79$)[a]	64.9 (8.7)	
ID Service		
Residential facility		22 (20.2)
Community facility		40 (36.7)
Outpatient		50 (44.6)

[a] Based on information provided by the ID facility.

feasibility of large scale biomarker testing and participation rates are limited, since hair and sweat patch analysis is costly and biomarker sampling is to some extent seen as invasive and intrusive (Fendrich et al., 2004).

In short, all three strategies to assess SU—self-report, collateral-report, and biomarker analysis—have their strengths and weaknesses, and it is unclear how they perform when applied to individuals with MBID. In this study, we compared (a) self-reported SU measured by the *Substance use and misuse in Intellectual Disability—Questionnaire* (*SumID-Q*, VanDerNagel, Kiewik, Van Dijk, De Jong, & Didden, 2011; VanDerNagel, Kemna, & Didden, 2013), (b) collateral-reported SU measured by the *Substance use and misuse in Intellectual Disability Collateral-report questionnaire* (*SumID-CR*), and (c) biomarker analysis of urine, hair, and sweat patches in individuals with MBID. Our main objectives were to compare willingness to participate in each strategy, to assess SU rates across the three strategies, and to explore agreement and disagreement among them. Regarding willingness to participate, we hypothesized that (1) there would be greater willingness to participate in *SumID-Q* than with biomarker analysis, and (2) willingness to participate in hair and sweat patch sampling would be lower compared to willingness to participate in urine sampling. Furthermore, we hypothesized that (3) SU rates measured with biomarkers would be higher than those measured with self-report and collateral-report, (4) SU rates measured with biomarker samples with a large window of detection (i.e., hair and sweat patch analysis) would be higher than those measured with samples with a smaller window of detection (i.e., urine analysis), and (5) self-reported lifetime SU would be higher compared to collateral-reported lifetime SU, especially for illicit substances. Regarding the agreement and disagreement between the three strategies, we hypothesized that there would be (6) moderate agreement between *SumID-Q* and biomarker analysis as well as between *SumID-CR* and biomarker analysis, (7) under-reporting of SU when *SumID-Q* and *SumID-CR* are compared with biomarkers, and (8) under-reporting of SU when collateral-report is compared to self-report.

2. Methods

2.1. Participants

Between November 2011 and December 2012, six organizations of the Dutch Association of Healthcare Providers for People with Disabilities invited 135 individuals with intellectual disabilities (ID) who had access to substances to participate. Two individuals refused to participate for unspecified reasons, three had no MBID, one individual withdrew consent during the *SumID-Q* interview and caregivers of two individuals withdrew consent to participate. Whilst 127 individuals (94%) completed the *SumID-Q*, 15 individuals (11% of the original sample) refused to provide any biomarker samples and as such were excluded from the study, leaving a total sample of 112 (83%) participants (see Table 1 for participant characteristics).

Collateral-report of SU was sought for the remaining 112 participants from the staff members, who were all involved in the day-to-day care of the participants. This resulted in 97 completed *SumID-CR* questionnaires (87%). A power analysis (with G*Power Version 3.1.92) assessing the ability to detect a statistically significant difference showed that testing 2×2 contingency tables with this sample size would yield a power of 0.89 at a medium effect size ($w = 0.30$, Cohen, 1992) and an α of 0.05.

2.2. Measurements

2.2.1. Self-report

The *SumID-Q* (VanDerNagel, Kiewik, Van Dijk et al., 2011; VanDerNagel et al., 2013) was used to assess lifetime, last month, and recent use of tobacco, alcohol, cannabis, and stimulants (cocaine and amphetamines), as they are the substances used most often by individuals with MBID residing in Dutch facilities (VanDerNagel, Kiewik, Buitelaar et al., 2011). To decrease self-report bias, the questionnaire comprised (1) adapted item structure and wording, (2) visual aids, and (3) a step-by-step non-confrontational approach (VanDerNagel et al., 2013). The *SumID-Q* assesses participants' familiarity with substances (by

showing a standardized set of substance-related pictures and asking participants to identify them), substance knowledge and attitudes, and SU in participants' social environment (e.g., peers', family members', and professional caregivers' SU). Although the results related to these items are not directly relevant to this study, they serve as a means to discuss SU openly. The participants were then asked 'Have you ever use this?' (lifetime use; yes/no response format) for each substance, and if so, 'Once or more often?' and 'How old were you (the first time)?'. Subsequently, SU patterns (frequency, quantity, circumstances) and past month and recent SU (yes/no) were assessed. To enable comparison of *SumID-Q* responses with urine samples, 'recent use' was defined differently for cannabis and tobacco (two weeks) and for alcohol and stimulants (two days). Participants who did not recognize any pictures of a certain type of substance were classified as self-reported 'non-users' of this substance. The administration of the *SumID-Q* took 45–60 min.

2.2.2. Collateral-report

The *Substance use and misuse in Intellectual Disability Collateral-report* (*SumID-CR*) was used to assess staff members' rating of participants' SU. For each substance (i.e., tobacco, alcohol, cannabis, and stimulants) and reference period (i.e., lifetime, previous month, and recent), three response options were given: 'probably used'/'probably did not use'/'do not know'. Responses in the latter category were excluded from the analysis. The collateral-report was completed within 10–15 min.

2.2.3. Biomarkers (urine, hair, and sweat patches)

Urine samples were analyzed using Gas Chromatography (GC) and immunoassay. Liquid Chromatography, coupled with tandem Mass Spectrometry (LC–MS/MS), was used to analyze relevant substances in hair and sweat patches (Koster et al., 2014a, 2014b). Tobacco use was measured by cotinine in urine, hair and sweat patches. Alcohol use was measured by ethanol in urine, but for technical reasons not in hair and sweat patches (Kintz & Nicholson, 2014; Koster et al., 2014a, 2014b). Cannabis use was measured by THC-COOH in urine and by THC in hair. THC and THC-COOH could not be measured in sweat patches because of technical issues, which have also been reported in previous studies (e.g., de la Torre & Pichini, 2004). In urine, sweat patches, and hair, amphetamines and benzoylecgonine, the main metabolite of cocaine, were used to measure stimulant use. Cut-off concentrations from our laboratory (Koster et al., 2014a, 2014b) and from well-established references (Bush, 2008; Cooper et al., 2012; Mayo Medical Laboratories, 2015) were used to classify test results as positive or negative. Detection of a substance with a concentration below its cut-off value was classified as negative. Recent SU according to biomarkers was defined as a positive urine test for each substance, and monthly use according to biomarkers was defined by any positive biomarker analysis for each substance. Unfortunately, two hair samples, two patches, and a urine sample were lost in the mail, six hair samples were not analyzed for technical reasons, and three sweat patch sets were not worn correctly. Therefore, our biomarker analysis included 104 urine samples, 44 hair samples (five without corresponding urine samples), and 27 sets of sweat patches.

2.3. Procedure

A trained research assistant visited participants at their residence or another venue of their choice. Following a video presentation explaining the content and aim of the study, informed written consent was obtained from participants as well as their legal representatives or primary caregiver. Participants with sufficient scalp hair length ($\geq$2 cm, $\pm$ 0.79 in.) completed the *SumID-Q* and provided a urine sample. During a second visit one month later, they provided a hair sample (diameter 0.5 cm, taken 1 cm from the scalp to minimize contamination). Participants with insufficient scalp hair length to provide a hair sample (<2 cm) were asked to wear a transdermal patch and change it weekly (this was supported by their primary caregiver). After wearing sweat patches for four consecutive weeks, participants completed the *SumID-Q* and provided a urine sample. Samples were sent to the laboratory by regular postal service and stored until the analysis. Primary caregivers completed the *SumID-CR* on the same day as the *SumID-Q* interview. Participants received a small gift (worth approximately €2,50/$2,70) after the second visit. The Medical Ethical Review Board Twente (NL27716.044.09) approved the study, which was conducted in accordance with the Declaration of Helsinki (World Medical Association, 2013).

2.4. Data analysis

The data were analyzed with IBM® SPSS® Statistics (version 21). Differences in willingness to participate between the *SumID-Q* interview and biomarker sampling and between different biomarker sampling methods were quantified using Chi-square tests. To test whether SU rates varied across various assessment strategies, Cochran's *Q* test statistic was calculated. To test the agreement corrected for chance agreement between the *SumID-Q*, the *SumID-CR*, and biomarker analysis, we calculated Cohen's kappas and used Landis and Koch's (1977) interpretation. To analyze under- and over-reports, we calculated percentages, comparing *SumID-Q* with biomarker analysis as the criterion as well as *SumID-CR* with the *SumID-Q* as the criterion. Statistical significance was set at an alpha level of 0.05. In case of post-hoc tests, Bonferroni correction was applied for multiple comparisons, by dividing the alpha level by the number of comparisons.

Table 2
SU rates according to *SumID-Q*, *SumID-CR* and biomarker analysis.

	Self-report ($n = 112$)	Collateral report($n = 97$)	Biomarkers[a]	$Q(1)$	p
Lifetime use					
Tobacco	86.6%	78.4%	NA	6.4	0.011
Alcohol	97.3%	88.7%		4.5	0.035
Cannabis	59.8%	41.2%		12.3	< 0.001
Stimulants	21.4%	17.5%		0.4	0.527
Monthly use			($n = 112$)	$Q(2)$[b]	p
Tobacco	68.8%	67.0%	70.5%	0.2	0.905
Alcohol	66.1%	67.0%	0.9%[c]	114.9	< 0.001
Cannabis	14.3%	13.4%	9.8%	3.2	0.199
Stimulants	2.7%	1.0%	2.7%	0.5	0.799
Recent use			($n = 104$)		
Tobacco	66.1%	64.9%	72.1%	6.5	0.039
Alcohol	25.9%	57.7%	1.0%	70.9	< 0.001
Cannabis	13.4%	11.3%	10.6%	2.4	0.301
Stimulants	0%	0%	1.0%	–	–

[a] Recent use based on 104 urine samples, monthly use based on 104 urine, 44 hair, and 27 sweat patch samples.
[b] Cochran's Q based on full data.
[c] Based on urine samples ($n = 104$) only.

3. Results

3.1. Differences between SumID-Q and biomarker analysis in willingness to participate

In addition to the fifteen individuals who refused to provide any biomarkers, several were not willing to participate in either the urine analysis or the hair/sweat patch analysis. This resulted in significantly lower biomarker participation (77% for urine sampling, 62% for hair or sweat patch sampling) compared to *SumID-Q* participation (94%, $\chi^2(2, n = 135) = 39.95$, $p < 0.001$). Post hoc tests revealed that willingness to participate in the *SumID-Q* was significantly higher compared to both the urine testing ($\chi^2(1, n = 135) = 15.85$, $p < 0.001$) and the combined hair or sweat patch testing (χ^2 $(1, n = 135) = 40.10$, $p < 0.001$). Willingness to participate was significantly lower in hair or sweat patch testing compared to urine testing (χ^2 $(1, n = 135) = 7.00$ $p < 0.001$).

3.2. SU rates according to the three measurements

Most participants reported smoking, drinking alcohol, and using cannabis at least once in their lives, and about a fifth had ever used stimulants. Self-reported lifetime SU rates were significantly higher compared to collateral-reported SU rates for smoking, alcohol use, and cannabis use but not for stimulant use (Table 2).

No significant differences in past month SU rates were found among self-report, collateral-report, and biomarker analysis, except for the lower alcohol use rates found with biomarkers (Table 2). Hair and sweat patch analysis identified four users of tobacco and two users of stimulants who had no positive urine samples. No participants tested positive for THC in their hair. As such, previous month SU rates based on all biomarkers did not differ significantly from those found with urine analysis alone ($Q(1) = 3.00$, $p = 0.083$ for smoking; $Q(1) = 0.00$, $p = 1$ for cannabis use; and $Q(1) = 2.00$, $p = 0.157$ for stimulant use).

Recent SU rates differed significantly across the three strategies for tobacco use and alcohol use (Table 2). Post hoc tests showed that self-reported recent smoking did not differ significantly from either collateral-report or biomarkers ($Q(1) = 1$, $p = 0.317$, and $Q(1) = 3$, $p = 0.083$, respectively). Collateral-reported recent smoking was not significantly lower compared to recent smoking according to biomarkers when Bonferroni correction was applied ($Q(1) = 4$, $p = 0.046$). Post hoc tests further showed that recent alcohol use rates found with urine analysis were lower compared to those found with the *SumID-Q* ($Q(1) = 25$, $p < 0.001$) and *SumID-CR* ($Q(1) = 49$, $p < 0.001$). Additionally, *SumID-CR* recent alcohol use rates were significantly higher than *SumID-Q* rates ($Q(1) = 26.5$, $p < 0.001$).

3.3. Agreement among the three measurements

The agreement among *SumID-Q*, *SumID-CR*, and biomarker analysis varied across substances and types of biomarkers (Table 3). Kappas were substantial for tobacco use and very low for alcohol use. Self-reported recent cannabis use agreed substantially with the results of urine analysis, whilst collateral-reported recent cannabis use agreed moderately. Agreement of self-reported and collateral-reported cannabis use in the previous month with hair analysis was non-significant. Agreement of self-reported and collateral reported stimulant use with biomarkers could not be assessed reliably due to the low rate of stimulant use among participants.

Table 3
Agreement between *SumID-Q* and *SumID-CR* and biomarker analysis.[a]

	n	TP	FP	FN	TN	kappa	95% CI
Tobacco							
Urine Cotinine vs							
SumID-Q	104	70	0	5	29	0.886	0.789–0.984
SumID-CR	86	59	0	4	23	0.888	0.780–0.995
Hair Cotinine vs							
SumID-Q	44	20	9	0	15	0.602	0.371–0.834
SumID-CR	38	16	7	0	15	0.643	0.405–0.882
Sweat Patch Cotinine vs							
SumID-Q	27	18	1	2	6	0.724	0.427–1
SumID-CR	26	17	1	2	6	0.719	0.421–1
Any Biomarker Cotinine vs							
SumID-Q	112	72	5	7	28	0.747	0.592–0.866
SumID-CR	95	61	4	5	25	0.779	0.642–0.916
Alcohol							
Urine Alcohol vs							
SumID-Q	104	1	25	0	78	0.057	0–0.379
SumID-CR	82	1	49	0	32	0.016	0–0.191
Cannabis							
Urine THC vs							
SumID-Q	104	11	4	0	89	0.824	0.656–0.993
SumID-CR	82	6	5	3	68	0.545	0.246–0.845
Hair THC vs							
SumID-Q	44	0	6	0	38	0	0–0.744
SumID-CR	36	0	5	0	31	0	0–0.813
Any biomarker THC vs							
SumID-Q	112	11	5	0	96	0.79	0.615–0.966
SumID-CR	88	6	7	3	72	0.483	0.211–0.755
Stimulant use							
Urine Stimulants vs							
SumID-Q	104	0	0	1	103	0	0–1
SumID-CR	83	0	0	0	83	–	–
Hair Stimulants vs							
SumID-Q	44	1	0	1	42	0.656	0–1
SumID-CR	36	0	0	1	35	0	0–1
Sweat Patch Stimulants vs							
SumID-Q	27	0	1	0	26	0	0–1
SumID-CR	24	0	0	0	24	–	–
Any biomarker Stimulants vs							
SumID-Q	112	1	2	2	107	0.315	−0.99
SumID-CR	91	0	1	1	89	−0.011	−0.031

TP—true positive, FP—false positive, FN—false negative, TN—true negative.
– Because of empty cells these values cannot be calculated.
[a] Using biomarker analysis as reference.

3.4. Under- and over-reporting of SumID-Q and SumID-CR compared with biomarkers

Disagreement between *SumID-Q* and biomarkers involved both under-reporting and over-reporting of SU, and varied by types of samples and reference periods (Table 3). Five participants reported that they did not smoke but tested positive for urine cotinine, giving an under-reporting rate of 7%. No over-reporting was found for recent smoking. The disagreement between cotinine in hair or sweat patches compared with the *SumID-Q* and *SumID-CR* involved two cases of under-reporting (9%) and ten cases of over-reporting (21%). Regarding alcohol and cannabis use, we found no under-reporting when *SumID-Q* and *SumID-CR* were compared with biomarkers. However, over-reporting of alcohol use was very high (96%): 26 participants stated that they had used alcohol in the days before urine sampling (mean number of units = 7, *SD* = 5.8), but only one urine specimen tested positive. In self-reported cannabis use, we found an over-reporting rate of 26% for recent use and 31% for monthly use (Table 3).

3.5. Agreement and under- and over-reporting of SumID-CR compared with SumID-Q

The agreement between *SumID-CR* and *SumID-Q* for smoking and cannabis use was moderate to almost perfect (see Table 4), with the largest disagreement for lifetime cannabis use (staff members under-reported by 34%). Agreement between self-reported and collateral-reported alcohol use was low for lifetime use, substantial for monthly use, and moderate for recent use. Staff members under-reported lifetime use but over-reported recent alcohol use compared to self-report. The

Table 4
Agreement between SumID-CR and SumID-Q.[a]

	n	TP	FP	FN	TN	Kappa	95% CI
Tobacco Lifetime	94	73	1	9	11	0.628	0.423–0.833
Monthly	95	65	0	1	29	0.975	0.928–1
Recent	94	63	0	1	30	0.976	0.928–1
Alcohol							
Lifetime	94	82	2	9	1	0.110	−0.154–0.374
Monthly	88	58	6	2	22	0.748	0.640–0.925
Recent	89	24	32	2	32	0.315	0.168–0.462
Cannabis							
Lifetime	73	29	1	15	28	0.577	0.407–0.747
Monthly	88	8	5	6	69	0.519	0.271–0.767
Recent	88	8	3	5	72	0.614	0.371–0.857
Stimulants							
Lifetime	72	9	6	4	53	0.557	0.314–0.800
Monthly	91	0	1	2	88	−0.015	−0.036–.006
Recent	90	0	0	0	90	–	–

TP—true positive, FP—false positive, FN—false negative, TN—true negative
– Because of empty cells, these values cannot be calculated.
[a] Using the SumID-Q as reference.

agreement between *SumID-Q* and *SumID-CR* was moderate for lifetime stimulant use, with more over-reporting than under-reporting by staff. For monthly use, no agreement was found between *SumID-Q* and *SumID-CR,* with under-report by staff members (Table 4).

4. Discussion

In this study, we compared self-reported SU (*SumID-Q*), collateral-report by staff members (*SumID-CR*), and urine, hair, and sweat patches collected from a sample of 112 individuals with MBID. To our knowledge, this is the first study to compare these strategies to assess SU in individuals with MBID.

Irrespective of the assessment strategy, high rates of alcohol and tobacco use and slightly lower – but nonetheless substantial – illicit drug use rates were found. In fact, with the exception of alcohol use, lifetime and past month SU rates among participants were higher compared to SU rates in the general population in The Netherlands (past month tobacco use ~25%, alcohol use ~77%, cannabis use ~5%, stimulant use <1%; Laar & Ooyen-Houben, 2015). Whether this is due to sampling characteristics or is actually indicative of higher SU rates in Dutch individuals with MBID than the general population remains to be seen. Higher levels of smoking in individuals with mild ID, however, have been reported before (Žunić-Pavlović, Pavlović, & Glumbić, 2013; Kiewik, VanDerNagel, Kemna, Engels, & de Jong, 2016). Also, it has been hypothesized that the cannabis possession policies in Holland (i.e., possession of small amounts of cannabis for personal use is not prosecuted) may lead individuals with MBID to assume that cannabis use is harmless (VanDerNagel, Kiewik, Buitelaar et al., 2011). This in turn could explain high levels of cannabis use in our study, as well as willingness to disclose this use.

Participation rates differed significantly across the three strategies. In accordance with epidemiological studies conducted in the general population (e.g., Fendrich et al., 2004), participation in biomarker sampling was lower compared to participation in self-report. This may be ascribed to the relatively invasive nature of biomarker sampling and to the additional visits required to collect sweat patches. In addition, even though confidentiality of participants' test results was assured, individuals with risky SU behavior and those who perceived more severe consequences of having their SU identified may have been less willing to participate in the biomarker analysis. For those who were not willing to provide any biomarker samples and hence were excluded from the study ($n = 15$, 11% of the original sample), this hypothesis could not be further explored. However, in participants who refused either the urine sample ($n = 8$, 7% of the final sample) or the hair or sweat-patch sample ($n = 28$; 25% of the final sample), no significant differences were found in self-reported SU rates compared with participants who did provide these samples ($\chi^2(1, n = 112)$, *p*ís 0.184–0.905).

Regarding SU rates, our hypothesis based on research involving non-ID participants, which proposed that SU rates found with biomarker analysis would be higher compared to self-reported and collateral-reported SU rates, was not confirmed. SU rates found with biomarker analysis did not differ significantly from self-reported or collateral-reported tobacco, cannabis, and stimulant use; and self-reported and collateral-reported alcohol use rates were higher compared to those in biomarkers. The agreement between the *SumID-Q* and *SumID-CR* versus biomarker analysis was also better than expected. Contrary to our expectations and findings in the literature (Akinci et al., 2001; de Beaurepaire et al., 2007), we did not find high levels of under-reporting with the *SumID-Q* or with the *SumID-CR*. In fact, under-reporting of recent smoking (7%) of the *SumID-Q* and *SumID-CR* may in fact be even lower, since three participants classified as false negatives for self- and collateral-reported recent smoking had urine cotinine levels of 11, 14, and 16 ng/ml, which may also be explained by environmental contamination of the biomarker samples (Moeller et al., 2008; Rebagliato, 2002). In addition, self-report compared to biomarkers indicated

over-reporting of monthly tobacco use (21%), cannabis use (31%), and recent and monthly alcohol use (96%). A possible explanation for both the higher than expected levels of self-reported SU and the high levels of agreement between self-report and biomarker analysis, is that our participants were less reluctant to report SU compared to participants in previous studies. This may be attributed to the context of this study, which assured participants that their data would remain confidential and that they would not face any consequences of SU disclosure. Also, the structure of the *SumID-Q*, which aims to facilitate an open discussion of SU through a rather indirect approach, may have contributed to these results. For cannabis, the low levels of under-reporting in this study may be related to Netherland's cannabis policies. The strikingly low rates of alcohol use found in biomarker analysis and the associated high levels of alcohol over-reporting may be attributed to a smaller window of detection of alcohol in urine (University Medical Center Groningen, 2016) and the lack of long-term biomarker analysis of alcohol use. Contrary to our hypothesis, the availability of hair and sweat analysis results for tobacco, cannabis, and stimulant use did not result in higher SU compared to urine analysis alone. For THC, the detection sensitivity of the hair analysis seems lower than for the urine analysis, as no subjects tested positive for THC in hair despite having corresponding positive urine samples.

When comparing self-report and collateral-report, as expected, lifetime self-reported SU was higher for lifetime tobacco use, alcohol use, and cannabis use. However, contrary to our expectations, self-reported and collateral-reported SU rates for past month and recent SU were similar, except for recent alcohol use. According to the staff members in our study, most participants who consumed alcohol in the last month had done so in the last few days. By contrast, self-reported recent use was much lower compared to self-reported monthly use. The cause of this discrepancy remains unclear and warrants further investigation. Individuals with MBID may only consume alcohol on special occasions, although staff members may assume that those who drink will do so on a regular basis. Another explanation may be that participants are willing to admit to occasional drinking but not to more frequent or recent drinking. However, these explanations remain speculative, and they will need to be explored in further research on self-reported and collateral-reported alcohol use patterns of individuals with MBID. Agreement between self-reported SU and collateral-reported SU was moderate to substantial for almost all substances and almost perfect for the most overt SU (i.e., smoking). For recent and past month illicit drugs (cannabis and stimulant use) as well as for lifetime SU, staff members were more likely to under-report rather than over-report. Despite risks of under-reporting being present, the relatively high degree of agreement between self-reported and collateral-reported SU is promising. When staff members are aware of SU among their clients who also feel free to discuss SU openly, there are procedures in place to stage interventions to prevent SU-related problems from worsening.

4.1. Limitations

The findings of this study should be interpreted in the light of several limitations. First, the number of completed cases was limited, predominantly due to lower willingness to participate in hair and sweat patch testing. Second, we limited our study to the substances used most often by individuals with MBID residing in Dutch facilities (VanDerNagel, Kiewik, Buitelaar et al., 2011), and thus did not include other substances such as non-prescription opioids. Third, given the low number of individuals who were found to be using stimulants, the findings from earlier studies (e.g., Fendrich et al., 2004), which showed that drug testing can identify under-reporting of this substance, could neither be confirmed nor rejected in this study. Fourth, participant's prior knowledge of biological testing may have increased self-report accuracy. Therefore this accuracy may not reflect that in day-to-day care. Fifth, biomarker analysis rendered less than optimal results for several substances and reference periods.

Accurate biomarker analysis relies upon ideal sampling conditions, and this means that urine samples should be collected soon after SU to avoid false negative results due to metabolism and excretion of the drug. In hair and sweat patch samples, traces of infrequent SU are diluted by periods of non-use, as the collected samples contained sweat or hair from previous weeks to months. Infrequent SU may not be detected by biomarker analysis (Fendrich et al., 2004; Hoiseth et al., 2008). Therefore, whilst positive biomarker analysis may confirm drug use and provide results consistent with self-reported SU, negative biomarker analysis results do not necessarily mean no SU has occurred and may therefore falsely contradict self-reported SU. Although repeated and frequent biomarker sampling (e.g., several times a week) would theoretically be a solution, this may not be feasible in most situations. Less invasive micro sampling techniques, such as dried blood spots (a finger prick and a single drop of blood on a special card) or oral fluid sampling in combination with the sensitive LC–MS/MS analysis technique might be an option for future biomarker analyses.

4.2. Conclusions

Comparing three strategies to assess SU in individuals with MBID, we found lower willingness to participate in biomarker sampling than in self-report. A rather large number of participants reported having used substances, and contrary to the findings in non-ID populations, biomarker analysis did not result in a higher number of identified substance users. It remains to be seen if this finding can be explained by characteristics of the MBID group, the methodology of the *SumID-Q*, or by factors related to the less than optimal performance of biomarker analysis. However, we can conclude that given the additional costs and lower participation rates of biomarker analysis (especially for hair and sweat patches), using this strategy in the future seems to be of limited additional value compared to self-report or collateral-report. Therefore, it appears that for

individuals with MBID, self-report combined with collateral-report can be used to assess current SU, and this combination may contribute to collaborative, early intervention efforts to reduce SU and its related harms in this vulnerable group.

Competing interests

None.

Acknowledgements

This study was funded by ZonMW, The Netherlands Organisation for Health Research and Development (project number 60-60600-97-158). The authors wish to thank participants, institutions, and research assistants for their contributions, and Neil B. McGillicuddy, Eric O. Noorthoorn, and Neomi van Duijvenbode for supporting this study and proofreading this manuscript.

References

Akinci, I., Tarter, R., & Kiriski, L. (2001). Concordance between verbal report and urine screen of recent marijuana use in adolescents. *Addictive Behaviors*, *26*, 613–619.
American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders: DSM-5*. Washington, DC: American Psychiatric Association.
Brahm, N. C., Yeager, L. L., Fox, M. D., Farmer, K. C., & Palmer, T. A. (2010). Commonly prescribed medications and potential false-positive urine drug screens. *American Journal of Health System Pharmacy*, *67*, 1344–1350.
Bush, D. M. (2008). The U.S: Mandatory guidelines for federal workplace drug testing programs: current status and future considerations. *Forensic Science International*, *174*, 111–119.
Carroll Chapman, S. L., & Wu, L.-T. (2012). Substance abuse among individuals with intellectual disabilities. *Research in Developmental Disabilities*, *33*, 1147–1156.
Cohen, J. (1992). A power primer. *Psychological Bulletin*, *112*, 155–159.
Connors, G. J., & Maisto, S. A. (2003). Drinking reports from collateral individuals. *Addiction*, *98*(Suppl. 2), 21–29.
Cooper, G. A. A., Kronstrand, R., & Kintz, P. (2012). Society of Hair Testing guidelines for drug testing in hair. *Forensic Science International*, *218*, 20–24.
Cotten, S. W., Duncan, D. L., Burch, E. A., Seashore, C. J., & Hammett-Stabler, C. A. (2012). Unexpected interference of baby wash products with a cannabinoid (THC) immunoassay. *Clinical Biochemistry*, *45*, 605–609.
de Beaurepaire, R., Lukasiewicz, M., Beauverie, P., Castéra, S., Dagorne, O., Espaze, R., et al. (2007). Comparison of self-reports and biological measures for alcohol, tobacco: and illicit drugs consumption in psychiatric inpatients. *European Psychiatry*, *22*, 540–548.
de la Torre, R., & Pichini, S. (2004). Usefulness of sweat testing for the detection of cannabis smoke. *Clinical Chemistry*, *50*, 1961–1962.
Fendrich, M., Johnson, T. P., Wislar, J. S., Hubbell, A., & Spiehler, V. (2004). The utility of drug testing in epidemiological research: results from a general population survey. *Addiction*, *99*, 197–208.
Hoiseth, G., Bernard, J. P., Stephanson, N., Normann, P. T., Christophersen, A. S., Morland, J., et al. (2008). Comparison between the urinary alcohol markers EtG, EtS: and GTOL/5-HIAA in a controlled drinking experiment. *Alcohol and Alcoholism*, *43*, 187–191.
Kiewik, M., VanDerNagel, J. E. L., Kemna, L. E. M., Engels, R. C. M. E., & DeJong, C. A. J. (2016). Substance use prevention program for adolescents with intellectual disabilities on special education schools: a cluster randomised control trial. *Journal of Intellectual Disability Research*, *60*, 191–200.
Kintz, P., & Nicholson, D. (2014). Testing for ethanol markers in hair: discrepancies after simultaneous quantification of ethyl glucuronide and fatty acid ethyl esters. *Forensic Science International*, *243*, 44–46.
Koster, R. A., Alffenaar, J.-W. C., Greijdanus, B., VanDerNagel, J. E. L., & Uges, D. R. A. (2014a). Fast and highly selective LC–MS/MS screening for THC and 16 other abused drugs and metabolites in human hair to monitor patients for drug abuse. *Therapeutic Drug Monitoring*, *36*, 234–243.
Koster, R. A., Alffenaar, J.-W. C., Greijdanus, B., VanDerNagel, J. E. L., & Uges, D. R. A. (2014b). Application of sweat patch screening for 16 drugs and metabolites using a fast and highly selective LC-MS/MS method. *Therapeutic Drug Monitoring*, *36*, 35–45.
Laar van, M. W., & Ooyen-Houben van, M. M. J. (2015). *Nationale drug monitor [National drug monitor]*. Accessed 04.03.16. https://assets.trimbos.nl/docs/24dd30ba-464f-4dcd-a740-20ac058d310b.pdf
Landis, J. R., & Koch, G. G. (1977). The measurement of observer agreement for categorical data. *Biometrics*, *33*, 159–174.
Mayo Medical Laboratories. (2015). *Test ID: NICOU, nicotine and metabolites, urine*. Accessed 04.03.16. http://www.mayomedicallaboratories.com/test-catalog/Clinical+and+Interpretive/82510
McGillicuddy, N. B. (2006). A review of substance use research among those with mental retardation. *Mental Retardation and Developmental Disabilities Research Reviews*, *12*, 41–47.
Moeller, K. E., Lee, K. C., & Kissack, J. C. (2008). Urine drug screening: practical guide for clinicians. *Mayo Clinic Proceedings*, *83*, 66–76.
Rebagliato, M. (2002). Validation of self reported smoking. *Journal of Epidemiology & Community Health*, *56*, 163–164.
Stasiewicz, P. R., Vincent, P. C., Bradizza, C. M., Connors, G. J., Maisto, S. A., & Mercer, N. D. (2008). Factors affecting agreement between severely mentally ill alcohol abusers' and collaterals' reports of alcohol and other substance abuse. *Psychology of Addictive Behaviors*, *22*, 78–87.
University Medical Center Groningen. (2016). *Bepalingenwijzer [Laboratory reference guide]*.. Accessed 04.03.16. http://www.umcg.nl/NL/UMCG/Afdelingen/Ziekenhuisapotheek/producten_diensten/Bepalingenwijzer/Paginas/Bepalingenwijzer.aspx
van Duijvenbode, N., VanDerNagel, J. E. L., Didden, R., Engels, R. C. M. E., Buitelaar, J. K., Kiewik, M., et al. (2015). Substance use disorders in individuals with mild to borderline intellectual disability: current status and future directions. *Research in Developmental Disabilities*, *38*, 319–328.
VanDerNagel, J., Kemna, L., & Didden, R. (2013). Substance use among persons with Mild Intellectual Disability: approaches to screening and interviewing. *NADD Bulletin*, *16*, 87–92.
VanDerNagel, J., Kiewik, M., Buitelaar, J., & De Jong, C. (2011). Staff perspectives of substance use and misuse among adults with intellectual disabilities enrolled in dutch disability services. *Journal of Policy and Practice in Intellectual Disabilities*, *8*, 143–149.
VanDerNagel, J., Kiewik, M., Van Dijk, M., De Jong, C., & Didden, R. (2011). *Handleiding SumID-Q, Meetinstrument voor het in kaart brengen van Middelengebruik bij mensen met een lichte verstandelijke beperking [Manual of the SumID-Q. An instrument to assess substance use in individuals with a mild intellectual disability]*. Deventer: Tactus.
Wojcik, M. H., & Hawthorne, J. S. (2007). Sensitivity of commercial ethyl glucuronide (ETG) testing in screening for alcohol abstinence. *Alcohol and Alcoholism*, *42*, 317–320.
World Medical Association. (2013). World Medical Association Declaration of Helsinki : ethical principles for medical research involving human subjects. *Journal of American Medical Association*, *310*, 2191–2194.
Žunić-Pavlović, V., Pavlović, M., & Glumbić, N. (2013). Drug use in adolescents with mild intellectual disability in different living arrangements. *Drugs: Education, Prevention and Policy*, *20*, 399–407.