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**Cognitive profiles and academic achievement
in school-age children with borderline intellectual functioning**

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ENGLISH SUMMARY

Characterized by an IQ between one and two standard deviations below average (APA, 2000), and adaptive functioning problems, borderline intellectual functioning (BIF) is a clinical condition often associated with behavioral and psychiatric disorders, and with negative effects on daily life. Judging from the Gaussian distribution of intelligence, individuals with BIF would account for 13.6% of the population, but this is not always confirmed in the literature and the real prevalence of the condition is probably lower, as suggested by some authors (e.g. Ninivaggi, 2001).

Even so, the prevalence of BIF and the problems often associated with it (e.g. cognitive and learning problems that may result in academic failure and early school dropout, behavioral and emotional problems, a higher risk for mental health problems and maladaptive behavior across the life-span) are far from negligible, but there is a paucity of research on BIF in the Italian and international literature. BIF thus remains a relatively unknown phenomenon, not only in research, but also in the clinical and educational settings.

The aim of the present doctoral thesis was to explore BIF by focusing particularly on cognitive profiles and learning skills. The studies described were conducted on school-age children attending Italian primary and lower secondary schools. Data were collected by screening archival data made available by several Italian mental health services for children and adolescents, and referring to school-age children assessed for screening or diagnostic purposes.

The study described in chapter 2 was conducted to examine the cognitive profile of BIF, exploring whether BIF is characterized by a specific WISC-IV profile, with peculiar strengths and weaknesses. The WISC-IV scores of 204 children who obtained a Full-Scale IQ between 70 and 85 were considered. The results showed a spiky profile, characterized by significant differences between the various indexes: the lowest scores were obtained for Working Memory, the highest for Perceptual Reasoning; Verbal Comprehension, Working Memory, and Processing Speed were in the borderline range, Perceptual Reasoning on the lower end of the normal range¹. Further analyses suggested that the BIF profile was relatively independent of the child's age and general level of intellectual functioning, supporting the likelihood of BIF coinciding with a specific WISC-IV cognitive profile.

A second study (chapter 3) was designed to explore inter-individual variability to identify any subgroups among children with BIF revealing similar patterns in their cognitive profiles. The data were analyzed using different cluster analyses. The results pointed to three different subgroups characterized by a similar Full-Scale IQ, but differences in their four-factor index profiles. One subgroup's performance in Verbal Comprehension and Working Memory was in the borderline range, while Processing Speed was on the boundary between borderline and normal ranges (PSI = 86), and Perceptual Reasoning was in the normal range (PRI = 95.6). The second subgroup included children with borderline Verbal Comprehension, Perceptual Reasoning, and Working Memory abilities, while their Processing Speed was not impaired (PSI = 92.4). The third subgroup revealed a borderline performance in terms of Working Memory and Processing Speed, and normal Verbal Comprehension and Perceptual Reasoning abilities (89.3, and 91.1, respectively). An exploratory analysis suggested that the

¹ In the present thesis, the term "normal range" refers to scores above 85.

clusters differed slightly in terms of the children's socio-economic status and any comorbid disorders.

A third study (chapter 4) was devised to analyze the learning skills of 133 school-age children with BIF, comparing them on reading and math measures with a group of 73 children matched for chronological age with learning disabilities and an IQ above 85. The results revealed both similarities and differences between the two groups. The BIF group's performance was weaker in reading comprehension and written math operations, while no significant differences emerged between the groups in terms of reading accuracy and speed, or mental calculation and arithmetical facts retrieval.

It is worth noting that the children with BIF who also had a clinically-diagnosed specific learning disorder (SLD) did not differ significantly from those with BIF who had not a diagnosis of SLD. These two BIF subgroups were similarly weak in reading comprehension and written math operations by comparison with a group with learning disorders but no BIF.

Taken together, these studies provide some insight on BIF in school-age children, particularly as concerns their cognitive profiles and learning skills. While the clinical importance of BIF is still unclear and individuals with this condition often remain confined to a "grey area", the present contribution supports the conviction that BIF is characterized by particular features that warrant specific attention and intervention, and an appropriate diagnostic and clinical acknowledgement.

ITALIAN SUMMARY

Caratterizzato da un QI tra una e due deviazioni standard sotto la media (APA, 2000) e difficoltà nel funzionamento adattivo, il Funzionamento Intellettivo Limite (FIL) è una condizione clinica spesso associata a disturbi di tipo psichiatrico e problemi comportamentali, accompagnata da effetti negativi nella vita quotidiana.

Sulla base della curva normale di distribuzione dell'intelligenza, si può stimare una prevalenza del 13.6%, tuttavia tale dato non viene sempre confermato in letteratura e la prevalenza del FIL sembrerebbe essere inferiore (e.g. Ninivaggi, 2009).

Nonostante la prevalenza del FIL, così come i problemi ad esso associati (per esempio le difficoltà cognitive e di apprendimento, che possono risultare in fallimenti e abbandono scolastico, i problemi emotivi e comportamentali, l'aumentato rischio di problemi psichiatrici o comportamenti disadattivi evidenti nell'intero arco di vita) siano tutt'altro che trascurabili, la ricerca sul FIL sia a livello nazionale che internazionale è ancora carente. Per questo motivo il FIL continua ad essere un fenomeno poco conosciuto, non solo in letteratura, ma anche nella pratica clinica ed educativa.

L'obiettivo della presente tesi di dottorato è stato quello di esplorare il FIL, ponendo particolare attenzione sui profili cognitivi e le abilità scolastiche. Nello specifico, gli studi che verranno di seguito presentati hanno considerato bambini e ragazzi frequentanti la scuola primaria e secondaria di primo grado.

I dati sono stati raccolti attraverso l'analisi di cartelle cliniche presso diversi Servizi e Strutture sanitarie per l'età evolutiva e si riferiscono a bambini e ragazzi valutati a fini di screening o diagnostici.

Lo studio descritto nel capitolo 2 è stato condotto con l'obiettivo di indagare il profilo cognitivo del FIL e, in particolare, di verificare se tale condizione presenta un profilo specifico alla WISC-IV, con punti di forza e debolezza peculiari. Sono stati considerati i profili WISC-IV di 204 bambini e ragazzi che avevano ottenuto un QI totale tra 70 e 85. I risultati hanno evidenziato la presenza di un profilo "altalenante", caratterizzato da differenze significative tra gli indici della WISC-IV (Comprensione Verbale, Ragionamento Percettivo, Memoria di Lavoro e Velocità di Elaborazione). L'indice di Memoria di Lavoro si caratterizza per i punteggi più bassi, mentre le prestazioni migliori sono relative all'indice di Ragionamento Percettivo. Gli indici di Comprensione Verbale, Memoria di Lavoro e Velocità di Elaborazione si collocano nel range borderline (tra una e due deviazioni standard sotto la media), mentre l'indice di Ragionamento Percettivo si caratterizza per un punteggio medio di 88. Ulteriori analisi hanno evidenziato come tale profilo sembri essere relativamente indipendente dall'età del bambino e dal livello di funzionamento intellettuale generale (ovvero, al confine tra la disabilità intellettiva – QI tra 70 e 75 - o più alto – QI tra 76 e 85), supportando l'ipotesi che il FIL sia caratterizzato da un profilo specifico alla WISC-IV.

Un secondo studio (capitolo 3) è stato condotto con lo scopo di indagare la variabilità inter-individuale, in termini di possibili sottogruppi caratterizzati da profili cognitivi uniformi, all'interno della condizione di FIL.

I dati sono stati analizzati mediante analisi dei cluster, che hanno evidenziato la presenza di tre diversi sottogruppi, differenti tra loro per profili WISC-IV, ma non per QI totale. Nello specifico, un sottogruppo risulta caratterizzato da punteggi nel range borderline negli indici di Comprensione Verbale e Memoria di Lavoro, mentre la

Velocità di Elaborazione si colloca al confine tra il range borderline e la norma² (IVE = 86), e le abilità di Ragionamento Percettivo risultano preservate (IRP = 95.6).

Un secondo sottogruppo include i bambini caratterizzati da prestazioni nel range borderline negli indici di Comprensione Verbale, Ragionamento Percettivo e Memoria di Lavoro, mentre la Velocità di Elaborazione risulta preservata (IVE = 92.4).

Il terzo sottogruppo, infine, si caratterizza per carenze negli indici di Memoria di Lavoro e Velocità di Elaborazione, che si collocano nel range borderline, ma prestazioni non borderline negli indici di Comprensione Verbale e di Ragionamento Percettivo (rispettivamente, 89.3 e 91.1). Ulteriori analisi di tipo esplorativo, hanno evidenziato alcune differenze tra i sottogruppi relative allo status socio-economico e alla presenza di disturbi in comorbidità.

Un terzo studio (capitolo 4) ha avuto l'obiettivo di indagare le abilità scolastiche di 133 bambini e ragazzi con FIL, confrontando le loro prestazioni in lettura e calcolo con quelle di un gruppo, appaiato per età cronologica, di 73 bambini e ragazzi con difficoltà e disturbo degli apprendimenti e un QI superiore a 85. I risultati hanno messo in luce somiglianze e differenze tra i due gruppi. Il gruppo con FIL ha ottenuto prestazioni inferiori in compiti di comprensione del testo e nel calcolo scritto, mentre non sono emerse differenze statisticamente significative tra i due gruppi negli indici di accuratezza e velocità di lettura, nel calcolo a mente e nel recupero di fatti numerici.

Va sottolineato che i bambini con FIL che presentavano una diagnosi clinica di disturbo specifico di apprendimento non differivano significativamente da quelli con FIL che non presentavano tale diagnosi. Entrambi i sottogruppi di FIL hanno evidenziato prestazioni inferiori nei compiti di comprensione del testo e calcolo scritto rispetto ai bambini con difficoltà e disturbi degli apprendimenti, ma QI superiore a 85.

² Con l'espressione "in norma" si fa riferimento, nella presente tesi, ad un punteggio superiore a 85.

Globalmente, gli studi condotti forniscono alcune indicazioni sul FIL, con particolare riferimento ai profili cognitivi e alle abilità di apprendimento. Se da un lato l'importanza clinica del FIL è ancora poco chiara e gli individui con questa condizione rimangono confinati in un'“area grigia” o di limbo, il presente contributo supporta la convinzione che il FIL presenti, nella sua generalità, caratteristiche particolari che richiedono un'attenzione e interventi specifici, nonché, preliminarmente, un appropriato riconoscimento clinico e diagnostico.

CHAPTER 1

BORDERLINE INTELLECTUAL FUNCTIONING: AN OVERVIEW

1. INTRODUCTION

This category can be used when an individual's borderline intellectual functioning is the focus of clinical attention or has an impact on the individual's treatment or prognosis. Differentiating borderline intellectual functioning and mild intellectual disability (intellectual developmental disorder) requires careful assessment of intellectual and adaptive functions and their discrepancies, particularly in the presence of co-occurring mental disorders that may affect patient compliance with standardized testing procedures (e.g. schizophrenia or attention-deficit/hyperactivity disorder with severe impulsivity) (American Psychiatric Association [APA], 2013, p. 727).

This is how the DSM-5, the Diagnostic and Statistical Manual of Mental Disorders, describes Borderline Intellectual Functioning, giving little or no clear information for the diagnosis and characterization of this clinical condition - even less than in the previous edition of the Manual (DSM-IV-TR, APA, 2000), which specified an IQ range from 71 to 84. In both versions, BIF finds a place in the V-code section,

defined in the former as “Other conditions that may be a focus of clinical attention” (APA, 2013), and in the latter as “Additional conditions that may be a focus of clinical attention” (APA, 1994, 2000).

Despite these latest two editions of the DSM, the classification and definition of what we now label as “BIF” has changed over time, starting out in the mental retardation (intellectual disability) category in the first two editions of the DSM.

In 1973 the American Association on Mental Deficiency omitted the “borderline mental retardation” category, which was introduced in the fifth edition of its manual. The cut-off of one standard deviation below the mean was abolished and a four-level classification of disability was re-introduced (i.e. mild, moderate, severe, and profound) (Grossman, 1973). This change was due to the fact that not all individuals with an IQ between one and two standard deviations below the mean have adaptive impairments. This change was followed by a drop in the number of diagnoses of mental retardation, and by a marked increase in the diagnoses of learning disabilities (Ferrari, 2009).

The point is that, though they are not eligible for a diagnosis of intellectual disability, individuals with BIF may be unable to cope with the demands of an increasingly complex society (Fennell & Ek, 2010) without specific educational or societal assistance.

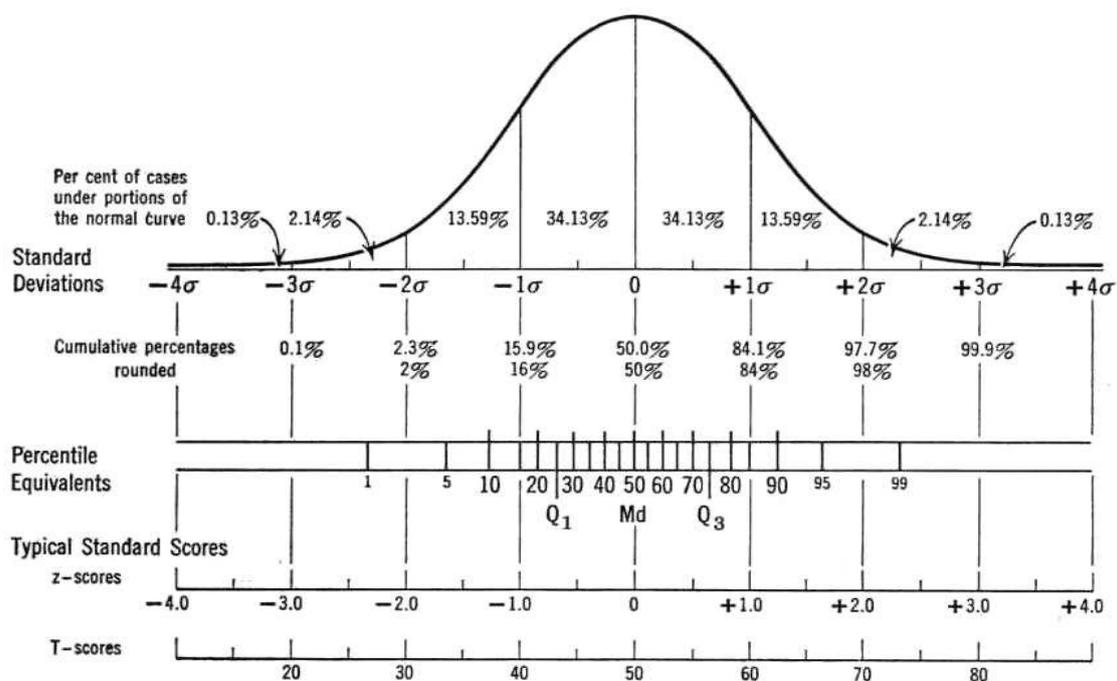
In the ICD Classifications, we can find something similar in the evolution of how Borderline Intellectual Functioning has been considered. It is now located in a loosely-defined code R41.8 (World Health Organization [WHO], 1992). BIF is described here exclusively in terms of its deficits in cognitive functioning and the involvement of awareness. A clear cut-off in terms of IQ is specified (71-84).

As mentioned earlier, the latest edition of the DSM (APA, 2013) gives us no clear idea of what BIF is or how it can be diagnosed, and no numerical range is given

for the IQ of individuals with BIF. Hence the lack of consensus on the criteria adopted to identify BIF in clinical practice and research. Several authors have suggested that, as we near intellectual quotients in the borderline range, impairment in adaptive functioning becomes important to define BIF as “clinical condition” (e.g. Ninivaggi, 2009; Vianello, Di Nuovo, & Lanfranchi, 2014; Salvador-Carulla et al., 2013). Other authors only considered cognitive limitations and an IQ in the borderline range to establish the presence of BIF (see for example Vianello et al., 2014, for a summary of previous studies).

This lack of information and attention clashes with the high prevalence of the phenomenon. Considering the normal distribution of intelligence, in fact, individuals with BIF account for about 14% of the population.

Figure 1. Normal distribution of intelligence.



As several authors have underscored, not all individuals with a borderline IQ have impairments in adaptive functioning or need special help (e.g. Peltopuro, Ahonen, Kaartinen, Seppälä, & Närhi, 2014; Salvador-Carulla et al., 2013; Vianello et al., 2014). Nevertheless, the prevalence of the phenomenon is not clear (e.g. Ferrari, 2009): different rates have been suggested, some higher, others lower than those expected on the basis of the normal distribution (ranging from 1-3% to 18% or more: see for example Vianello et al., 2014; Salvador Carulla et al., 2013). The recent Italian Ministerial Directive on Special Educational Needs stated that pupils with BIF account for 2.5% of all Italian school-aged children (Ministero dell'Istruzione, dell'Università e della Ricerca [MIUR], 2012).

This absence of agreement on how to quantify the prevalence of BIF could be due to methodological differences between studies (e.g. recruitment processes, or different ages considered), or to the lack of consensus on the terminology regarding BIF (Salvador-Carulla et al., 2013). Different names have been used to indicate individuals with BIF, including borderline mental retardation, borderline intelligence, borderline intellectual functioning, slow learners, grey area kids, invisible children, etc. (e.g. Jankowska, Bogdanowicz, & Shaw, 2012; Peltopuro et al., 2014). BIF is also often masked by other clinical conditions that share the similar symptoms.

So, BIF remains a misdiagnosed and unrecognized condition (Ferrari, 2009; Wieland & Zitman, 2016), given little consideration in clinical and research fields. Like the little space dedicated to BIF in the diagnostic manuals, so too in the literature publications on this topic are still few and far between (e.g. Fenning, Baker, Baker, & Crnic, 2014; Wieland, 2016).

Some information about BIF can be inferred from studies on intellectual disabilities. Individuals with BIF are often not the focus of such studies, but they may

be considered as a control or comparison group in research on intellectual disability or specific learning disorders, or they may be pooled together in such studies (see Peltopuro et al., 2014). Other information about BIF emerges from clinical practice, in terms of its more common features observed by clinicians and other professionals (e.g. Ninivaggi, 2009).

In agreement with the CONFIL 2007, a Spanish consensus group, BIF could be described as “a health meta-condition that requires specific public health, education and legal attention” (Salvador-Carulla et al., 2013). It is neither a mental disorder nor an intellectual disability, but a complex condition that could be associated with several neurodevelopmental disorders and impairments across the lifespan (Jankowska et al., 2012).

BIF is a pervasive condition that may influence a person’s overall functioning. Nouwens, Lucas, Embregts, and van Nieuwenhuizen (2017) found that individuals with BIF had more family and social problems than individuals with mild intellectual disability. In the sample considered, the average age at which these individuals first received care from a professional care-provider was 18 years old.

Families with a child with BIF seem to differ in their parent-child interactions from either families with typically-developing children or those with children who have intellectual disabilities. In particular, parents are more likely to show a less positive engagement and higher rates of negative behavior when they interact with a six-year-old child with BIF (Fenning et al., 2014). The poor-quality parenting among mothers of 5-year-old children with BIF is not associated with higher rates of difficult behavior among these children – in this sense, they are no different from typically-developing children (Fenning, Baker, Baker, & Crnic, 2007). According to the authors, differences in maternal involvement could be associated with the degree of awareness of the child’s

difficulties (which was found inadequate in mothers of children with BIF). Indeed, mothers who expressed early awareness of their child's difficulties were more likely to exhibit a more positive parenting behavior (Fenning et al., 2007).

For these reasons too, several authors have claimed that the early identification of cases of BIF, followed by the provision of adequate support, could help to prevent the negative evolution of this condition (Salvador-Carulla et al., 2013). This could have an important influence on the developmental trajectories of individuals with BIF (Jankowska, 2016).

The late detection of BIF is due largely to the lack of a specific phenotype or visible features (e.g. Karande, Kanchan, & Kulkarni, 2008), or discriminating behavioral characteristics (Salvador-Carulla et al., 2013), combined with the fact that individuals with BIF may respond positively to certain environmental demands, and may not reveal clear-cut developmental delays in early childhood (Farhadifar et al., 2011; Karande et al., 2008).

BIF is often recognized when a child starts school, when they are slow or unable to acquire basic academic skills (Jankowska, 2016, Ninivaggi, 2009; MacMillan, Gresham, Bocian, & Lambros, 1998). These children may reach the end of primary school or even be further into their education without their BIF being recognized (Salvador-Carulla et al., 2013), partly because of a delay between when learning problems are noticed and when a child is referred for a clinical assessment (Karande et al., 2008). Despite their difficulties in achieving their education milestones, pupils with BIF do not receive adequate educational and societal assistance (e.g. Jankowska, 2016). The gap between their knowledge and abilities and those of their peers consequently widens. They experience a series of failures (Salvador-Carulla et al., 2013), which cause emotional distress and a lack of self-esteem and self-competence (Karande et al., 2008).

They suffer from grade retention, and higher school dropout rates, ultimately becoming poorly educated (e.g. Jankowska, 2016), with lifelong negative effects (e.g. Jankowska et al., 2012).

1.1 Causes, risk factors, and comorbidity

Like the case of intellectual disabilities, no single, isolated cause has been found for BIF (e.g. Ninivaggi, 2009). Several causes may be recognized (e.g. Di Blasi, Savelli, Zingale, Buono, & Di Nuovo, 2014; Ninivaggi, 2009; Salvador-Carulla et al., 2013), and more than one may be present at the same time in the same person (e.g. Vianello et al., 2014).

Some of the causes of BIF can be classified as biological and environmental (e.g. Blasi, Baglio, Baglio, Canevini, & Zanette, 2017). Then there are motivational and affective factors, as well as indirect effects of other neurodevelopmental disorders (such as specific learning disorder or attention deficit hyperactivity disorder), which can exacerbate weaknesses in a child's development and intellectual impairments (Vianello et al., 2014).

The biological causes are distinguishable as genetic and non-genetic factors, which are much the same as those associated with intellectual disabilities. Generally speaking, the genetic causes include genetic syndromes typically responsible for intellectual disabilities, or that involve intellectual functioning, and autism spectrum disorders. Then there are several non-genetic biological risk factors that, as in the case of intellectual disabilities, include a number of negative conditions that may occur pre-, peri- or postnatally, influencing the child's development (e.g. Vianello et al., 2014), and increasing the risk of intellectual disability, or BIF, or other neurodevelopmental disorders.

Prematurity, low birth weight, chronic disease, asphyxia, maternal drug or alcohol abuse during pregnancy, and exposure to toxic materials are just some of the factors often associated with an increased risk of impairment or delay in a child's development (e.g. Delpisheh, Attia, Drammond, & Brabin, 2006; Durkin et al., 2000), and consequently of BIF too.

There is some evidence in the literature to suggest an influence of birth weight on impairment in intellectual functioning. Chen, Lawlor, Duggan, Hardy, and Eaton (2006), for example, found low birth weight more frequent among 4-year-old children with BIF or intellectual disabilities than in typically-developing children. But when Farhadifar et al. (2011) analyzed risk factors in school-aged children with BIF they found no significant relationship between low birth weight and BIF, or between prematurity and BIF. The authors did find several other factors associated with a higher risk of BIF, however, such as prenatal care, maternal drug use during pregnancy, birth order (seventh or more), family history of psychiatric disorders or intellectual disability, history of stuttering or head trauma, complications during pregnancy, maternal disease before and during pregnancy, and serious disease in their offspring.

Considering school-age children with BIF, Karande et al. (2008) found that most children with BIF had a normal perinatal history, typical developmental milestones, and a normal physical appearance. Soft neurological signs were identified in 61.8% of their sample, however. On the other hand, some authors have suggested that there are early developmental delays in individuals with BIF (see Peltopuro et al., 2014, for a review). These different results regarding potential risk factors for BIF could be due to the specific samples considered (for example, Karande and colleagues considered individuals living in families with upper or middle-upper socioeconomic conditions, while prematurity and other problems relating to perinatal history were more likely in

families with a low socioeconomic status), or to the cut-off used in the different studies to establish low birth weight, for example (Farhadifar et al., 2011).

As for environmental causes of BIF, it is recognized that the environment influences children's development. For example, children living in families with a higher socioeconomic status (SES) outperform their lower-SES peers in several cognitive measures (e.g. Bradley & Corwyn, 2002). Differences in SES are also seen in academic attainment and years of education (e.g. Brooks-Gunn & Duncan, 1997; Battin-Pearson et al., 2000), as well as in socio-emotional development (Bradley & Corwyn, 2002).

Negative effects of the environment (e.g. environmental disadvantage, little stimulation from by parents or caregivers, etc.) are more likely to be behind a condition of BIF than to cause intellectual disability (e.g. Vianello et al., 2014). In the above-cited study, Farhadifar et al. (2011) also found an influence of environmental factors. Their results indicated that parents' illiteracy, a family history of psychosis or intellectual disability, and a low SES were significantly associated with borderline intelligence in children.

BIF seems to be overrepresented in populations characterized by a low SES and poverty. Emerson, Einfeld, and Stancliffe (2010) found, for example, in a large sample of Australian four- or five-year-old children that those with BIF and intellectual disability were more likely to be exposed to socioeconomic disadvantage than their peers with on-average intelligence. In their study, the authors considered several variables associated with the family's socioeconomic position, such as income poverty, material hardship (assessed, for example, in terms of difficulty paying bills on time), subjective poverty, and parents' education.

Vianello and colleagues found similar results in their sample of preschoolers and children in the early years of primary school: a large percentage of the children with BIF lived in low-SES families (see Vianello et al., 2014).

Whatever the causes that may underlie BIF, the condition is often associated with different clinical conditions or neurodevelopmental or neuropsychiatric disorders. According to the CONFIL 2007 consensus group, the neurodevelopmental disorders most commonly associated with BIF include generalized developmental disorders, and specifically high-functioning autism, Asperger syndrome, and non-specific generalized developmental disorder, specific learning disorders, attention deficit and hyperactivity disorder, and fetal alcohol syndrome, as well as some genetic syndromes that are often associated with intellectual disabilities (Salvador-Carulla et al., 2013).

Several researchers analyzed the prevalence of mental disorders in individuals with BIF. Their results suggest that people with BIF have higher rates of mental health problems than the general population in childhood, adolescence and adulthood (e.g. Chen et al., 2006; Emerson et al., 2010; Hassiotis et al., 2008). These mental health problems are likely to be a consequence of the features of BIF. For example, Masi, Marcheschi, and Pfanner (1998) reported a higher incidence of psychiatric disorders in adolescents with BIF. These authors suggested that individuals with BIF have fewer mental resources to cope with internal and external conflicts because of their limited intellectual functioning. It may be that clinical features already present at an early age (such as learning difficulties or attention problems) that result in scholastic failure are accentuated in adolescence by a subsequent loss of emotional and behavioral control, or by growing feelings of incompetence and failure.

BIF has been found associated with higher rates of drug and alcohol use and abuse (Hassiotis et al., 2008), of suicidal behavior (Hassiotis, Tanzarella, Bebbington, &

Cooper, 2011), and of antisocial behavior and criminal acts (see Peltopuro et al., 2014, for a review) than in the general population.

1.2 Clinical and cognitive features associated with borderline intellectual functioning in school-aged children

Research and clinical practice have pinpointed some clinical features often associated with BIF. As previously reported, BIF typically comes to the clinician's attention during school years, when children begin to reveal impairments in their academic achievements, given that they usually do not show any clear physical signs of their disorder (e.g. Karande et al., 2008; Salvador-Carulla et al., 2013).

That is why they have been called “six-hour retarded children”, to indicate that these children show problematic or maladaptive functioning at school, but an almost normal functioning outside school (e.g. MacMillan et al., 1998). School-aged children with BIF are described as having an erratic academic performance, with mild but generalized learning problems (Ninivaggi, 2009). The literature on their academic achievements is not always clear, however, particularly as concerns reading and spelling. For example, some studies found poor reading skills in children with BIF, when compared with children with average intellectual functioning (e.g. Claypool, Marusiak, & Janzen, 2008), and weaknesses in spelling (e.g. MacMillan et al., 1998). But other studies produced different findings, with children with BIF performing on a par with those of average intelligence in measures of reading and spelling (e.g. Kortteinen, Narnhi, & Ahonen, 2009). On the other hand, performance in mathematics is always poorer in children with BIF than in their peers with average intellectual functioning, and the same applies to reading comprehension ability (e.g. Claypool et al., 2008; Kortteinen et al., 2009; MacMillan et al., 1998).

Their academic problems may be linked to a lower than average intelligence, but also to other cognitive features of individuals with BIF. The literature provides evidence of limitations in short-term and working memory when children and adolescents with BIF were compared with typically-developing children on both verbal and visuospatial components (e.g. Alloway, 2010; Bonifacci & Snowling, 2008; Schuchardt, Gebhart, & Maehler, 2010; but see Henry, 2001 and Kortteinen et al., 2009 for two exceptions regarding visual and spatial memory). Children with BIF generally had a higher performance than their peers with intellectual disabilities, however (e.g. Henry, 2001; Schuchardt et al., 2010). Their impairments in WM seem to be consistent with their general intellectual impairment (Schuchardt, Maehler, & Hasselhorn, 2011).

Alloway (2010) studied executive functions in children with BIF, comparing them with a group of typically-developing peers, and found performance significantly worse in the BIF group in all the tasks presented.

Children with BIF are slower in processing speed tasks, and poorer in sustained attention, than their peers with average intellectual functioning, or peers with reading disorder (Bonifacci & Snowling, 2008). Individuals with BIF also seem to have more problems with abstract and logical thought, and a general tendency towards more concrete thoughts, which means they are more confident with practical tasks. Given their lower intellectual level, individuals with BIF are significantly delayed in reaching the Piagetian stage of concrete operational thought, while formal operational thought is rarely acquired (Masi et al., 1998).

Another clinical feature often associated with BIF seems to be a lower capacity to generalize information from one learning context to another (Ninivaggi, 2009), again indicating a certain rigidity of thought.

In the study by Baglio et al. (2016), children with BIF showed a worse performance in tasks assessing theory of mind than their typically-developing counterparts. The authors suggested that this deficit could be related to their impaired executive functioning.

Children with BIF also revealed higher levels of school anxiety and depressed mood than their peers with average intellectual functioning (Alesi, Rappo, & Pepi, 2015). Emerson et al. (2010) found higher rates of psychiatric disorders in 6- to 7-year-old children with BIF than in their typically-developing peers. When compared with children with intellectual disabilities, children with BIF showed a similar prevalence of conduct and emotional problems, lower levels of hyperactivity and peer problems, but even less prosocial behavior.

In short, school-aged children with BIF show higher rates of learning and cognitive problems than their peers with normal intellectual functioning. This increases their risk of educational failure, grade retention and school drop-out (e.g. Fernell & Ek, 2010; Karande et al., 2008), particularly when adequate support is not provided. The effects may be recognizable throughout their lives and in different areas of functioning, also affecting their quality of life.

2. BORDERLINE INTELLECTUAL FUNCTIONING IN THE ITALIAN SCHOOL SYSTEM

In the Italian formal education system, almost all pupils with special educational needs attend mainstream schools. Over the years, several laws and ministerial directives have been adopted to support their inclusion at school.

In accordance with Law 104/1992 and all subsequent legislation and decrees, children with disability are generally enrolled in mainstream school at all educational levels. They are supported by additional teachers specially trained to facilitate their inclusion. Children with BIF are usually not considered eligible for such support, however. Another piece of legislation (Law 170/2010) recognized dyslexia, dysgraphia, dysorthographia and dyscalculia as specific learning disorders. Supported and completed by subsequent regulations and decrees, this law stated that students with specific learning disorders do not need a support teacher, but they may take advantage of a series of educational measures that facilitate their teaching and learning.

No specific requirements were available for children with BIF until the enactment of the Ministerial Directive of 27 December 2012, on ‘Measures for pupils with special educational needs and local organizations for school inclusion’, which marked an important shift in the right direction. It created a macro-category covering all kinds of difficulties at school, both temporary or permanent, and established the importance of recognizing and supporting all pupils with different special needs, even those without a disability proper.

The Directive explicitly dedicates a paragraph to pupils with BIF, considering them as individuals who require special attention and adequate support to reach better academic outcomes and a better quality of life. According to the Directive and its subsequent specifications, the school takes action autonomously to provide support for children with BIF and nurture their development by means of tailored educational plans that allow for their schooling to be customized and adapted to the student’s actual characteristics and needs (MIUR, 2012, 2013a, 2013b).

In the light of these considerations, further research on BIF should clarify the characteristics of this condition in order to help schools plan adequate educational support better tailored to the abilities and needs of these children.

3. OUTLINE OF THIS DISSERTATION

The above-described issues associated with BIF, and particularly the lack of attention it has received in the literature, raise important questions, and highlight the need for further research on this sensitive condition.

The results of the studies described in this dissertation contribute to the growing body of knowledge regarding this topic, and focus specifically on the cognitive profiles of school-age children with BIF.

The choice of this particular age group derives from a personal interest in developmental age and the conviction that the early identification of individuals with developmental problems can positively influence their subsequent outcomes. Indeed, failures experienced during a child's school career can influence their development in terms of their motivation, self-esteem, and self-concept. They may result in an early school drop-out and a consequent lack of an adequate education, which in turn carries a greater risk of negative outcomes throughout the individual's lifespan.

Data were collected thanks to the cooperation and consent of several mental health services for children and adolescents. Archival data on children assessed for clinical purposes between 2013 and 2016 were screened to identify children attending primary or early secondary school with an IQ in the borderline range, as obtained by administering the WISC-IV (Wechsler, 2003). We focused on this test in order to obtain

a more complete measure of the children's intellectual abilities, so that their intellectual profiles could be assessed in terms of strengths and weaknesses (the aim of the first study described in the dissertation). Only children assessed with this test were considered in order to ensure that they were comparable.

Data regarding children with a FSIQ between 70 and 85 scores were collected in a dataset. For each participant, the information concerned their WISC-IV performance, age, school grade, socio-demographic details (e.g. parents' education and occupation, and children's and parents' nationality), information available from interviews with parents about the children's adaptive behavior (i.e. standardized measures of adaptive behavior were only available for a minority of the cases examined), the reasons for requesting a clinical assessment, and the clinical diagnosis, if any, were collected.

The first two studies focus on intellectual profile(s), as assessed with the WISC-IV (Wechsler, 2003). Specifically, in the first study the WISC-IV profile of individuals with BIF was examined to explore the possibility of a specific profile for this condition. The specificity of the BIF profile was also examined in terms of the level of general impairment or full-scale IQ (i.e. considering children with an IQ at the lower end of the borderline range, near the cut-off for a diagnosis of intellectual disability, and children with higher IQ scores), and different ages (in terms of school grades). The developmental trajectories of the intellectual components were considered.

The second study focused on inter-individual variability in cases of BIF, and particularly explored whether this variability is associated with the existence of homogeneous subgroups characterized by different patterns of intellectual functioning.

The third aim of the dissertation was to investigate learning abilities in pupils with BIF. School performance is one of the most crucial aspects in children with BIF, and one of the first evident symptoms to prompt their referral for a clinical assessment.

Several reports have concluded that children with BIF have academic difficulties, but few studies have focused on these children's particular learning abilities. Given the frequent association between BIF and specific learning disorders (SLD), in terms of both comorbidity and the difficulties encountered, and bearing in mind that these conditions are often confused, another purpose of the study was to investigate differences in learning abilities between children with BIF and those with SLD and an IQ above 85. The third study specifically focused on analyzing the learning abilities of individuals with BIF, and their reading and mathematics skills in particular.

In summary, the studies presented in this dissertation addressed three general aims:

1. to investigate the presence of a specific intellectual profile, characterized by peculiar strengths and weaknesses, in individuals with BIF;
2. to explore the presence of different subgroups with homogeneous intellectual profiles, among the cases of BIF; and
3. to analyze the learning abilities of children with BIF and identify any similarities and differences vis-à-vis children with specific learning difficulties and disorders, and an IQ above 85.

CHAPTER 2

STUDY 1:

INTELLECTUAL PROFILE IN SCHOOL-AGED CHILDREN WITH BORDERLINE INTELLECTUAL FUNCTIONING

1. INTRODUCTION

Borderline Intellectual Functioning (BIF) is a complex condition characterized by an intellectual functioning below the normal range, but above the cut-off for the diagnosis of intellectual disability, which means an intellectual quotient between one and two standard deviations below the mean (APA, 2000; WHO, 1992). As several authors have suggested, impairment in intellectual functioning has to be associated with impairments in adaptive functioning to consider “clinical” the condition of BIF (e.g. Ninivaggi, 2009; Vianello et al., 2014).

In recent decades, BIF has been excluded from classifications of intellectual disability (or mental retardation, or mental deficiency, as it was called in the past) (e.g. Ferrari, 2009; Wieland & Zitman, 2016), and become a “marginal” condition without a

clear definition or categorization, as we can see from the DSM-5 (APA, 2013), which does not give specific criteria for defining this condition. Little attention has been dedicated to this condition in the literature, and even when it was considered, it was not always the specific focus of study (Peltopuro et al., 2014).

BIF seems to characterize a broad percentage of the population, however (e.g. Emerson et al., 2010; Ferrari, 2009; Salvador-Carulla et al., 2013). Individuals with BIF can be considered vulnerable or at risk of negative outcomes across the life span, such as low academic achievement, school dropout, behavioral and societal problems, and psychiatric disorders (e.g. Emerson et al., 2010; Fernell & Ek, 2010; Hassiotis et al., 2008; Masi et al., 1998; Peltopuro et al., 2014).

Intelligence is one of the most studied constructs in the field of psychology. There is no single definition of intelligence, but experts generally agree to consider intelligence as the individual's ability to adapt to environmental demands and to learn from experience (Sternberg & Detterman, 1986). Intellectual quotient is often measured by tasks that assess different components of intelligence (e.g. verbal, non-verbal, working memory and processing speed, using the Wechsler scales), even though some tests were developed to assess specific aspects of intelligence (e.g. Raven's Colored Progressive Matrices, CPM, Raven, Raven, & Court, 1998; or Logical Operation and Conservation test, LOC, Vianello & Marin, 1998). Individual differences in intellectual functioning go beyond a mere difference in IQ, and are better interpreted in terms of specific strengths and weaknesses.

In line with this assumption, several researchers have attempted to analyze cognitive profiles in individuals with neurodevelopmental disorders. Their results showed, for example, that individuals with intellectual disabilities of different etiology had some characteristics in common, but performed differently in the components

considered (e.g. Di Nuovo & Buono, 2009a; Vianello, 2008). An example comes from Williams and Down syndromes, which are known to feature almost opposite cognitive profiles (e.g. Paterson, 2001). Specifically, individuals with Down syndrome often show a relative weakness in verbal abilities, whereas their visuo-spatial skills seem to be relatively well preserved; the opposite is true in Williams syndrome (e.g. Dykens, Hodapp, & Finucane, 2000).

In the case of BIF too, a clear understanding of the related cognitive profile in terms of strengths and weaknesses would probably shed more light on the functioning of these individuals.

The current study thus aimed to investigate the possibility of a specific profile of intellectual functioning in individuals with BIF, based on the WISC-IV indexes.

The WISC-IV (Wechsler, 2003) is currently the most often used tool for the clinical cognitive assessment of children between 6 and 16 years old (e.g. Bremner, McTaggart, Saklofske, & Janzen, 2011), to obtain a measure of intelligence not only when intellectual disability is suspected, but also when diagnosing specific learning disorder, attention-deficit and hyperactivity disorder, or other neurodevelopmental disorders.

As well as the Full-Scale IQ (FSIQ), which is a measure of overall intellectual ability, the WISC-IV allows to calculate four main indexes (similar to the additional indexes in the WISC-III, Wechsler, 1991) that relate to different, separate cognitive abilities, i.e. the Verbal Comprehension Index (VCI), the Perceptual Reasoning Index (PRI), the Working Memory Index (WMI), and the Processing Speed Index (PSI), which replace the previous Verbal IQ and Performance IQ. Working Memory and Processing Speed load the General IQ more than in the previous edition (being represented by four subtests instead of two), but - by eliminating the dual structure (i.e.

Verbal IQ and Performance IQ) characteristic of the WISC-III - the WISC-IV enables a better estimation of verbal comprehension and perceptual reasoning, less influenced by working memory and processing speed (Raiford, Weiss, Rolfhus, & Coalson, 2005).

Several authors have suggested that the FSIQ provides little information about intellectual functioning in clinical populations (e.g. Fiorello et al., 2007). In these cases it could be more suitable to use the four main indexes, or the two additional global indexes that can be calculated on the basis of the WISC-IV subtest, i.e. the General Ability Index (GAI) and the Cognitive Proficiency Index (CPI). The former was first developed for the WISC-III by Prifitera, Weiss, and Saklofske (1998) to obtain a measure of general cognitive ability uninfluenced by the Arithmetic and Coding subtests. Using the WISC-IV, the GAI is obtained by considering the Verbal Comprehension and Perceptual Reasoning indexes (Raiford et al., 2005). The GAI thus provides a measure of global cognitive functioning that is less influenced by working memory and processing speed (Cheremie, Stafford, & Mire, 2008). The CPI summarizes the working memory and processing speed performances (Weiss & Gabel, 2008).

Analyzing the WISC-IV findings in several clinical conditions led to the identification of different profiles. When specific learning disorders (SLD) are considered as a single group, for instance, this profile seems to feature a better performance in Verbal Comprehension (VC) and Perceptual Reasoning (PR) than in Working Memory (WM) and Processing Speed (PS) (e.g. Cornoldi, Giofrè, Orsini, & Pezzuti, 2014; Toffalini, Giofrè, & Cornoldi, 2017). A similarly spiky profile emerged for children with ADHD (e.g. Fenollar-Cortés, Navarro-Soria, Gonzáles-Gómez, & García-Sevilla, 2015; Thaler, Bello, & Etcoff, 2012), whereas individuals with

intellectual disabilities showed a flatter profile, marked by generalized and homogeneous weaknesses across all indexes (e.g. Cornoldi et al., 2014).

For now, little is known about the WISC-IV profile in children with BIF. In a previous study that compared the intellectual profiles of children with SLDs and with intellectual disabilities, Cornoldi et al. (2014) had a subgroup of children with BIF in the sample of children with a clinical diagnosis of SLD. Comparing the WISC-IV profile of these three subgroups, the authors found that individuals with BIF had a WISC-IV profile more similar to that of individuals with SLDs, with higher scores for VC and PR than for WM and PS, whereas the profile of individuals with intellectual disability was flat. It may be, however, that these results were influenced by the sample selection criteria: these BIF participants were originally selected because they had a diagnosis of SLD, so their WISC-IV profile may have been influenced by the fact that they had a comorbidity between BIF and SLD. Indeed, the authors of the study identified a small subgroup of children with BIF (FSIQ range from 70 to 75) in the sample with a clinical diagnosis of intellectual disability, whose profile was flat. This is in line with what found by Bremner et al. (2011) when they considered the additional indexes.

In the light of these results, the aim of the present study was to explore the WISC-IV profile (considering the four main factor indexes) of individuals with BIF. In particular, this study aimed to explore whether the cognitive profile of these individuals resembles the one found for intellectual disabilities (that seems to be flat, characterized by general and homogeneous weaknesses), or the one found for specific learning disorders (characterized on the whole by significantly higher verbal comprehension and perceptual reasoning abilities than working memory and processing speed) (Cornoldi et al., 2014). Or BIF may have its own specific profile, with peculiar characteristics that

make it differ quantitatively and qualitatively from that of typical development too (on the basis of standardization criteria; i.e. with no differences between the four indexes, since the scores for all indexes in each age group are given on a distribution with a mean of 100 and a standard deviation of 15).

Several authors have suggested that working memory is impaired in individuals with BIF. For example, Alloway (2010) compared children with BIF and typically-developing children matched for chronological age, and found a worse performance in both verbal and visuo-spatial short-term and working memory in the former. Similar results were obtained by Schuchardt et al. (2010), who found a worse performance in children with BIF compared with typically-developing children matched for chronological age, while the BIF group outperformed their peers with mild intellectual disability. Children with BIF, however, did not differ from typically-developing children matched for mental age in terms of the central executive and visuo-spatial working memory (Schuchardt et al., 2010), or in verbal working memory (Schuchardt et al., 2011). Overall, these and other studies have shown a weaker working memory functioning in individuals with BIF than in typically-developing children of the same chronological age (e.g. Alloway, 2010; Bonifacci & Snowling, 2008; Mäehler & Schuchardt, 2009). In line with this evidence, albeit with a general weakness intrinsic in BIF, the WISC-IV profile of individuals with BIF might be expected to reveal a relatively more impaired WM index.

Again, in line with research on the WISC-IV profile in clinical conditions, the study also focused on the additional scores that can be calculated from the WISC-IV subtest results, i.e. the GAI and CPI. On average, the GAI could be expected to be higher than the CPI in BIF, due largely to the predicted impairment in WM.

Secondly, potential differences linked with the level of intellectual functioning (as emerged from the FSIQ) were investigated. Specifically, given the overlap with intellectual disabilities in terms of IQ, children with a FSIQ at the lower end of the borderline range (from 70 to 75) were compared with children with higher IQ scores (from 76 to 85) to explore whether their intellectual profile could be influenced by the level of impairment. No specific hypothesis was advanced on this issue, but - in line with the main hypothesis of the study – there might be no differences between the two groups. This result would confirm and strengthen the idea of BIF identifying a specific population, not just a deviation in terms of IQ.

Finally, it was looked into age-related differences in the WISC-IV profile. The developmental trajectories of the main indexes were investigated. Children with BIF seem to develop more slowly than their typically-developing peers, but they show a certain stability in terms of their IQ (Jankowska, Bogdanowicz, & Takagi, 2014).

Experiences influence various aspects of functioning at different levels. The environment, for example, seems to have more effect on the crystallized component than on fluid intelligence (e.g. Duyme, Dumaret, & Tomkiewicz, 1999; Rindermann, Flores-Mendoza, & Mansur-Alves, 2010), with a higher impact on verbal skills. It could be that intellectual profiles remains similar across different ages, even though differences between the various indexes might gradually change, with verbal components gradually improving, also thanks to the positive effects of school and peer relationships, and perceptual reasoning (which is more associated with fluid abilities) gradually declining. This would reflect not a deterioration in intellectual functioning, but a slower rate of development, which would widen the gap vis-à-vis typical development, as seen in other clinical conditions, such as Down syndrome (Tsao & Kindelberger, 2009).

2. METHOD

2.1 Participants

Data on WISC-IV assessment of 204 children and adolescents attending mainstream primary and lower secondary schools, who were assessed at Italian child and adolescent - public and private - mental health services, were collected. All participants were assessed from 2013 to 2016 for screening or diagnostic purposes, due to learning or behavioral problems in the vast majority of cases, as reported by parents and/or teachers.

The inclusion criteria were: the availability of a complete WISC-IV assessment (with the 10 core subtests administered); a FSIQ between one and two standard deviations below the mean (from 70 to 85; the standard error was not considered in order to reduce overlaps); and attendance at primary and lower secondary school. The FSIQ had to be considered instead of the diagnosis because BIF is often not specified among the clinical diagnoses, by Italian mental health services at least (i.e. in many cases, the presence of BIF or an IQ in the borderline range was not reported in the clinical diagnosis).

The sample was between 6 and 15;6 years old (mean age = 9;11, SD = 28 months), and included 138 males and 66 females. This discrepancy between males and females is not surprising and it is consistent with other evidence of more males having intellectual disabilities (e.g. APA, 2013) and other clinical conditions (e.g. ADHD; Nøvik et al., 2006; Willcutt, 2012).

Most of the participants were Italian, with both parents Italian ($n = 131$); 10 were first-generation immigrants (children born abroad, who then migrated to Italy with their parents), or adopted by Italian natives ($n = 4$); and 53 were second-generation

immigrants, i.e. born in Italy from immigrant parents, or mixed Italian-immigrant parents. Information about nationality was missing for 6 participants.

Unfortunately no standardized assessment of adaptive functioning was available for the majority of the participants, but some qualitative information was available from interviews with parents. Given the specific recruitment process (i.e. data were collected in a clinical setting, and children had all been referred for clinical purposes), it is safe to say that all participants showed impairments in adaptive functioning, in terms of academic achievement at least.

The reason for their assessment was stated in 190 of the 204 clinical files considered. The most common reason was learning impairment or delay (about 71% of participants), followed by referred attention or behavioral problems (such as inattention, hyperactivity or deficit in impulse control, lack of respect for rules, difficulties in social interactions), often associated with referred learning difficulties (about 49% of participants). Other reasons for the clinical assessment were identified as developmental delays ($n = 6$), language impairment ($n = 5$), or gross and fine motor difficulties ($n = 2$). In some cases, the children were being evaluated following previous assessments ($n = 27$).

The parents or teachers providing information on individual functioning reported difficulties in communication skills ($n = 41$), social competence ($n = 70$), emotional control and motivation ($n = 52$), and self-care ($n = 33$). In many cases, the children were reportedly lacking in the ability to organize their own activities and materials ($n = 38$).

2.2 Instrument

The Italian standardization of the WISC-IV (Orsini, Pezzuti, & Picone, 2012) was used for the study. This is the most often used tool for assessing intellectual

functioning in individuals aged from 6 years to 16 years and 11 months. The WISC-IV is composed of 15 subtests, 10 core subtests, and 5 additional subtests. The last of these can be administered in addition to the core subtests to obtain more information on a child's intellectual functioning, or they can be used in lieu of the core subtests, subject to certain rules (see Manual). Like the American standardization (Wechsler, 2003), the Italian one confirms the four-factor structure of the scale.

Administering the 10 core subtests enables four main indexes to be computed, as concerns: Verbal Comprehension (comprising the Similarities, Vocabulary, and Comprehension core subtests), Perceptual Reasoning (with the Block Design, Picture Concepts, and Matrix Reasoning subtests), Working Memory (including the Digit Span and Letter-Number Sequencing subtests), and Processing Speed (consisting of the Coding and Symbol Search subtests).

A Full-Scale Intelligent Quotient (FSIQ) can also be calculated, which represents overall intellectual functioning.

No supplemental subtests were included in this study, because they were only available for a small number of children, and because they are not needed to calculate the main and additional indexes, and the FSIQ.

The various indexes and FSIQ are expressed in terms of standard scores with a mean of 100 and standard deviation of 15; subtest raw scores could be transformed into scaled scores, with a mean of 10 and a standard deviation of 3.

2.3 Data analysis plan

First participants were considered all together to seek a specific intellectual profile in BIF.

In a second step, to see whether the level of impairment was associated with the trend of the profile, the sample was divided into two groups on the basis of the children's FSIQ. One group included children with a FSIQ ranging from 70 to 75, the other involved the children with a FSIQ ranging from 76 to 85.

Finally, age was taken into consideration to examine the trajectories of the cognitive development of school-age children with BIF. Specifically, participants were divided into 3 groups by school grade, according to the Italian school cycles: the first group includes children attending their first and second year of primary school; the second group consisted of children enrolled in the third, fourth, and fifth years of primary school; and children attending lower secondary school (grades 6-8) formed the third group.

3. RESULTS

3.1 WISC-IV profile in BIF

Table 1 shows descriptive statistics for the WISC-IV indexes and subtests, as well as the results for the single-sample *t* tests comparing the BIF group's scores with those of the WISC-IV standardization sample (i.e. mean = 100 and standard deviation = 15, for composite indexes; mean = 10 and standard deviation = 3, for subtests).

Table 1

Means (standard deviations), and results of single-sample t-tests (expected mean 100 for composite indexes, and 10 for subtests)

	<i>M (SD)</i>	<i>t (df = 203)</i>	<i>p</i>
VCI	84.35 (9.39)	-23.799	< .001
PRI	88.37 (10.11)	-16.422	< .001
WMI	77.04 (9.67)	-33.913	< .001
PSI	83.46 (11.86)	-19.911	< .001
FSIQ	78.45 (4.46)	-68.987	< .001
GAI	84.38 (6.66)	-33.486	< .001
CPI	75.15 (9.58)	-37.052	< .001
<i>Subtest</i>			
SI	7.67 (2.02)	-16.468	< .001
VC	7.11 (2.02)	-20.458	< .001
CO	7.40 (2.60)	-14.275	< .001
BD	7.72 (2.58)	-12.610	< .001
PCm	8.53 (2.31)	-9.078	< .001
MR	8.41 (2.49)	-9.093	< .001
DS	6.08 (2.25)	-24.840	< .001
LN	6.25 (2.28)	-23.467	< .001
CD	6.97 (2.74)	-15.834	< .001
SS	7.41 (2.55)	-14.529	< .001

Note. VCI = Verbal Comprehension Index, PRI = Perceptual Reasoning Index, WMI = Working Memory Index, PSI = Processing Speed Index; FSIQ = Full-Scale IQ, GAI = General Ability Index, CPI = Cognitive Proficiency Index; SI = Similarities, VC = Vocabulary, CO = Comprehension, BD = Block Design, PCm = Picture Concepts, MR = Matrix Reasoning, DS = Digit Span, LN = Letter-Number Sequencing, CD = Coding, and SS = Symbol Search.

As shown in the table, the BIF group's performance was significantly worse than the standardization sample in all indexes and subtests.

From a descriptive point of view, the Perceptual Reasoning component appears to coincide with the highest scores, at the lower end of the normal range³. In the other cases, the indexes were below one standard deviation from the mean, with the lowest performance for Working Memory.

In order to analyze the WISC-IV profile, and particularly identify any differences between the four indexes, a repeated-measures ANOVA was run, considering the four main indexes (VCI, PRI, WMI, and PSI) as the within-group factor. Bonferroni's correction for multiple comparisons was applied.

The results showed a statistically significant main effect of Index, $F(3, 609) = 36.01, p < .001, \eta^2_p = .15$. Subsequent comparisons showed that participants performed better in Perceptual Reasoning than in Verbal Comprehension ($MDiff. = 4.03, p_{adjusted} = .002$), Working Memory ($MDiff. = 11.33, p_{adjusted} < .001$), or Processing Speed ($MDiff. = 4.92, p_{adjusted} < .001$). Working Memory differed significantly from Verbal Comprehension ($MDiff. = -7.30, p_{adjusted} < .001$) and Processing Speed ($MDiff. = -6.42, p_{adjusted} < .001$), the former coinciding with the lowest scores. No significant differences were found between Verbal Comprehension and Processing Speed ($p_{adjusted} = 1$).

A further repeated-measures ANOVA was run considering the GAI and CPI as the within-group factor. The main effect of index was statistically significant, $F(1, 203) = 89.87, p < .001, \eta^2_p = .31$. The analysis of these additional indexes showed higher scores in the GAI than in the CPI ($MDiff. = 9.24, p_{adjusted} < .001$), and both scores were more than one standard deviation below the mean. This result is not surprising, given

³ In the present thesis, the term "normal range" refers to scores above 85.

the initial hypothesis of a relatively broad impairment in working memory abilities, and it partly confirms the uneven profiles in the sample considered.

Finally, a repeated-measures ANOVA was run considering the GAI, CPI, and FSIQ as within factor. The results showed a statistically significant main effect of index, $F(2, 406) = 90.03, p < .001, \eta^2_p = .31$. Subsequent comparisons showed that the GAI was significantly higher than the FSIQ ($MDiff. = 5.94, p_{adjusted} < .001$), and the latter was significantly higher than the CPI ($MDiff. = 3.30, p_{adjusted} < .001$).

Cohen's (1988) d -values were calculated to analyze the effect size of the differences between the four main indexes, and between the two additional indexes.

The effect sizes were large for the comparison between the PRI and the WMI ($d = 1.11$), medium for the comparisons between the VCI and the WMI ($d = .68$), and between the PSI and the WMI ($d = .58$), while the other comparisons yielded small effect sizes ($d_{(VCI-PRI)} = -.37; d_{(VCI-PSI)} = .07; d_{(PRI-PSI)} = .41$). As for the comparison between the two additional indexes, the effect size was large ($d_{(GAI-CPI)} = .94$), confirming the discrepancy between the components of the FSIQ. The difference between the GAI and CPI is due largely to the much lower WMI, rather than to the influence of greater processing speed difficulties.

3.2 Is the profile stable across different levels of FSIQ?

The above-cited study by Cornoldi et al. (2014) indirectly suggested that the intellectual profile of individuals with BIF could change depending on their level of impairment. To seek any difference in the profiles between children with a FSIQ at the lower end of the borderline range and children with a "higher" FSIQ, participants were divided into two groups: one group ($n = 66$) had a FSIQ between 70 and 75; in the other ($n = 138$) it was between 76 and 85.

Table 2

Sex distribution and mean with standard deviation of chronological age, and WISC-IV indexes in Lower IQ (70-75) and Higher IQ (76-85) groups.

	Lower IQ (<i>n</i> = 66)		Higher IQ (<i>n</i> = 138)	
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>
Sex (m/f)	40/26		98/40	
Chronological age	121.52	27.73	118.44	28.37
FSIQ	73.09	1.90	81.01	2.71
VCI	81.74	10.31	85.59	8.69
PRI	83.04	9.89	90.93	9.20
WMI	73.86	9.83	78.57	9.25
PSI	78.26	10.05	85.95	11.89
GAI	80.09	6.13	86.44	5.90
CPI	69.68	8.91	77.76	8.78

Note. Chronological age in months.

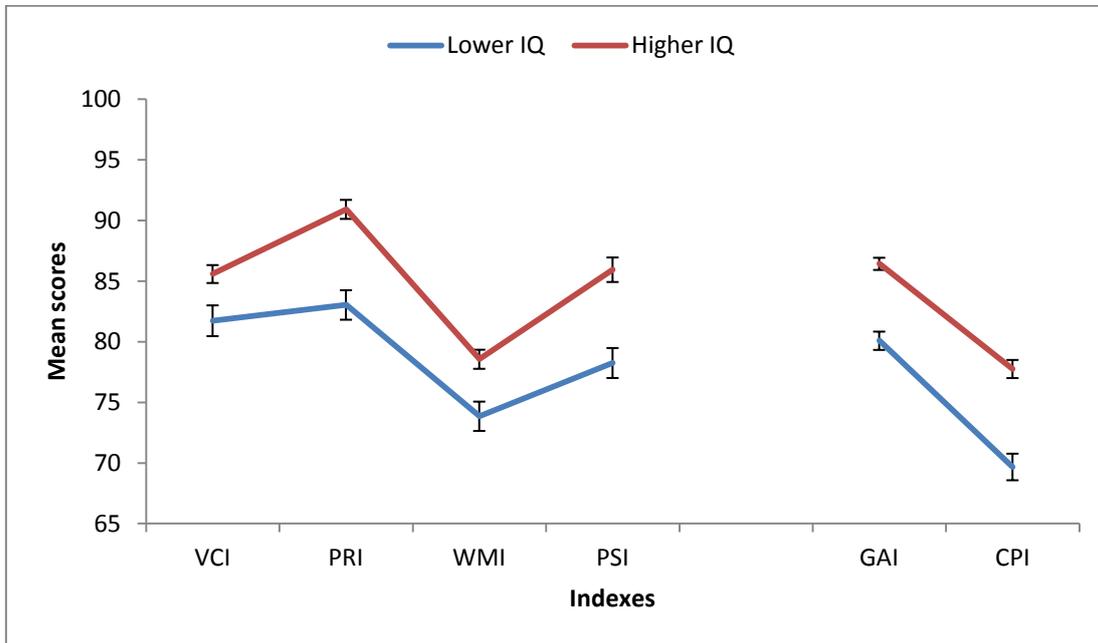
FSIQ = Full-Scale IQ; VCI = Verbal Comprehension Index, PRI = Perceptual Reasoning Index, WMI = Working Memory Index, PSI = Processing Speed Index; GAI = General Ability Index, CPI = Cognitive Proficiency Index

A 2 Groups x 4 Indexes mixed ANOVA was run, with FSIQ Group as the between factor and the Indexes as the within factor variables. Post-hoc comparisons were analyzed by applying Bonferroni's corrections for multiple comparisons. Not surprisingly, a statistically significant main effect of Group was found, $F(1, 202) = 349.76, p < .001, \eta^2_p = .63$, with the children with a higher IQ outperforming those with a lower IQ ($MDiff. = 6.03, p_{adjusted} < .001$). The main effect of Index was also significant $F(3, 606) = 29.14, p < .001, \eta^2_p = .13$, and showed a pattern similar to the one found for the overall sample, with lower scores for WM than for VC, PR, and PS ($MDiff. = -7.45,$

$p_{adjusted} < .001$; $MDiff. = -10.77$, $p_{adjusted} < .001$; $MDiff. = -5.89$, $p_{adjusted} < .001$, respectively), and higher scores in the PRI than in the VCI or PSI ($MDiff. = 3.32$, $p_{adjusted} = .03$; $MDiff. = 4.88$, $p_{adjusted} < .001$, respectively). The Group x Index interaction, $F(3, 606) = 1.52$, $p = .208$, $\eta^2_p = .007$, was not significant, indicating similar profiles in the two groups.

A similar pattern of results emerged considering the two additional indexes. The 2 x 2 mixed ANOVA showed a statistically significant main effects of Group, $F(1, 202) = 280.46$, $p < .001$, $\eta^2_p = .581$, and Index, $F(1, 202) = 83.85$, $p < .001$, $\eta^2_p = .29$. The children with a higher IQ outperformed those with a lower IQ ($MDiff. = 7.21$, $p_{adjusted} < .001$), and higher scores were obtained for GAI than for CPI ($MDiff. = 9.54$, $p_{adjusted} < .001$). The Group x Index interaction was not significant, $F(1, 202) = .69$, $p = .406$, $\eta^2_p = .003$. The profiles of the two groups are shown in Figure 1.

Figure 1 – Means and profiles for the two groups. Standard errors are represented in the figure by the error bars attached to each column.



Note. VCI = Verbal Comprehension Index, PRI = Perceptual Reasoning Index, WMI = Working Memory Index, PSI = Processing Speed Index; GAI = General Ability Index, CPI = Cognitive Proficiency Index

3.3 Analysis of the profile’s stability across age groups

Participants were divided into three groups, based on their school grade. Table 2 shows the demographic information and WISC-IV performance (in terms of the main and additional indexes) for the three groups.

First a one-way ANOVA was run to compare the three groups, and the results showed no significant differences in FSIQ between them, $F(2, 201) = 0.11, p = .894, \eta^2_p = .001$.

Table 3

Sex distribution and mean (with standard deviation) chronological age, and main and additional WISC-IV indexes, by age group (school cycles)

	1st cycle (n = 57) <i>M (SD)</i>	2nd cycle (n = 95) <i>M (SD)</i>	Lower secondary school (n = 52) <i>M (SD)</i>
Sex (M/F)	37/20	65/30	36/16
Chronological age	89 (8)	117 (13)	158 (14)
VCI	82.25 (9.86)	84.71 (8.55)	86.00 (10.08)
PRI	91.79 (9.06)	87.72 (9.96)	85.85 (10.65)
WMI	76.32 (9.63)	77.29 (9.91)	77.39 (9.40)
PSI	82.96 (12.54)	83.45 (11.04)	84.02 (12.74)
FSIQ	78.68 (4.15)	78.35 (4.45)	78.37 (4.88)
GAI	84.93 (6.94)	84.20 (6.27)	84.12 (7.13)
CPI	74.33 (9.24)	75.36 (9.30)	75.65 (10.53)

Note. Chronological age in months.

VCI = Verbal Comprehension Index, PRI = Perceptual Reasoning Index, WMI = Working Memory Index, PSI = Processing Speed Index; FSIQ = Full-Scale IQ, GAI = General Ability Index, CPI = Cognitive Proficiency Index

To find any differences in the WISC-IV profiles between the three groups, a 4 x 3 mixed ANOVA was run, with Index (VCI, PRI, WMI, and PSI) as the within-group factor, and Group (1st cycle, 2nd cycle, and lower secondary school) as the between-group factor. The main effects and the interactions were analyzed using post-hoc comparisons, and applying Bonferroni's correction for multiple comparisons.

The main effect of Index was significant, $F(3, 603) = 34.58, p < .001, \eta^2_p = .147$. As seen for the whole group, significantly higher scores were obtained for Perceptual Reasoning than for Verbal Comprehension, Working Memory, or Processing Speed ($MDiff. = 4.13, p_{adjusted} < .001$; $MDiff. = 11.45, p_{adjusted} < .001$; $MDiff. = 4.97, p_{adjusted} <$

.001, respectively), and for Verbal Comprehension and Processing Speed than for Working Memory ($MDiff. = 7.32$, $p_{adjusted} < .001$; $MDiff. = 6.48$, $p_{adjusted} < .001$, respectively), with no significant differences between the Verbal Comprehension and the Processing Speed indexes ($p_{adjusted} = 1$).

The effect of Group, $F(2, 201) = .002$, $p = .998$, $\eta^2_p < .001$, and the Index x Group interaction, $F(6, 603) = 2.048$, $p = .058$, $\eta^2_p = .02$, were not significant.

Similar results were obtained considering the two additional indexes, with a significant main effect of Index, $F(1, 201) = 84.49$, $p < .001$, $\eta^2_p = .296$: taken together participants obtained higher scores for the GAI than for the CPI ($MDiff. = 9.30$, $p_{adjusted} < .001$). Neither the effect of Group, $F(2, 201) = 0.05$, $p = .956$, $\eta^2_p < .001$, nor the Index x Group interaction, $F(2, 201) = 0.39$, $p = .678$, $\eta^2_p = .004$, were significant.

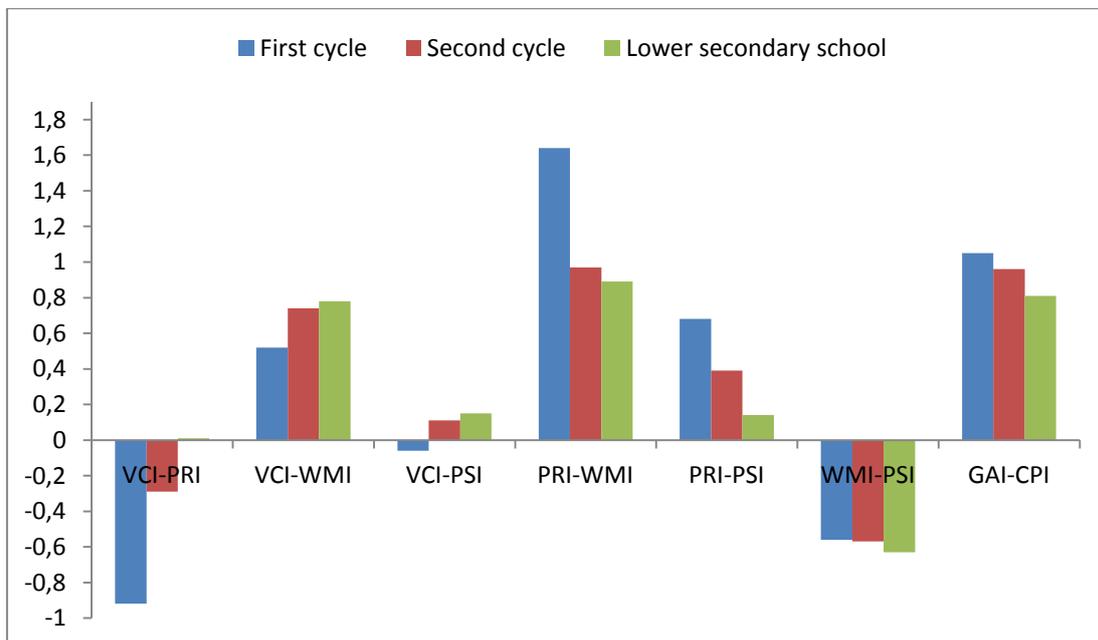
Cohen's (1988) d -values were calculated to analyze the effect size of the differences between the four main indexes, and between the two additional indexes, within each age group. Figure 2 shows the d -values obtained for each comparison in the three groups.

In the comparison between indexes in the First cycle group (grades 1-2), the effect sizes were large for the comparisons between the VCI and the PRI ($d = -0.92$), and between the PRI and the WMI ($d = 1.64$); they were medium for the comparisons between the VCI and the WMI ($d = 0.52$), the PRI and the PSI ($d = 0.68$), and between the PSI and the WMI ($d = 0.56$); and the comparison between the VCI and the PSI ($d = -0.06$) yielded a very small effect size. In the comparisons between the two additional indexes, the effect size was large ($d_{GAI-CPI} = 1.05$)

Similar patterns of effect sizes emerged for the differences between the indexes within the other two groups (i.e. Second cycle and Lower secondary school), i.e. with large effect sizes for the PRI-WMI comparisons ($d_{Second\ cycle} = 0.97$; $d_{Lower\ sec.\ school} =$

0.89); medium effect sizes for the comparisons between the VCI and the WMI ($d_{\text{Second cycle}} = 0.74$; $d_{\text{Lower sec. school}} = 0.78$), and between the PSI and the WMI ($d_{\text{Second cycle}} = 0.57$; $d_{\text{Lower sec. school}} = 0.63$); and very small effect sizes for the comparison between the VCI and the PSI ($d_{\text{Second cycle}} = 0.11$; $d_{\text{Lower sec. school}} = 0.15$). The only exceptions regard the comparisons between the PRI and the VCI, and between the PRI and the PSI, which revealed effect sizes that were small in the second cycle group ($d_{\text{PRI-VCI}} = 0.29$; $d_{\text{PRI-PSI}} = 0.39$), and very small in the Lower secondary school group ($d_{\text{PRI-VCI}} = -0.01$; $d_{\text{PRI-PSI}} = 0.14$). As in the first group, the effect sizes were large for the GAI and CPI comparisons ($d_{\text{Second cycle}} = .96$; $d_{\text{Lower sec. school}} = 0.81$).

Figure 2 – Comparisons between the WISC-IV indexes by age group (school cycles), using Cohen’s d .



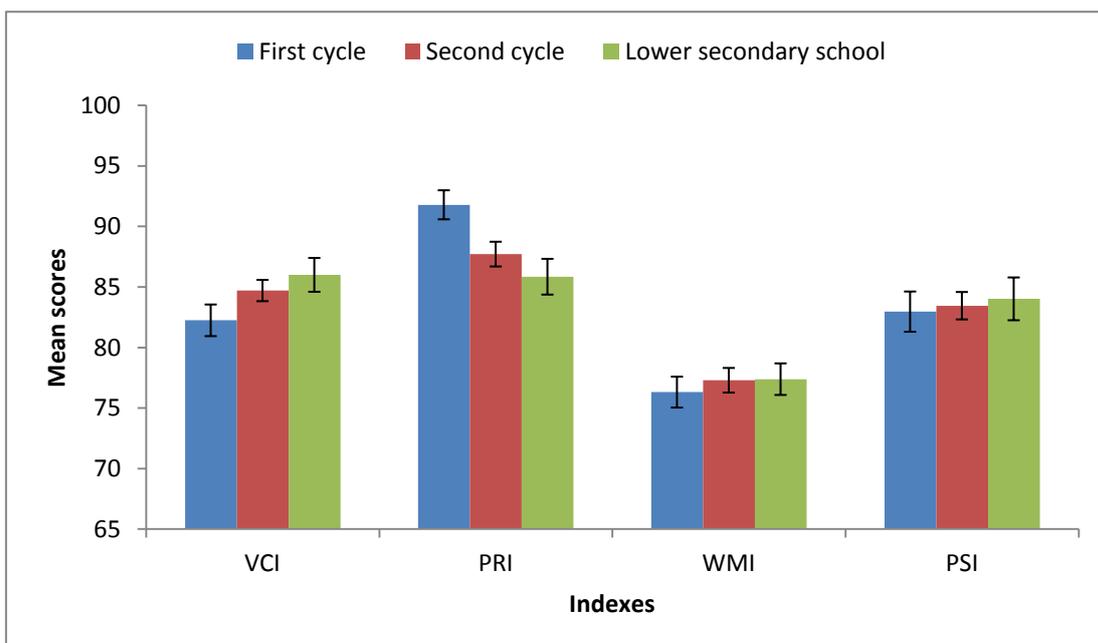
Note. VCI = Verbal Comprehension Index, PRI = Perceptual Reasoning Index, WMI = Working Memory Index, PSI = Processing Speed Index; GAI = General Ability Index, CPI = Cognitive Proficiency Index

3.3.1 Trajectories of the indexes

To explore changes in each component across different age groups, a series of one-way ANOVAs was run, considering age groups as independent variable and index as the dependent variable. The results showed no differences between the three age groups in the Verbal Comprehension, Working Memory and Processing Speed indexes ($p = .10$, $p = .799$, $p = .899$, respectively), while there was a statistically significant difference in Perceptual Reasoning between the groups, $F(2, 201) = 5.30$, $p = .006$, $\eta^2_p = .05$. Subsequent post-hoc comparisons with Bonferroni's correction showed that the younger children outperformed those in grades 3-5 (Second cycle) ($MDiff. = 4.07$, $p_{adjusted} = .045$), and those in grades 6-8 (Lower secondary school) ($MDiff. = 5.94$, $p_{adjusted} = .006$), while the latter two did not differ ($p_{adjusted} = .83$).

Figure 3 shows the developmental trajectory of each index.

Figure 3 – Means of the WISC-IV indexes across the different age groups. Standard errors are represented in the figure by the error bars attached to each column.



Note. VCI = Verbal Comprehension Index, PRI = Perceptual Reasoning Index, WMI = Working Memory Index, PSI = Processing Speed Index.

4. DISCUSSION

The present study analyzed the intellectual profile of school-age children with BIF attending Italian primary and lower secondary schools.

Apart from their below-average general intellectual functioning, little is known about the intellectual profile of individuals with BIF. This study aimed to examine whether BIF is associated with a specific WISC-IV profile.

This scale was chosen because it is the most often used tool for assessing intelligence in children and adolescents between 6 and 16 years old (e.g. Bremner et al., 2011), and because it provides a more complete picture of their intellectual abilities, considering different aspects of intellectual functioning – unlike other tests such as Raven’s progressive matrices (e.g. Raven et al., 1998).

The developmental trajectories of different components of intellectual functioning were analyzed. The results confirmed that the BIF profile differs from that of typical development, both quantitatively and qualitatively.

First of all, individuals with BIF had lower scores in all four WISC-IV indexes examined than those expected of a typical population (on the basis of standardization criteria i.e. the scores for the four main indexes, the FSIQ, and the two additional indexes are given on a distribution with a mean of 100 and a standard deviation of 15). Moreover, the profile revealed relative strengths and weaknesses, rather than being flat. This profile showed differences between almost all the indexes, except for the comparison between verbal comprehension and processing speed, which were similar. The trend was confirmed by the additional indexes, with a significant difference between the GAI and the CPI.

Among the four main indexes in the WISC-IV, the BIF profile coincided with highest scores for Perceptual Reasoning. This index concerns tasks that primarily measure nonverbal fluid reasoning and perceptual organization abilities (Wechsler, 2003). The lowest scores emerged in the Working Memory Index, confirming the impairment of this cognitive ability in BIF, as reported in several studies. Difficulties were found in both verbal and visuo-spatial components when individuals with BIF were compared with typically-developing peers (e.g. Alloway, 2010, Schuchardt et al., 2010).

Poor working memory may be considered a commonality with other clinical conditions, such as intellectual disability and specific learning disorders, but the WISC-IV profile that emerged differs from the one found in previous studies on intellectual disability and specific learning disorders (e.g. Cornoldi et al., 2014). Intellectual disability seems to be characterized by an overall impairment in WISC-IV components, showing an almost flat profile. Specific learning disorders, on the whole, have a profile showing strengths in verbal comprehension and perceptual reasoning (which are not discrepant with each other), and weaknesses in working memory and processing speed, which are similar in terms of mean scores (Cornoldi et al. 2014).

A similar picture emerged when different levels of FSIQ were considered, indicating that these peculiar traits persist for different levels of severity of the intellectual impairment. Given the overlap in terms of IQ (due to the standard error) between the group with an IQ of 70-75 and individuals with mild intellectual disability, this result again confirms the differences between the two conditions, and suggests that BIF is neither the least severe form of intellectual disability, nor at the bottom of the distribution of typical intelligence.

The BIF profile was not significantly different between the three different age groups considered, between 6 and 15 years old at least. When the developmental trajectories of the indexes were considered, some differences emerged between the various components of intellectual functioning. The only significant change with age concerned Perceptual Reasoning, which gradually decreased, particularly between the youngest children (mean 7;5 years old) and the other two groups (mean 9;9 and 13;2 years old, respectively), which did not differ significantly from each other. This result is at odds with a follow-up study by Jankowska et al. (2014) on 30 children with BIF, administered with the WISC-R at 8, 10;8, and 13;6 years of age. The authors found a slight improvement in non-verbal IQ, while verbal IQ declined with age. It is noteworthy that, in the study by Jankowska et al. (2014), the improvement in non-verbal intelligence was only significant between 8 and 10;8 years old, while it slightly decreased again between 10;8 and 13;6 years old. It is important to emphasize that a different test was used, however. Indeed, the WISC-R non-verbal scale does not include exactly the same subtests as the Perceptual Reasoning Index in the WISC-IV. Unlike the present study, moreover, Jankowska et al. conducted a longitudinal study, with participants assessed three times.

It may be that the decrease seen in Perceptual Reasoning in the present study is associated with a slowing in relation to typical development, with a consequent widening of the gap between children with BIF and typically developing children as they grow older (standard scores were considered for the purposes of the present study). Taken together, the present results would confirm that the BIF population has a specific cognitive profile, with its own peculiar characteristics. This means that it is important to better differentiate BIF from either intellectual disability or other clinical conditions

(such as specific learning disorders), both in research and in clinical and educational practice.

What could the implications be from a clinical and educational point of view?

First, obtaining information about a profile “specific” to a certain condition could help clinicians and other professionals plan appropriate training based on strengths and weaknesses. As written previously, BIF is often misrecognized, and masked by other clinical disorders. In such cases it is important to understand whether BIF is the primary cause of an individual’s problems, or whether it is a comorbid, secondary condition (Ninivaggi, 2009). In other words, we need to know whether a child’s difficulties that prompt a clinical assessment (e.g. delayed or impaired academic achievement, behavioral problems, attention problems) are due to a lower IQ or have other origins.

Considering the WISC-IV profile could help to differentiate individuals with BIF from those with mild intellectual disability, particularly when the FSIQ is at the bottom end of the borderline range (i.e. between 70 and 75) or at the top end of the range for intellectual disability.

A limitation of the present study concerns the absence of a control group of typically developing children, though the use of standardized WISC-IV scores presumably suffices to confirm the results, bearing in mind that the mean of each index in a typical population is, by definition, 100.

Finally, it would be useful to see whether a cognitive profile similar to the one found in the present study would also emerge using different tools to assess the same components as those examined by the WISC-IV.

CHAPTER 3

STUDY 2:

COGNITIVE PROFILES IN BORDERLINE INTELLECTUAL FUNCTIONING: ANALYSIS OF SUBTYPES

1. INTRODUCTION

Borderline intellectual functioning (BIF) is a condition with different etiologies and a distinctive cognitive phenotype of individuals with BIF is still not clear (Di Blasi et al., 2014).

No single cause has been found at the origin of BIF (Ninivaggi, 2009). Like intellectual disability, BIF is not a syndrome, but rather the end result of different causal factors (e.g. Vianello et al., 2014). BIF is neither a mental disorder nor an intellectual disability (e.g. Ferrari, 2009; Jankowska, 2016; Vianello, 2008), but it is recognized as a condition that may require clinical attention (e.g. APA, 2013), and it is often associated with other disorders (e.g. Salvador-Carulla et al., 2013; Vianello et al., 2014).

As stated by the American Association on Intellectual and Developmental Disabilities, the causes of intellectual disabilities may be distinguished on the basis of

their nature, e.g. biomedical, social, behavioral, or educational, and of the period of exposure, i.e. prenatal, perinatal, and postnatal. This is probably true of the causes and risk factors for BIF too.

Specifically, BIF may have biological (genetic or not genetic) and environmental causes (e.g. Ninivaggi, 2009; Peltopuro et al., 2014; Farhadifar et al., 2011). Among the environmental causes, socio-economic and cultural disadvantage seems to have a negative influence on intellectual functioning, leading to a lower intellectual quotient (e.g. Duyme et al., 1999; Vianello, 2012). Research has suggested that environmental influences are not uniform across different neurocognitive systems and they seem more marked on some components or abilities than others (e.g. Eilertsen et al., 2016; Farah et al., 2006). BIF could also be caused by the indirect effects of other neurobiological disorders, such as specific learning disorder (SLD), attention deficit and hyperactivity disorder (ADHD), or language and communication disorder, or by the negative influence of emotional problems on intellectual functioning (Vianello et al., 2014).

More than one cause could be behind a poor intellectual functioning in the same person (e.g. Vianello et al., 2014). BIF may be associated with other developmental disorders, such as SLD, ADHD, or autism spectrum disorders, but also with genetic syndromes characterized by a broad variability in intellectual functioning like the Fragile X, Prader-Willi, and Williams syndromes (e.g. Ninivaggi, 2009; Salvador-Carulla et al., 2013).

The different conditions associated with BIF, and/or its different causes conceivably engender or influence heterogeneous cognitive and behavioral profiles. A deeper understanding of the numerous phenotypic manifestations of BIF could help in the planning of adequate training or support measures.

For example, Vianello et al. (2014), hypothesized that different subtypes of BIF could be recognized by considering the causes behind the condition. Specifically, we can distinguish between a type of BIF due to genetic causes, another due to non-genetic biological causes, a type of BIF associated with or caused by socio-economic disadvantage, one due to the indirect effects of SLD, ADHD or other neurodevelopmental disorders, another due to intellectual inhibition caused by motivational or affective issues, and finally a “pure” type of BIF. Each type has partially different behavioral and cognitive characteristics (Vianello, 2008; Vianello et al., 2014).

In the field of neuropsychological research, several studies have aimed to investigate the inter-individual variability of different disorders and clinical conditions. Given the observation that, beyond some common features or symptoms characteristic of each specific condition and enabling its diagnosis, individuals may be extremely diverse in their cognitive and/or behavioral or adaptive functioning (e.g. Goldstein, 2013). Some attempts have been made to clarify the variability within clinically-definable neuropsychological disorders by analyzing subgroups with Autism Spectrum Disorders, SLD, traumatic brain injury, and schizophrenia, for example. Brennan, Barton, Chen, Green, and Fein (2015) investigated whether subgroups could be identified among children with pervasive developmental disorder – not otherwise specified. They found three possible clusters, characterized by different patterns of symptoms, and cluster membership at 2 years old had a predictive value on outcome at the age of 4.

Another attempt to explore inter-individual variability by analyzing subgroups involved children with Down syndrome (Tsao & Kindelberger, 2009). The Authors suggested that the broad variability characterizing this syndrome could be associated

also to the presence of homogeneous subgroups with different patterns of cognitive development.

The present study aimed to find evidence of different subgroups of BIF. Based on the WISC-IV main indices, we attempted to find homogeneous subgroups characterized by different patterns of cognitive functioning in an effort to contribute to research on BIF and the understanding of this condition from a clinical point of view. Clarifying the characteristics of children with diverse presentations enables a more clear and comprehensive understanding of the unique profiles and needs of children in this diagnostic category (Brennan et al., 2015). This might shed some light on the broad inter-individual variability characteristic of BIF that makes it difficult to obtain a clear profile of this condition (e.g. Di Blasi et al., 2014; Salvador-Carulla et al., 2013).

Using WISC-IV profiles, Thaler et al. (2012) analyzed the association between intellectual profiles and diagnostic frequency, symptoms, and outcome in children with ADHD (inattentive and combined subtypes), finding different clusters that might be useful for predicting the symptoms and functional outcome in children with ADHD. Similar results were obtained by Fenollar-Cortes et al. (2015), who compared inattentive and combined ADHD subtypes. They found differences in the WISC-IV profiles, particularly for working memory and processing speed, that showed opposite trends between the two groups.

Other authors compared different specific learning disorders (coming under the diagnostic category of Specific Learning Disorder in the DSM-5; APA, 2013) in terms of their WISC-IV profiles, and found subgroups characterized by partially different profiles. For example, Poletti (2016) studied the intellectual profile of Italian children with a clinical diagnosis of SLD assessed with the WISC-IV, comparing them by category (i.e. reading, written expression, or mathematics disorder, isolated or in

combination). The author found partially different cognitive profiles between these subtypes of SLD, with the exception of writing and reading disorders, which did not differ from each other.

Similarly, Toffalini et al. (2017) found some shared features, and some different patterns in the intellectual abilities of children with different SLD subtypes. The authors compared the WISC-IV profiles of different SLD categories (i.e. identified with the ICD-10 coding system as: specific reading disorder, specific spelling disorder, specific disorder of arithmetical skills, and mixed disorder of scholastic skills). Their results showed similar weaknesses across the categories, with the working memory and processing speed indexes always obtaining the lowest scores, while the verbal comprehension and perceptual reasoning indexes were relatively more differentiated across the categories. For example, the perceptual reasoning index was higher than the verbal comprehension index in the reading disorder subtype, and vice versa in the children with an arithmetical disorder.

Unlike these other clinical conditions, no different subtypes of BIF have been clinically defined to date. The only information available comes from an unavailable work cited by Jankowska (Jankowska, 2011, cited in Jankowska, 2016), who suggested that there are at least three different profiles of BIF, judging from the WISC-R scores. One profile is flat, with an overall performance ranging between one and two standard deviations below average. A second cognitive profile is more uneven, characterized by more impaired and below average verbal abilities, while performance IQ is relatively well preserved, on the threshold of the normal range. A third profile is spiky, with impairments in both verbal and non-verbal abilities, but more severe weaknesses in the Arithmetic, Coding, Information, and Digit Span WISC-R subtests.

Several changes have been made to the Wechsler scales since the WISC-R (Wechsler, 1974). One of particular relevance for the present study is that the WISC-R could be used to compute a verbal IQ (which also included a subtest for assessing working memory), and a performance IQ (which included a subtest of processing speed), while the WISC-IV has abandoned this dual structure in favor of a four-factor structure, and a broader consideration of working memory and processing speed. This poses the question of whether using the WISC-IV scale - with its four-factor structure to measure the different cognitive abilities influencing overall intellectual functioning - will produce similar profiles to those suggested by Jankowska (2016).

Cornoldi et al. (2014) found that individuals with BIF and SLD had an intellectual profile similar to that of children with SLD and average intelligence, with higher scores for verbal comprehension and perceptual reasoning than for working memory and processing speed. In the same study, the authors also had a small subgroup of individuals with a clinical diagnosis of intellectual disability, but an IQ in the borderline range. This last subgroup showed a flat profile, with no significant differences between the indexes (Cornoldi et al., 2014). Similar results were found by Bremner et al. (2011), who found no significant differences between the general ability index and the cognitive proficiency index in their subgroup of children with BIF, suggesting a flat profile. These findings support the hypothesis that it is possible to find different subgroups of BIF, in terms of WISC-IV profiles.

In the light of these considerations, the present study aimed to analyze the existence of different subgroups, within the broad category of BIF, characterized by a homogeneous pattern of intellectual strengths and weaknesses.

2. METHOD

Archival data held by several private and public mental health services for children and adolescents that agreed to participate in the study were screened to extrapolate data for school-age children attending primary and lower secondary school with a FSIQ score of between 70 and 85 on administration of the WISC-IV. The sample's WISC-IV scores, socio-demographic details (i.e. parents' education and occupations), and clinical information (i.e. clinical diagnoses) were collected.

2.1 Participants

Participants were the same 204 school-age children involved in study 1 (for a more detailed description of the sample, see chapter 2).

The main indexes obtained by administering the 10 core subtests of the WISC-IV were used for the purposes of the present study. Standardized scores were considered.

2.2 Data analysis plan

To seek to distinguish between subgroups with BIF, different cluster analysis procedures were used, considering the four main WISC-IV indexes (i.e. Verbal Comprehension, Perceptual Reasoning, Working Memory, and Processing Speed). The SPSS (v. 23) was used for the statistical analysis.

Cluster analysis is a grouping technique that enables homogenous subgroups to be identified within diverse samples based on shared characteristics or similarities (Allen & Goldstein, 2013). The aim of cluster analysis is to create the best possible grouping of observations such that the degree of association between observations within a given group is maximized, while the association or similarity between different

clusters is minimized (Cross, 2013). Since cluster analyses may be considered an exploratory technique, some precautions are necessary. As stated by Thaler et al. (2012), for a clustering solution to be relevant, the cluster obtained has to be theoretically interpretable, comprise at least 10% of the overall sample, and remain stable with alternative clustering solutions. There are different cluster methods available, and applying them to the same data may yield different results (Fisher et al., 2000). One of the approaches suggested to establish the stability of a clustering solution is to run different clustering procedures on the same dataset and cross-tabulate the solutions. This enables a check on whether different clustering procedures result in similar profiles, and whether high percentages of cases are assigned to the same cluster.

In the present study, both hierarchical and non-hierarchical cluster analyses were conducted, following the method used by other authors (e.g. Fisher et al., 2000; Wasserman & Holmbeck, 2016), to identify and confirm the number of subgroups.

In a first step, a hierarchical agglomerative cluster analysis was run to identify a clustering solution, with no restriction on number. Squared Euclidean distance was used as a similarity measure, and Ward's method (Ward, 1963) was chosen for the first cluster analysis, as an exploratory procedure, to be compared with other methods. This clustering method begins with each observation represented as a single cluster, then cases are grouped stepwise until the variance among observations within each cluster reaches a defined minimum level (Cross, 2013). Ward's method was chosen for this purpose, because it is commonly used in social science and neuropsychological research (e.g. Allen et al., 2010; Goldstein, 2013; Lewandowski, Sperry, Cohen, & Öngür, 2014; Thaler et al., 2012). Another hierarchical clustering analysis was conducted with the average-linkage, within-group method to validate the first cluster solution. Then a non-hierarchical cluster procedure was used to validate the solution using Ward's method.

K-Means clustering techniques are designed to group items or subjects into a specified number of clusters. The different solutions obtained with the different methods were then compared with those derived using Ward's method to assess the stability of the clusters.

Once the cluster solution had been identified (using the results obtained with Ward's method), a mixed ANOVA was run with Group (i.e. clusters) as between factor and Index (i.e. VCI, PRI, WMI, and PSI) as within factor to explore any difference in the profiles of the three groups obtained. Additional indexes were considered as well for the sake of completeness.

3. RESULTS

Visual inspection of the dendrogram suggested solutions with two, three or four clusters. Specifically, there seemed to be two macro-groups, which were then divided into groups, generating three or four clusters. All the possible solutions (with two, three, or four clusters) were then considered to establish which clustering solution best fitted the data. For every solution, each participant was recorded as a member of a given cluster. The four-cluster solution yielded four groups characterized by sample sizes of 45, 26, 82, and 51 participants. The three-cluster solution combined two groups in the four-cluster solution into a single cluster, generating three groups comprising 45, 77, and 82 participants. Finally, the two-cluster solution yielded a first cluster that coincided with the group of 82 participants that emerged for the four- and three-cluster solutions, and combined the other two (or three) groups into the second cluster.

When the solutions obtained with the average-linkage method, used as a confirmatory procedure, were compared with their counterparts generated using Ward's method, it emerged that 62% of participants were classified in similar clusters (i.e. as having similar cognitive profiles) (with a kappa agreement of .49) when the four-cluster solution was considered, while this was true of 83% (kappa agreement .74) for the three-cluster solution, and 95% (kappa agreement .89) for the two-cluster solution. Finally, three K-means cluster analyses were run for the four-, three- and two-cluster solutions. Comparing the different groups generated with Ward's method with those obtained with the K-means analyses, the two methods classified 55%, 79%, and 60% of the participants as having similar cognitive profiles for the four-, three-, and two-cluster solutions, respectively.

In the light of the results of the two confirmatory analyses (i.e. hierarchical, average-linkage, and non-hierarchical, K-means), the three-cluster solution was considered for further investigation. It seemed more interesting from a clinical standpoint because the three groups were of clinical interest. Table 1 shows the characteristics of the three clusters in terms of sample's chronological age, and mean scores and standard deviations in the main WISC-IV indexes.

Table 1

Means and standard deviations for the sample's chronological age and scores on the WISC-IV indexes, in the three-cluster solution.

	Cluster 1 (<i>n</i> = 45)		Cluster 2 (<i>n</i> = 77)		Cluster 3 (<i>n</i> = 82)	
	M	SD	M	SD	M	SD
Chronological age	117.91	26.95	118.88	27.64	120.79	29.46
VCI	76.53	5.11	83.62	7.95	89.32	9.40
PRI	95.62	7.59	81.26	7.67	91.09	9.22
WMI	80.67	8.43	77.48	10.16	74.65	9.25
PSI	86.04	6.47	92.35	10.07	73.69	7.71
FSIQ	79.58	4.23	78.03	4.53	78.22	4.47
GAI	83.84	5.17	80.10	5.62	88.69	5.54
CPI	78.69	5.98	81.05	8.11	67.66	7.21

Note. Chronological age in months.

VCI = Verbal Comprehension Index, PRI = Perceptual Reasoning Index, WMI = Working Memory Index, PSI = Processing Speed Index; FSIQ = Full-Scale IQ, GAI = General Ability Index, CPI = Cognitive Proficiency Index.

The three groups did not differ in terms of chronological age, $F(2, 201) = 0.175$, $p = .84$, $\eta^2_p = .002$, or FSIQ, $F(2, 201) = 1.91$, $p = .15$, $\eta^2_p = .019$.

On a descriptive level, participants in the first group had an uneven profile characterized by higher scores in Perceptual Reasoning, which were within the normal range, and a performance in Verbal Comprehension and Working Memory tasks in the borderline range (the former being particularly low). Scores for Processing Speed were on the threshold between borderline and normal.

Participants in the second group generally had a similar performance in Verbal Comprehension and Perceptual Reasoning, with both in the borderline range, as well as Working Memory, which was characterized by the lowest scores. On the other hand, participants in this group were on average for Processing Speed.

The third group consisted of children who performed similarly on Verbal Comprehension and Perceptual Reasoning tasks (both in the normal range), while Working Memory and Processing Speed were both weak, and to much the same degree (both in the borderline range).

To better analyze the differences between the three profiles, a 3 x 4 mixed ANOVA was run with Group (the three clusters) as the between-group factor, and Index (VCI, PRI, WMI, and PSI) as the within-group factor. The main effects and the interactions were analyzed using post hoc analyses, and applying Bonferroni's adjustment for multiple comparisons.

The main effect of Group was statistically significant, $F(2, 201) = 8.65, p < .001, \eta^2_p = .08$. Subsequent comparisons showed that the first and second groups outperformed the third ($MDiff. = 2.53, p_{adjusted} < .001$; $MDiff. = 1.49, p_{adjusted} = .021$, respectively), with no statistically significant difference between the first and second ($p_{adjusted} = .32$).

The main effect of Index, $F(3, 603) = 52.97, p < .001, \eta^2_p = .209$, was significant. Participants' performance was generally better on Perceptual Reasoning than on the three other indexes ($MDiff_{PRI-VCI} = 6.17, p_{adjusted} < .001$; $MDiff_{PRI-WMI} = 11.73, p_{adjusted} < .001$; $MDiff_{PRI-PSI} = 5.29, p_{adjusted} < .001$), better on Verbal Comprehension than on Working Memory ($MDiff. = 5.56, p_{adjusted} < .001$), and better on Processing Speed than on Working Memory ($MDiff. = 6.43, p_{adjusted} < .001$), while no

significant differences emerged between Verbal Comprehension and Processing Speed ($p_{adjusted} = 1$).

The Group x Index interaction was significant too, $F(6, 603) = 52.76, p < .001, \eta^2_p = .344$. Subsequent post-hoc comparisons, after applying Bonferroni's adjustment for multiple comparisons, showed that participants in the first cluster performed significantly better on Perceptual Reasoning than on the three other indexes ($MDiff_{PRI-VCI} = 19.09, p_{adjusted} < .001$; $MDiff_{PRI-WMI} = 14.96, p_{adjusted} < .001$; $MDiff_{PRI-PSI} = 9.58, p_{adjusted} < .001$), and better in Processing Speed than in Verbal Comprehension tasks ($MDiff = 9.51, p_{adjusted} < .001$), with no significant differences between the other components.

Participants in the second cluster performed significantly better on Processing Speed than on Verbal Comprehension, Perceptual Reasoning, or Working Memory ($MDiff_{PSI-VCI} = 8.73, p_{adjusted} < .001$; $MDiff_{PSI-PRI} = 11.09, p_{adjusted} < .001$; $MDiff_{PSI-WMI} = 14.87, p_{adjusted} < .001$), and better in Verbal Comprehension than in Working Memory tasks ($MDiff = 6.14, p_{adjusted} < .001$). There were no significant differences between the Verbal Comprehension and Perceptual Reasoning ($p_{adjusted} = 1$), nor between the Working Memory and Perceptual Reasoning indexes ($p_{adjusted} = .23$).

Finally, the profile of participants in the third cluster featured no significant differences between Verbal Comprehension and Perceptual Reasoning abilities ($p_{adjusted} = 1$), both of which were significantly better than working memory or processing speed ($MDiff_{VCI-WMI} = 14.67, p_{adjusted} < .001$; $MDiff_{VCI-PSI} = 15.62, p_{adjusted} < .001$; $MDiff_{PRI-WMI} = 16.44, p_{adjusted} < .001$; and $MDiff_{PRI-PSI} = 17.39, p_{adjusted} < .001$). No significant differences emerged between performance in the Working Memory and Processing Speed tasks ($p_{adjusted} = 1$).

The differences between the three clusters were also analyzed for each index. For Verbal Comprehension, participants in cluster 3 outperformed participants in clusters 1 and 2 ($MDiff. = 12.78, p_{adjusted} < .001$; $MDiff. = 5.69, p_{adjusted} < .001$, respectively), and participants in cluster 2 outperformed participants in cluster 1 ($MDiff. = 7.09, p_{adjusted} < .001$). For Perceptual Reasoning, participants in cluster 1 outperformed participants in clusters 2 and 3 ($MDiff. = 14.36, p_{adjusted} < .001$; $MDiff. = 4.54, p_{adjusted} = .048$, respectively), while participants in cluster 3 outperformed those in cluster 2 ($MDiff. = 9.83, p_{adjusted} < .001$). Different patterns emerged for Working Memory: there was only a significant difference in the comparison between cluster 1 and cluster 3, with the former obtaining higher scores than the latter ($MDiff. = 6.02, p_{adjusted} = .012$). As for Processing Speed, participants in cluster 2 obtained significantly higher scores than those in clusters 1 and 3 ($MDiff. = 6.31, p_{adjusted} < .001$, $MDiff. = 18.66, p_{adjusted} < .001$, respectively), and participants in cluster 1 outperformed those in cluster 3 ($MDiff. = 12.35, p_{adjusted} < .001$).

A subsequent 3 x 2 mixed ANOVA was run, with Group (the three clusters) as the between-group factor, and Index (i.e. the two additional indexes, GAI and CPI) as the within-group factor, to analyze the trend in the two additional indexes. Post-hoc comparisons were analyzed using Bonferroni's adjustment for multiple comparisons.

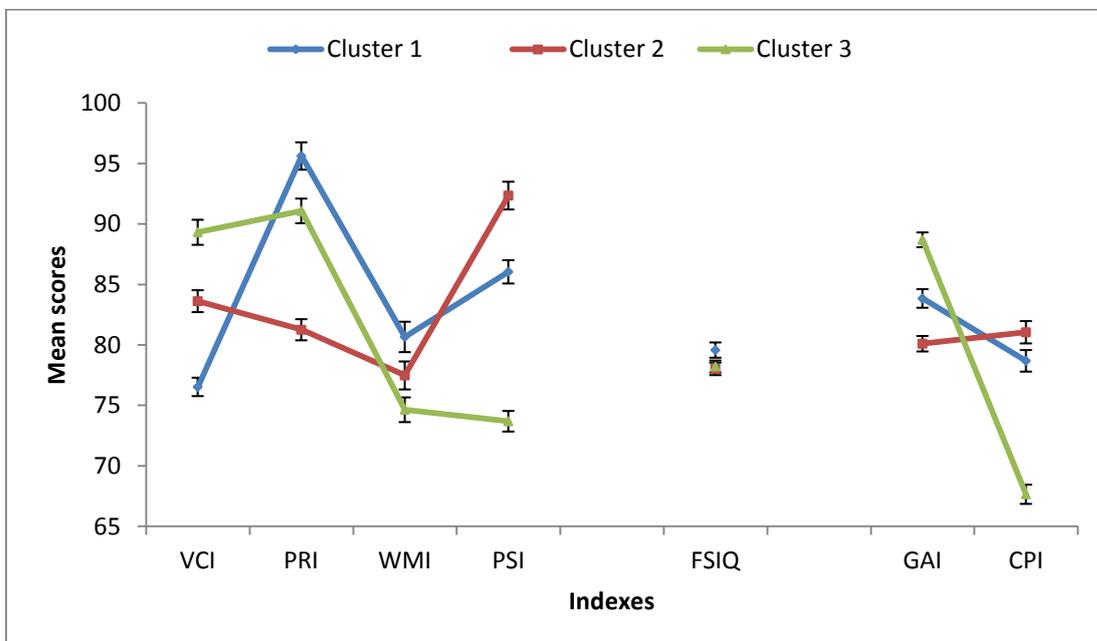
The main effect of Group, $F(2, 201) = 9.93, p < .001, \eta^2_p = .09$, was significant. Participants in cluster 3 were outperformed by those in clusters 1 and 2 ($MDiff. = -3.09, p_{adjusted} < .001$; $MDiff. = -2.40, p_{adjusted} = .001$, respectively), while participants in the first and second clusters showed no statistically significant difference in their performance ($MDiff. = .69, p_{adjusted} = 1$).

The main effect of Index was significant too, $F(1, 201) = 141.11, p < .001, \eta^2_p = .412$. Generally speaking, participants performed better on the GAI than on the CPI ($MDiff. = 8.42, p_{adjusted} < .001$).

The Group x Index interaction was significant, $F(2, 201) = 105.78, p < .001, \eta^2_p = .513$. Subsequent post-hoc comparisons showed that clusters 1 and 3 scored higher on the GAI than on the CPI ($MDiff. = 5.16, p_{adjusted} < .001$; $MDiff. = 21.04, p_{adjusted} < .001$, respectively), while the scores obtained by participants in cluster 2 did not differ significantly between the two additional indexes ($p_{adjusted} = 1$).

Figure 1 shows the profiles of the three groups in terms of the main and additional indexes.

Figure 1 - WISC-IV profiles of the three clusters in terms of main and additional indexes. Standard errors are represented in the figure by the error bars.



Note. VCI = Verbal Comprehension Index, PRI = Perceptual Reasoning Index, WMI = Working Memory Index, PSI = Processing Speed Index; FSIQ = Full-Scale IQ, GAI = General Ability Index, CPI = Cognitive Proficiency Index

3.1 Additional analyses

After finding the three profiles associated with the three clusters, it was explored whether the three clusters were characterized by different distributions of contextual or clinical variables. Given the frequent association between BIF and learning and behavioral disorders or difficulties (e.g. Salvador-Carulla et al., 2013), and between BIF and low socio-economic status (e.g. Emerson et al., 2010; Vianello et al., 2014), the family's socio-economic status and the children's clinical diagnoses were considered.

Socio-economic status (SES) was judged from the parents' educational level and occupational level. In line with Miconi, Moscardino, Ronconi, and Altoè (2017), the educational level was considered as the highest level of formal education achieved by either of the parents, and classified on three levels: low (i.e. elementary school or less), medium (i.e. secondary education/middle school), and high (i.e. pre-university education or college/university). The occupational level of the family was considered as the highest level of occupation of either parent, and classified as: low (i.e. unskilled work), medium (i.e. skilled work or white collar), and high (professional work). Educational and occupational levels were then averaged, and three categories were created for our purposes: low, medium, and high SES. Information about the family's SES was available for 153 of the 204 participants. Missing data were evenly distributed in the three clusters, and concerned 13 of 45 participants in the first cluster (28.9%), 17 of 77 in the second (22.08%), and 21 of 82 in the third cluster (25.6%). Overall, 64 children were from low SES families, 45 from medium SES families, and 44 from high SES families. Table 2 shows the distribution of the three SES levels within each cluster.

Table 2

Characteristics of the three clusters in terms of SES (i.e. low, medium, and high level) and clinical status (i.e. with versus without an associated clinical diagnosis other than BIF).

		Cluster 1	Cluster 2	Cluster 3
		n (%)	n (%)	n (%)
SES	Low	18 (56.3)	25 (41.7)	21 (34.4)
	Medium	9 (28.1)	17 (28.3)	19 (31.2)
	High	5 (15.6)	18 (30)	21 (34.4)
	Total	32	60	61
Clinical status	Without comorb.	17 (44.7)	32 (45.1)	21 (27.3)
	With comorb.	21 (55.3)	39 (54.9)	56 (72.7)
	Total	38	71	77

All the SES levels were represented in each cluster ($p = .08$), but some differences emerged for the distribution of SES levels within each cluster⁴. SES levels were not evenly distributed in the first cluster ($\chi^2 = 8.31$, $p = .016$), which consisted primarily of children from low SES families (56.3%), while 28.1% were from medium SES families, and only 5 (15.6%) from high SES families. The three SES levels were more evenly represented in cluster 2 ($\chi^2 = 1.90$, $p = .39$), with 41.7% of children from low SES families, 28.3% from medium SES families, and 30% from high SES families. The same applied to cluster 3 ($\chi^2 = 0.13$, $p = .94$), with equal proportions of children from low and high SES families (34.4% each), and 31.1% from medium SES families.

Similarly, the correspondence between cluster membership and clinical diagnoses was explored. A clinical diagnosis was found for 186 of the 204 cases

⁴ Bonferroni's corrections were applied and the α -value was set at 0.017.

examined. In the others, either the diagnostic process was underway at the time of the data collection or no diagnosis was reported in the clinical records, or - in cases of SLD, for example - the child was too young to be diagnosed, or a session of treatment was needed to confirm the clinical hypotheses.

The sample was divided into two categories on the basis of any available clinical diagnosis: with comorbidities ($n = 116$) or without comorbidities ($n = 70$). Both categories were represented in all three clusters (Table 2), but they were not evenly distributed ($\chi^2 = 6.01, p = .049$). The first cluster contained much the same proportions of participants with and without a comorbidity (55.3% and 44.7% of the participants, respectively; $\chi^2 = .42, p = .52$), and so did the second cluster (54.9 and 45.1% respectively; $\chi^2 = .69, p = .41$). The third cluster, on the other hand, included a higher proportion of children with a comorbidity, i.e. 72.7% versus 27.3% without any known comorbidity ($\chi^2 = 15.91, p < .001$). The most common primary diagnoses associated with BIF in this cluster were SLD and ADHD (accounting for 45% and 27% of the comorbid disorders, respectively).

4. DISCUSSION

The present study aimed at analyzing inter-individual variability in BIF, and the possibility of homogeneous subgroups being detectable within the general population of children with BIF, that might be characterized by different patterns of intellectual abilities. When the four main WISC-IV indices (i.e. Verbal Comprehension, Perceptual Reasoning, Working Memory, and Processing Speed) were considered, cluster analysis revealed three different subgroups, characterized by different WISC-IV profiles. All

three subgroups were characterized by an uneven profile, but with different patterns of strengths and weaknesses.

The first subgroup featured significantly higher scores in Perceptual Reasoning than on the three other indexes, and better scores for Processing Speed than for Verbal Comprehension (in which the sample's performance was the most impaired). Participants' Perceptual Reasoning scores were in the normal range, their Working Memory and Verbal Comprehension scores were both between one and two standard deviations below average (in the borderline range), and their Processing Speed was on the threshold between borderline and normal range.

The second subgroup scored highest on Processing Speed (within normal range), while these children's performance was borderline on the Verbal Comprehension and Perceptual Reasoning indexes, and for Working Memory (the index with the lowest scores).

The third subgroup grouped children with an average performance on Verbal Comprehension and Perceptual Reasoning, and lower scores for Working Memory and Processing Speed, with the latter both in the borderline range.

So, Working Memory scores were below average in all three subgroups (i.e. less than 85), and in the borderline range, while the other indexes varied from borderline to normal across the three groups. This result would confirm the working memory impairment identified in individuals with BIF (e.g. Alloway, 2010; Maehler & Schuchardt, 2009), regardless of their different phenotypic manifestations. However, some differences in the three subgroups' working memory did come to light, with subgroup 1 scoring significantly higher than subgroup 3.

The three clusters did not differ in terms of full-scale IQ. This suggests that differences between individuals with BIF are not only quantitative, or merely an issue

of general intellectual impairments, supporting the findings of a previous study (Study 1 in this dissertation), from which differences in the cognitive profiles of children with BIF did not depend on their general IQ.

The three profiles identified here are conceivably associated with other shared characteristics, or shared origins. The first cluster might particularly reflect the effects of the environment. Several studies have suggested that environmental factors have broader influences on some abilities or neurocognitive systems than others, with a stronger effect on the crystallized than on the fluid component of intelligence (e.g. Duyme et al., 1999; Eilertsen et al., 2016). Children in the first cluster scored lower on the Verbal Comprehension index, which is more representative of crystallized intelligence.

Although children from low SES families were represented in all three clusters, statistical analyses showed that the first cluster contained a higher proportion of these children, and only a few from high SES families. The other two clusters were more balanced in terms of the proportions of the three SES levels. It is important to bear in mind, however, that the family's SES was only available for 153 of the 204 children. Our result partially supports the impression that family SES influences the intellectual profile of children with BIF, and performing such analyses on a larger sample of participants could help to clarify this issue.

The third cluster had a similar profile to the one found in previous studies for children with SLD and ADHD (e.g. Cornoldi et al., 2014; Fenollar-Cortés et al., 2015), who had similar verbal comprehension and perceptual reasoning scores, which were higher than their scores for working memory and processing speed (and the latter two did not differ from each other).

When participants in the present study were grouped by presence or absence of a clinical diagnosis associated with their BIF, the third cluster contained more children with than without comorbidities, while the proportions of children with and without comorbidities were evenly distributed in the other two clusters. The group with comorbidities included children with several different associated conditions: considering the primary diagnoses coded according to the ICD-10, 65 had SLD, 29 had ADHD or conduct disorders, 9 had specific developmental disorders of speech and language, 8 had emotional disorders, 1 was diagnosed with autistic disorder, 1 had an unspecified childhood disorder of social functioning, 1 was diagnosed with specific developmental disorder of motor function, 1 had spastic hemiplegic cerebral palsy, and 1 had Sotos syndrome. The vast majority of the primary clinical diagnoses were therefore SLD and ADHD (about 81% of the comorbid diagnoses).

Children in the second cluster had more generalized impairments, with the exception of Processing Speed, i.e. their Verbal Comprehension, Perceptual Reasoning, and Working Memory scores all fell in the borderline range.

Unlike the report from Jankowska (2011; cited in Jankowska, 2016), no flat profile was found in the present study, and this could be due to differences between the WISC-R and WISC-IV scales.

When the two additional indexes (the GAI and CPI) were considered, however, cluster 2 was the only one of the three clusters emerged in the present study with no discrepancies, and with a profile similar to the one found by Bremner et al. (2011) in their subgroup of individuals with BIF. Without other data available, we can only hypothesize that this cluster might group the children defined by Vianello et al. (2014) as cases of “pure” BIF, without a clear and well-defined etiology. Future studies could address this issue.

Finally, it is important to bear in mind that BIF may be associated with a broad array of clinical and neurodevelopmental conditions that may affect the individual's cognitive and behavioral profile, and that were not represented in the sample considered here (e.g. children with autism spectrum disorder, or children with genetic syndromes), so we cannot exclude the possibility of further subgroups.

It would be interesting to see whether the three clusters of intellectual profiles were also associated with specific behavioral profiles and cognitive characteristics not assessed by the WISC-IV (e.g. planning or problem-solving abilities, or academic achievement). Future studies should address this issue in order to better analyze the phenotypic manifestations of BIF.

Due to the analyses adopted, the present study has an exploratory value, but the findings could help to promote a greater clinical awareness of the complexities of BIF.

CHAPTER 4

STUDY 3:

BORDERLINE INTELLECTUAL FUNCTIONING AND ACADEMIC ACHIEVEMENT

1. INTRODUCTION

One area in which an impaired functioning is particularly important in childhood is academic performance (Colomer, Re, Miranda, & Lucangeli, 2013). The literature suggests that school-age children with borderline intellectual functioning (BIF) are characterized by difficulties at school and delays in reaching their academic milestones (e.g. Salvador-Carulla et al., 2013). Although this condition should be present earlier on, BIF is often only recognized when a child starts school. Children with BIF may have problems with academic demands or fail to acquire the basic academic skills, such as reading, writing, and math, typical of their age and education (e.g. Jankowska, 2016; MacMillan et al., 1998). The diagnostic delay is because there are often no phenotypic characteristics associated with BIF, and the individuals affected may not reveal serious or marked delays in their early years (Karande et al., 2008; Salvador-Carulla et al., 2013).

Based on the normal distribution of intelligence, about 14% of the population have an intellectual quotient in the borderline range, so around three children with BIF might be expected in an average classroom of 20 to 25 children. Cooter and Cooter (2004) reported that, in a typical US schoolroom of 25 pupils, three or four pupils might be expected to have BIF, with higher numbers in areas of poverty and in low-income urban areas. A different prevalence is reported by the Ministerial Directive on special educational need that indicated that the pupils with BIF should account for 2.5% of the Italian student population (MIUR, 2012). There are several reasons why this estimation was probably too “optimistic”, and only considered children who had already been attested with BIF, or completed a clinical assessment (Vianello et al., 2014).

Be that as it may, cases of BIF seem to account for a large proportion of the children who have academic problems (MacMillan et al., 1998). Karande et al. (2008) found that 20.1% of the children referred for clinical assessment due to poor school performance had BIF. MacMillan et al. (1998) found an even higher prevalence: when they considered children at risk of having special education placement, who were referred for learning, behavioral, or other problems by their regular class teachers, 48% had an intellectual quotient in the borderline range.

Students with BIF are described as having generalized difficulties in almost all areas of learning, unlike children with specific learning disorders (SLD) (Ninivaggi, 2009). Ninivaggi (2009) described some features of BIF that may emerge during clinical assessments, such as a tendency to concrete-mindedness, difficulties in combining new knowledge with previously-learned information, and in generalizing learned knowledge and skills to new problems or situations, low frustration tolerance, irritability, low self-esteem, and low motivation (also due to perceived inadequacies), and poor common sense, to cite just a few.

Several authors have reported academic and learning difficulties in this population, but only a few studies have analyzed the learning profiles of school-age children with BIF (Zuccarello et al., 2015), and such studies often did not focus directly on BIF. The present study aimed at analyzing learning achievement in school-age Italian children with BIF to better understand the nature of this population's difficulties at school.

In the previously-mentioned study by Karande et al. (2008), 89% of their sample of children with BIF were described by the school authorities as being poor in all academic subjects. When different academic abilities were considered, 92.7% had writing difficulties, 76.4% had problems with mathematics, and 25.5% reported difficulties in reading. In another of the above-mentioned studies, MacMillan et al. (1998) also found that children with BIF performed less well than their peers with typical intellectual functioning in all the learning skills examined (i.e. reading, arithmetic, and spelling). Similarly, Claypool et al. (2008) found that students with borderline (IQ 70-79) and low average intelligence (IQ 80-89) had a worse performance than their peers with average intelligence in both reading and mathematics. Zuccarello et al. (2015) reported that the performance of their subgroup of children with BIF in reading tasks (i.e. text reading, word and pseudo-word reading, and text comprehension) was worse than could be expected on the basis of norms (i.e. using z-scores and an expected mean of 0). Kortteinen et al. (2009) compared adolescents with BIF (with and without reading disability) and adolescents of average IQ (with and without reading disability) on a series of cognitive and academic measures. They found no significant interaction between reading disability and intellectual functioning, concluding that the relationship between IQ and the cognitive and learning skills examined in the study remained much the same, irrespective of any reading disability. Their results showed a

significant effect of IQ in reading comprehension, and arithmetic, since participants with borderline IQ performed less well than those of average intelligence in these tasks, whereas their performance in a text reading task was similar. Participants with BIF and reading disability performed less well than their peers of average IQ with reading disability in the spelling of non-words, but not in the spelling of words (Kortteinen et al., 2009).

The relationship between intelligence and academic achievement is recognized in the literature, and intelligence has been considered hugely important to children's academic success (e.g. Deary, Strand, Smith, & Fernandes, 2007). So children with BIF presumably struggle to learn because of their weak cognitive skills, with negative outcomes for their academic achievement (e.g. Jankowska, 2016). This assumption contrasts, however, with the fact that some children show a clear discrepancy between their intellectual functioning and their learning achievements (e.g. Mäehler & Schuchardt, 2016). A good example comes from children with SLD, who show impairments in one or more learning skills that cannot be explained by an impaired intelligence. SLD are among the most common developmental disorders diagnosed in school-age children (Moll, Kunze, Neuhoff, Bruder, & Schulte-Körne, 2014). A criterion conventionally used to diagnose SLD was a discrepancy between an average intelligence and a poor academic performance (APA, 2000). This criterion is no longer mentioned in the DSM-5 (APA, 2013).

According to the CONFIL Spanish consensus group, one of the most frequent neuropsychological disorders associated with BIF is SLD (Salvador-Carulla et al., 2013). It is noteworthy that the two conditions (BIF and SLD) share similar manifestations, specifically regarding academic performance. Moreover, some studies found similar working memory and short-term memory functioning in children with

BIF and children with SLD (e.g. Bonifacci & Snowling, 2008; Mäehler & Schuchardt, 2009). Bonifacci and Snowling (2008), for example, found no differences between children with BIF and children with dyslexia in the digit span task. Similarly, Mäehler and Schuchardt (2009) compared children with mild-to-borderline intellectual disabilities and learning difficulties, and children with mixed disorder of scholastic skills and average IQ, finding no differences in their working memory functioning.

With the exclusion of the discrepancy criterion, there could be a higher risk of confusing these two conditions, and failing to treat them as a comorbidity, i.e. SLD could be diagnosed in children with BIF whose difficulties might be due to their weaker cognitive abilities, and this could affect the adequacy of their treatment and support measures. It is questionable whether the learning difficulties of children with BIF are qualitatively and quantitatively similar to those of children with learning difficulties or disorders and a normal intellectual functioning or there may be some differences.

In the above-mentioned study by Bonifacci and Snowling (2008), children with BIF and children with dyslexia (and a normal IQ) revealed similar reading skills, with both groups performing significantly less well than a control group of typically-developing peers.

Going beyond the comparison between BIF and SLD, it is worth noting that an effect of IQ seems more likely for some learning skills, such as mathematic and reading comprehension, than for others (e.g. Kortteinen et al., 2009). For example, reading comprehension involves higher-level abilities and reasoning than reading alone (in terms of decoding). Individuals have to connect single parts of a text to construct a meaningful message (e.g. Kintsch & Rawson, 2005). Together with the involvement of working memory, the ability to infer information not directly provided in the passage,

and an understanding of grammar rules are just some of the abilities needed to understand a text (e.g. van Wingerden, Segers, van Balkom, & Verhoeven, 2014).

The aim of the present study was to analyze learning skills in children with BIF, and particularly to investigate the commonalities and differences between children with this condition and those with SLD. It was assumed here that pupils with BIF have basic learning skills similar to those of children with SLD, but the former will presumably have greater difficulty with higher-level academic skills. In particular, children with BIF will conceivably have more difficulty with reading comprehension than children with SLD. Similarly, children with BIF can be expected to be less capable in mathematics than children with SLD, specifically when all subtypes of SLD are considered as a group.

2. METHOD

Archival data on children assessed between 2013 and 2016 at several Italian mental health services for children and adolescents were screened to identify children attending Italian primary and lower secondary schools with BIF and children with learning difficulties and disorders and a normal intellectual functioning.

For the BIF group, the inclusion criterion was a full-scale IQ (FSIQ) between 1 and 2 standard deviations below average based on the administration of the 10 core subtests of the WISC-IV (Wechsler, 2003). A control group consisted of children with learning difficulties or disorders and a normal intellectual functioning ($IQ > 85$) (the LD

group), matched for chronological age with the BIF group. This group was selected on the basis of a clinical judgment and diagnosis, and a FSIQ higher than 85.

All participants had been assessed for clinical or diagnostic purposes.

2.1 Participants

The BIF group consisted of 133 school-age children with a mean age of 10 years and 1 month ($SD = 25$ months, age range = 6;4 – 15;5 years), who obtained a FSIQ between 70 and 85 on using the WISC-IV (mean FSIQ = 79.09, $SD = 4.35$). The group included 92 males and 41 females. Some of the participants considered in the present study were also involved in the studies discussed in the previous chapters of this dissertation.

The LD group included 73 school-age children with a mean age of 9 years and 11 months ($SD = 24$ months, age range = 6;2 – 13;10 years), 47 males and 26 females, assessed for diagnostic purposes because they had learning difficulties. The children in the LD group were attending Italian mainstream primary and lower secondary schools, and had a mean FSIQ of 105.11 ($SD = 10.71$; IQ range = 87 – 136). The inclusion criteria for the control group were: the presence of marked learning difficulties or a clinical diagnosis of SLD in accordance with the ICD-10 code (F81.0, F81.1., F81.2, F81.8, F81.3)⁵; and a full assessment of intellectual functioning based on the administration of the WISC-IV (to ensure that no variables linked with the use of any specific intelligence scale could influence the total IQ score).

⁵ We considered not only participants with a clinical diagnosis, but also those with a stronger clinical suspicion of SLD ($n = 4$), but not diagnosed as such due to their young age. This was needed to obtain groups with a similar chronological age range. However, the vast majority of the children met the requirements for a diagnosis in the F81 code. When the analyses reported in the Results section were run after excluding the 4 cases of ‘suspected’ SLD, there were no differences in the outcomes.

2.2 Measures

Several tasks are used by Italian clinicians to assess learning abilities in school-age children. The tasks to consider were decided a priori, before starting any data collection, bearing in mind the tools most commonly used to assess learning skills in Italian children.

A text reading task and a text comprehension task were chosen as measures of speed, accuracy, and reading comprehension. To assess mathematical abilities, tasks of written operations, mental operations, and retrieval of arithmetic facts were considered. In order to avoid the source of measurement error linked with the use of different tasks, it was decided to consider the same task for all participants.

2.2.1 Reading abilities

Reading speed and accuracy

Reading speed and accuracy were assessed with the narrative passage reading test of the MT batteries (Cornoldi & Colpo, 1995, 2011), which are the most psychometrically valid Italian tools for examining passage reading speed and accuracy (Tressoldi, Stella, & Faggella, 2001). Different passages are available for each school grade, with the number of syllables per passage increasing for higher school grades. The children are asked to read the passage coinciding with their school grade. The instructions emphasize the need for both accuracy and speed (“Read aloud as accurately and rapidly as you can”). The task enables reading speed to be calculated in terms of syllables per second (the number of syllables read divided by the time in seconds taken to read them), and accuracy (the number of mistakes made while reading).

In accordance with standardization data, raw scores can be converted into z-scores and percentiles, or into performance bands (i.e. optimal performance, adequate performance, needs monitoring, needs immediate action). For the purposes of our analysis, all scores were converted into z-scores. Reading accuracy was calculated by considering the number of words misread, so a higher z-score corresponded to a worse performance. Reading speed was calculated in terms of syllables read per second, so in this case a lower z-score coincided with a worse performance.

Reading comprehension

Tasks from the MT batteries (Cornoldi & Colpo, 1995, 1998) were used as measures of reading comprehension. The children are asked to read a text silently and then answer some multiple-choice questions related to the passage. They are given an unlimited amount of time to complete the task, and they are encouraged to consult the text as often as they wished while answering. The texts and the number of questions varied with school grade. The total score is the number of correct answers. Raw scores could be interpreted in terms of performance bands (i.e. immediate intervention required, monitoring required, adequate, and optimal). For statistical purposes, the raw scores were converted into z-scores according to standard reference data (Cornoldi & Colpo, 1995, 1998); the higher the z-score, the better the performance.

2.2.2 Arithmetical academic achievement

Tasks involving written operations, mental calculation, and arithmetical fact retrieval were considered as measures of mathematical skills. All the tasks came from the AC-MT 6-11 (Cornoldi, Lucangeli, & Bellina, 2002; 2012), and AC-MT 11-14

(Cornoldi & Cazzola, 2004), two Italian batteries for assessing mathematical abilities in pupils attending primary and lower secondary schools (grades 1 to 8).

Written operations

This task aims to examine children's ability to apply the procedures needed to complete written computational operations and the automatism involved. The children are shown different written multi-digit calculations. The number and type of the calculations changed for different school grades: children in first and second grades are asked to solve four operations (additions and subtractions); those in grades 3 to 8 are asked to solve eight operations (additions, subtractions, multiplications, and divisions). The parameter considered was the number of correct answers. *Z*-scores were calculated on the basis of the normative sample by school grade; the higher the *z*-score, the better the performance.

Mental calculation

Children are asked to do some calculations in their heads. The items changed with school grade: children attending grades 1 to 5 are asked to complete 6 operations (3 additions and 3 subtractions); those in grades 6 to 8 are presented with 4 operations (an addition, a subtraction, a multiplication, and a division). Two measures are obtained from this task, i.e. time (total time taken to answer), and number of wrong answers for the children in grades 1 to 5, or number of right answers for those in grades 6 to 8. For each operation, the time is measured from the moment the operator finishes saying the numbers involved in the operation aloud to the moment when the child answers. The time limit for each calculation is 30 seconds for the children in grades 1 to 5, and 60 seconds for those in grades 6 to 8.

Z-scores were calculated on the basis of the normative sample by school grade. To obtain a single measure of accuracy, the z-scores for the children in grades 6 to 8 were inverted. For the time parameter, higher z-scores coincided with a slower, and consequently weaker performance. Similarly, higher z-scores for the accuracy parameter coincided with more wrong answers, and therefore with less accuracy.

Arithmetical facts retrieval

This task is used to investigate the children's knowledge of number facts, and their ability to access them. The children are asked to complete several verbally-presented arithmetical operations and given 5 seconds to answer for each operation. The items changed with the school grades: the children in grades 1 and 2 are presented with 6 operations (additions and subtractions); for those in grades 3 to 5 the task includes 12 operations (additions, subtractions, and multiplications); and for children in grades 6 to 8 there are 24 operations (additions, subtractions, multiplications, and divisions). Examples of arithmetical facts are simple operations such as 7×7 , $8 + 2$, $10 - 5$, $83 + 17$, or $54:9$.

Here again, the number of wrong answers are considered for the children in grades 1 to 5, and the number of right answers for the older children. Z-scores were calculated on the basis of the normative sample by school grade. As for the mental operations task, the z-scores of the children in grades 6 to 8 were inverted. Higher z-scores coincided with a worse performance.

2.3 Data analysis plan

For statistical purposes, the raw scores for all the measures were converted into *z* scores to compare children of different ages. The SPSS-23 software was used for the statistical analyses.

Multivariate analyses of variance (MANOVA) were conducted on the data. Reading and math abilities were treated separately. Effect sizes were also calculated using Cohen's *d* to analyze the effect sizes of the differences between the groups' learning abilities. Effect sizes of 0.2–0.3 were considered “small”, those around 0.5 “medium”, and those of 0.8 and higher were interpreted as “large” (Cohen, 1988).

When the clinical diagnoses were considered, it was found that a subgroup of children in the BIF group (*n* = 48) had also been diagnosed with SLD. To see if the BIF-related learning difficulties were associated with the condition per se, or specific to a subsample with associated SLD, it was started by considering the BIF group as a single group, then subsequently considered the children with a borderline IQ and an associated diagnosis of SLD separately (the BIF/SLD group).

3. RESULTS

3.1 Comparison between BIF and LD

Descriptive statistics of the learning measures in terms of mean and standard deviations for each group are presented in Table 1.

A series of one-sample *t*-tests was run (with 0 as the expected mean). The results showed that both groups had a significantly worse performance than expected for typically developing children (based on normative data; i.e. by considering *z*-scores,

with 0 as the expected mean, and standard deviation = 1) in all the measures analyzed (see table 1).

Table 1

Mean performance (and standard deviations) in the reading and math tasks for the BIF and LD groups.

	BIF group (n = 133) M (SD)	LD group (n = 73) M (SD)
Reading accuracy (errors)	0.686 (1.365)***	0.614 (1.724)**
Reading speed (syllables/sec.)	-1.227 (0.787)***	-1.209 (0.915)***
Reading comprehension (right answers)	-1.255 (0.974)***	-0.274 (0.953)*
Written operations (right answers)	-1.757 (1.359)***	-0.916 (1.379)***
Mental calculation accuracy (errors)	0.952 (1.40)***	0.469 (1.354)**
Mental calculation speed (total time)	1.558 (1.772)***	1.428 (2.338)***
Arithmetical facts (errors)	1.632 (1.418)***	1.139 (1.425)***

Note. * $p < .05$; ** $p \leq .01$; *** $p \leq .001$.

A multivariate analysis of variance (MANOVA) was run to analyze the differences between children with BIF as a whole and children with LD and on-average intelligence, on the reading measures (i.e. accuracy, speed, and comprehension). All comparisons were analyzed considering Bonferroni's correction for multiple comparisons.

At a multivariate level, the results showed a significant effect of group, Wilks' Lambda = .799, $F(3, 202) = 16.89$, $p < .001$, $\eta^2_p = .201$. Several univariate analyses of variance (ANOVA) were run by applying Bonferroni's correction for multiple

comparisons. Subsequent univariate ANOVAs showed that the two groups differed in reading comprehension, $F(1, 204) = 48.57, p_{\text{adjusted}} < .001, \eta^2_p = .192$; the LD group outperforming the BIF group ($MDiff. = .98, p_{\text{adjusted}} < .001$). No significant differences emerged for the other measures (i.e. reading accuracy and speed: $F(1, 204) = 0.109, p_{\text{adjusted}} = 1, \eta^2_p = .001$; $F(1, 204) = 0.02, p_{\text{adjusted}} = 1, \eta^2_p < .001$, respectively).

To see whether IQ influenced the two groups' performance on the reading measures, a MANCOVA was run with IQ as covariate. The results showed no significant effect of group, Wilks' Lambda = .965, $F(3, 201) = 2.44, p = .065, \eta^2_p = .035$.

A further MANOVA was run to analyze the differences between the two groups in the math measures considered. The results showed a significant effect of group, Wilks' Lambda = .904, $F(4, 201) = 5.31, p < .001, \eta^2_p = .096$. Subsequent univariate ANOVAs and post-hoc comparisons showed differences between the two groups only in the written operations task, $F(1, 204) = 17.84, p_{\text{adjusted}} < .001, \eta^2_p = .08$, with the LD group outperforming the BIF group ($MDiff. = .84, p_{\text{adjusted}} < .001$). No significant differences emerged for the other measures (i.e. mental calculation accuracy and speed, and arithmetical fact retrieval: $F(1, 204) = 5.75, p_{\text{adjusted}} = .068, \eta^2_p = .027$; $F(1, 204) = 0.20, p_{\text{adjusted}} = 1, \eta^2_p = .001$; $F(1, 204) = 5.67, p_{\text{adjusted}} = .072, \eta^2_p = .027$, respectively).

To investigate whether IQ influenced the two groups' performance on the arithmetic measures, a MANCOVA was run with IQ as covariate. The results showed no significant effect of group Wilks' Lambda = .971, $F(4, 200) = 1.52, p = .199, \eta^2_p = .029$.

Cohen's (1988) d values were calculated to measure the effect size of the differences between the BIF and LD groups on the measures of learning ability. Among

the reading measures, the effect sizes were large for reading comprehension ($d = 1.02$), and very small for the other two measures, i.e. accuracy and speed ($d = .05$, $d = .02$, respectively). As for the math measures, the effect sizes were medium for written math operations ($d = .61$), and small for the other measures ($d_{\text{mental calc. accuracy}} = .35$, $d_{\text{mental calc. speed}} = .06$, $d_{\text{arithmetical fact}} = .35$, respectively).

3.2 Comparison between children with BIF, children with BIF and SLD, and children with LD.

To analyze whether the learning difficulties of children with BIF were associated with their BIF per se or with the co-presence of SLD, the BIF group was split into two subgroups on the basis of the presence or absence of a clinical diagnosis of SLD.

Table 2 shows the z -scores obtained by the three groups in the learning tasks considered.

Table 2

Mean (standard deviations) scores in learning tasks by group

	BIF/SLD	BIF/no-SLD	LDs
	n = 48	n = 85	n = 73
	M (SD)	M (SD)	M (SD)
Reading accuracy (errors)	0.877 (1.411)	0.578 (1.335)	0.614 (1.724)
Reading speed (syllables/sec.)	-1.405 (0.889)	-1.126 (0.709)	-1.209 (.915)
Reading comprehension (right answers)	-1.407 (1.038)	-1.17 (0.93)	-0.274 (.953)
Written operations (right answers)	-1.607 (1.369)	-1.841 (1.355)	-0.916 (1.379)
Mental calc. accuracy (errors)	1 (1.354)	0.925 (1.433)	0.469 (1.354)
Mental calculation speed (total time)	1.475 (1.788)	1.605 (1.772)	1.428 (2.338)
Arithmetical facts (errors)	1.557 (1.412)	1.674 (1.429)	1.139 (1.425)

Note: BIF/SLD = borderline intellectual functioning with an associated clinical diagnosis of SLD; BIF/no-SLD = borderline intellectual functioning with no associated diagnosis of SLD; LDs = learning difficulties or disorders and normal IQ.

To identify any differences in the learning achievements between the three groups, two multivariate analyses of variance (MANOVAs) were run on the z-scores obtained in the tasks, separately considering the measures of reading (i.e. accuracy, speed, and comprehension), and math (i.e. math operations, accuracy and speed in mental operations, and arithmetical fact retrieval).

A MANOVA was run to test for differences in the three groups' reading accuracy, speed, and comprehension. On a multivariate level, a significant effect of group emerged, Wilks' Lambda = .781, $F(6, 402) = 8.82$, $p < .001$, $\eta_p^2 = .116$. Subsequent univariate analyses of variance were run, applying Bonferroni's correction for multiple comparisons. The results showed a significant effect of group for reading

comprehension, $F(2, 203) = 25.31$, $p_{adjusted} < .001$, $\eta^2_p = .20$. Subsequent post-hoc comparisons showed that the LD group outperformed the other two groups (i.e. BIF no-SLD, and BIF/ SLD) ($MDiff. = 0.9$, $p_{adjusted} < .001$; $MDiff. = 1.13$, $p_{adjusted} < .001$, respectively), while no significant differences were seen between the two BIF groups ($MDiff. = 0.24$, $p_{adjusted} = .527$).

No significant differences emerged for the reading accuracy and speed measures, $F(2, 203) = 0.67$, $p_{adjusted} = 1$, $\eta^2_p = .007$; $F(2, 203) = 1.74$, $p_{adjusted} = .53$, $\eta^2_p = .017$, respectively.

A second MANOVA was run to analyze performance in the math tasks. A significant effect of group emerged, Wilks' Lambda = .896, $F(8, 400) = 2.81$, $p = .005$, $\eta^2_p = .053$. Subsequent univariate ANOVAs, with Bonferroni's correction for multiple comparisons, showed a significant effect of group for the written operations task, $F(2, 203) = 9.37$, $p_{adjusted} < .001$; $\eta^2_p = .084$. Post-hoc comparisons showed that participants in the LD group obtained significantly higher scores than participants in the BIF no-SLD group ($MDiff. = 0.93$, $p_{adjusted} < .001$) or BIF/SLD group ($MDiff. = 0.69$, $p_{adjusted} = .021$), while the two BIF subgroups did not differ significantly ($p_{adjusted} = 1$). No significant differences were found between the three groups for the other math measures; i.e. accuracy, $F(2, 203) = 2.91$, $p_{adjusted} = .23$, $\eta^2_p = .03$, and speed in mental operations, $F(2, 203) = 0.17$, $p_{adjusted} = 1$, $\eta^2_p = .002$, and arithmetical fact retrieval, $F(2, 203) = 2.93$, $p_{adjusted} = .22$, $\eta^2_p = .03$.

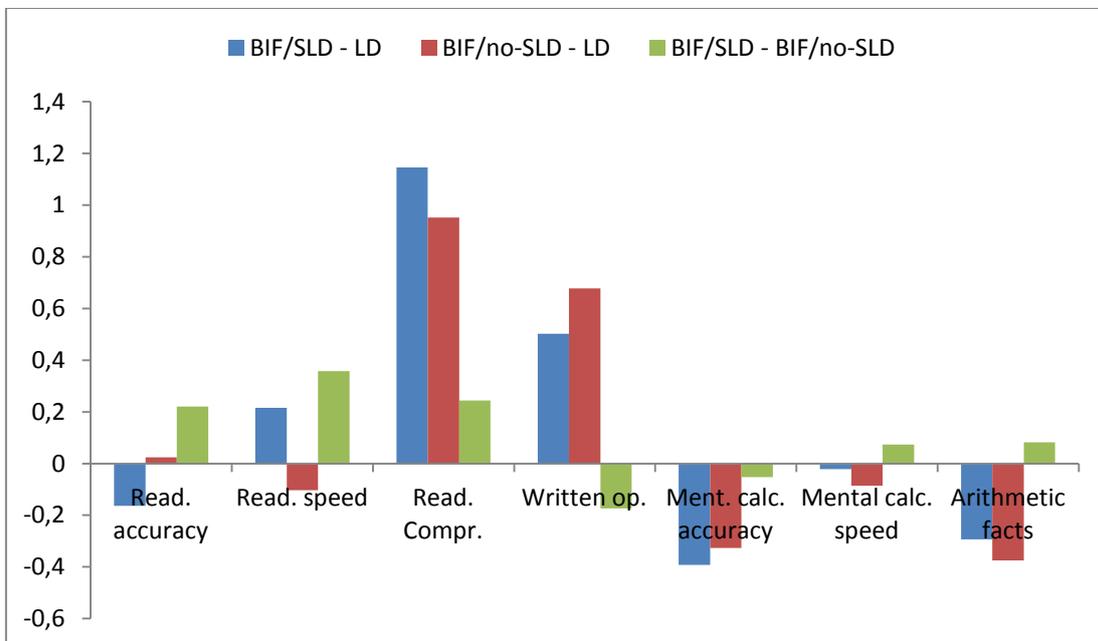
Cohen's (1988) d -values were calculated to analyze the effect sizes of the differences between the three groups for each measure analyzed. In the comparison between the BIF/SLD and BIF no-SLD groups, the effect sizes were small or very small for all tasks ($d_{reading\ acc.} = .22$, $d_{reading\ speed} = .36$, $d_{reading\ compr.} = .24$, $d_{written\ op.} = -.17$, $d_{mental\ calc.\ accuracy} = -.05$, $d_{mental\ calc.\ speed} = .07$, $d_{arithmetical\ facts} = .08$).

The comparison between the BIF/SLD and LD groups yielded a large effect size for reading comprehension ($d = 1.15$), a medium one for the written math operation ($d = .50$), and small and very small effect sizes for the other measures ($d_{\text{reading acc.}} = -.16$, $d_{\text{reading speed}} = .22$, $d_{\text{mental calc. accuracy}} = -.39$, $d_{\text{mental calc. speed}} = -.02$, $d_{\text{arithmetical facts}} = -.29$).

In the comparison between the BIF no-SLD and LD groups, the effect sizes were large for reading comprehension ($d = .95$), medium for the written operations ($d = .68$), and small or very small for reading accuracy and speed ($d = .02$, $d = -.10$, respectively), and for mental calculation and arithmetical fact retrieval ($d_{\text{mental calc. accuracy}} = -.33$, $d_{\text{mental calc. speed}} = -.09$, $d_{\text{arithmetical facts}} = -.38$).

Figure 1 shows the d values obtained from the comparisons.

Figure 1 – Cohen’s d effect sizes of the differences between the measures for the three groups.



Note: BIF/SLD = borderline intellectual functioning with an associated clinical diagnosis of SLD; BIF/no-SLD = borderline intellectual functioning with no associated diagnosis of SLD; LD = learning difficulties or disorders and normal IQ.

4. DISCUSSION

The present study analyzed learning skills in children with BIF, as compared with a sample of peers with learning disorders or difficulties (LD group), and an IQ above 85, seeking similarities and differences between these two conditions. The study was motivated by the acknowledgement that the two conditions often overlap from a clinical point of view, and that SLD is one of the most frequent comorbidities in children with BIF (Salvador-Carulla et al., 2013).

The results revealed both similarities and differences in the academic achievements of children with BIF and those with SLD.

First, children with BIF and children with SLD showed a similar performance in reading speed and accuracy measures, and both groups performed less well than expected on the basis of norms. This result is in line with the report from Bonifacci and Snowling (2008), who found no difference in text reading performance between BIF and SLD (dyslexia).

Significant differences came to light, however, between the two groups' reading comprehension skills, with the BIF group showing a poorer performance. Even though this ability may be impaired in children with SLD (e.g. APA, 2013), and decoding ability plays a part in understanding the meaning of a text (e.g. children with reading disorder need more cognitive resources to decode words in a text), intellectual functioning has a greater influence on comprehension than on reading decoding or reading speed. Reading comprehension is a complex cognitive ability (e.g. Carretti, Caldarola, Tencati, & Cornoldi, 2014) that demands different skills, such as working memory, language skills (e.g. knowledge of vocabulary, and of grammatical structures),

and higher-level text-processing skills (e.g. inference of information, and comprehension monitoring) (e.g. Cain, Oakhill, & Bryant, 2004).

The present findings are in line with those of previous studies on typical development. For example, Kortteinen et al. (2009) found a significant effect of IQ on reading comprehension, but not on text reading, and concluded that IQ has an effect on reading comprehension irrespective of any presence of reading disorder.

In the present study, after controlling for IQ, no significant effect of group emerged on a multivariate level ($p = .065$). Previous studies had identified weaker mathematical skills in children with BIF (e.g. Kortteinen et al., 2009; Claypool et al., 2008) than in children of average intelligence. Only one measure of mathematical skills was used, however, without differentiating between different tasks. In the present study, children with BIF performed less well than might have been expected for their school grade (based on normative data) in all the measures considered. When our BIF and LD groups were compared, the results showed significant differences for written operations, but no significant differences for mental calculations or arithmetical fact retrieval. There are several different components involved in math performance, such as basic knowledge of numbers, working memory, memory for arithmetical facts, knowledge of procedures and the ability to follow them, for a start (e.g. Commodari & Di Blasi, 2014; Cornoldi & Lucangeli, 2004). Different tasks involve different procedure and processes (Cornoldi et al., 2002). It may be that children with BIF have more difficulty than peers with SLD in understanding, using, and automating procedures that enable them to complete adequately written math operations. As for the reading measures, once the effect of IQ had been controlled for, no significant differences emerged between the two groups at a multivariate level ($p = .199$).

A subgroup of participants in the BIF group also had a clinical diagnosis of SLD (i.e. coded as F81.0, F81.1, F81.2, F81.3, F81.8), and their performance was consequently expected to be more impaired than that of their counterpart with no SLD. Instead, our results unexpectedly revealed no significant differences between the two subgroups with BIF, and both performed less well in reading comprehension and written math operations than children with LD and a normal IQ.

These results prompt some considerations on the clinical diagnosis of children with BIF. Without the discrepancy criterion, concomitant SLD may be diagnosed in children with BIF, as various authors have pointed out (e.g. Salvador-Carulla et al., 2013; Vianello et al., 2014). It would be important to differentiate between cases of BIF in which the conditions are comorbid and those in which the child's learning difficulties are due to their impaired intellectual functioning alone, in order to plan adequate support measures.

In short, schoolchildren with BIF have no significant different performance to that of their peers with SLD in academic tasks that demand less reasoning ability, while some differences appear when higher-level skills or the use of procedures are needed.

The present study adds some information about BIF and its manifestation during a child's school years. Some limitations of the study need to be acknowledged, however. First of all, the absence of a typically-developing control group prevented us from better analyzing the strengths and weaknesses of pupils with BIF with regard to learning skills. Using *z*-scores nonetheless enabled us to obtain a measure of their difficulties in each specific learning ability on the basis of normative data.

Second, and most importantly in our opinion, there is the problem relating to the fact that LD were considered as a whole, without distinguishing between the subtypes

of this disorder. So it may be that impairments in each academic skill were masked by the average performance of children not showing a given specific impairment. Having said that, the BIF/SLD group also consisted of children with different subtypes of SLD. It is not easy to consider the different subtypes of SLD separately, and doing so would mean reducing the total number of participants, partly because more than one SLD is often found in the same child, and because some subtypes of SLD (e.g. dyscalculia) are uncommon in isolation.

Future studies could focus on better exploring the relationship between BIF and academic achievement, also considering other cognitive or environmental components that might affect academic performance.

CHAPTER 5

GENERAL CONCLUSION

1. RESEARCH OVERVIEW

The present dissertation aimed at increasing current knowledge of Borderline Intellectual Functioning (BIF) by focusing on analyzing the cognitive profiles and learning skills of school-age children with BIF.

Three general questions were addressed: whether school-age children with BIF have a specific intellectual profile (chapter 2); whether subgroups characterized by distinct, homogeneous cognitive profiles, can be recognized within the broad category of BIF (chapter 3); and how are the academic achievements of children with BIF, particularly when compared with children with learning difficulties and an IQ above 85.

1.1 Intellectual profiles of school-age children with BIF

Apart from their having an IQ between 1 and 2 standard deviations below average, little is known about the intellectual profile of children with BIF.

The WISC-IV (Wechsler, 2003) performance of 204 school-age children who obtained an IQ between 1 and 2 standard deviations below average was analyzed to explore the hypothesis of a specific cognitive profile, characteristic of BIF. The results showed an uneven profile with significant differences between the various indexes, with the sole exception of the Verbal Comprehension and Processing Speed indexes, which

did not differ. The lowest score was obtained for Working Memory, the highest for Perceptual Reasoning. To be specific, the group's performance in Verbal Comprehension, Working Memory, and Processing Speed was between one and two standard deviations below average (i.e. in the borderline range), while for Perceptual Reasoning they scored at the lower end of normal range.

This profile differs, both quantitatively and qualitatively, from the one emerging from previous studies on intellectual disabilities or other neurodevelopmental disorders, such as SLD or ADHD.

A difference was also found when the two additional WISC-IV indexes were considered, with scores on the General Ability Index (GAI) proving significantly higher than those on the Cognitive Proficiency Index (CPI). The profile identified when the children with BIF were considered as a whole seemed to apply regardless of their age (i.e. from 1st to 8th grades), or the severity of their intellectual impairment (i.e. there were no significant differences between children with an IQ of 70-75 and those with an IQ of 76-85). These results support the assumption that BIF has characteristics that make it different from intellectual disability (e.g. Contena & Taddei, 2017).

1.2 Cognitive profiles in BIF: Analysis of subtypes

The third chapter addressed another important issue, since individuals with BIF have been described by many authors as a heterogeneous population (e.g. Salvador-Carulla et al., 2013; Vianello et al., 2014) with no clear cognitive profile and characteristics.

The aim of the study described in chapter 3 of this dissertation was to identify any different subtypes of BIF characterized by homogeneous cognitive profiles, based on the four main factor indexes of the WISC-IV. The results suggested at least three

subgroups can be distinguished, each with their own different relative strengths and weaknesses. One subgroup's abilities were borderline for Verbal Comprehension, Working Memory, and for Processing Speed (which was only slightly above the boundary between borderline and normal range), while they scored higher (within normal range) on the Perceptual Reasoning component. The participants in a second subgroup had scores on Verbal Comprehension, Perceptual Reasoning, and Working Memory in the borderline range, and an average Processing Speed. A third subgroup had a profile characterized by low Working Memory and Processing Speed scores (both in the borderline range), while their Verbal Comprehension and Perceptual Reasoning abilities were relatively well preserved (in the normal range).

Scores on the Working Memory index were impaired in all three subgroups, in line with the children's full-scale IQ (i.e. between 1 and 2 standard deviations below the mean), but differed between the three subgroups, being significantly higher in the first than in the third.

The presence or absence of any clinical comorbidities and family socioeconomic status (SES) were not evenly distributed within each subgroup. The first subgroup consisted primarily of children living in low SES families, with only a small proportion (15.6%) of them from high SES families, whereas the three levels of SES considered (i.e. high, medium, and low) were more equally represented in the second and third subgroups. It may be that the first subgroup's profile is more typical of children from families with a low SES. This would be consistent with studies suggesting more evident effects of SES on some cognitive components rather than others (e.g. Farah et al., 2006), and particularly on crystallized rather than fluid intelligence (e.g. Eilertsen et al., 2016; Rindermann et al., 2010).

Similarly, the prevalence of children with and without comorbid disorders was not evenly distributed within each of the three subgroups. The third subgroup included a significantly larger proportion of children with a comorbid diagnosis than of those with BIF alone, whereas the proportions of cases of BIF with and without comorbidities were similar in the other two subgroups. It is worth adding that most of the comorbid conditions were diagnosed as cases of SLD, followed by and ADHD. The profile of the third subgroup might therefore be more typical of children with BIF who also have another disorder, and seems similar to the profile described in children with SLD or ADHD in previous studies (e.g. Cornoldi et al., 2014; Fenollar-Cortés et al., 2015).

Though only exploratory, the above-described evidence suggests that different cognitive profiles in BIF may be associated with different causative variables or different comorbid disorders. These findings, emerging from a first attempt to clarify the heterogeneity of BIF, should be developed further.

1.3 BIF and academic achievements

Several authors have said that BIF is characterized by learning difficulties, but not many studies have addressed the issue in depth in this specific population.

“Six-hour retarded children”, “Slow learners”, and other expressions coined to describe children with BIF highlight their difficulties at school. One of the more obvious effects of BIF in children and adolescents relates to their academic achievements, and in many cases learning difficulties are the first symptom of BIF to be recognized (e.g. Jankowska, 2016; Salvador-Carulla et al., 2013).

In the study described in chapter 4 of this dissertation, children with BIF were compared with a group of children with learning disabilities and a normal IQ (the LD

group)⁶. The results revealed similarities and differences between the two groups' performance. Children with BIF performed less well than children with LD in reading comprehension tasks, but were comparable in terms of reading accuracy and reading speed.

As for math abilities, the BIF and LD groups showed similar performance in three of the four measures considered (i.e. accuracy and speed in mental calculations, and arithmetical fact retrieval), while significant differences emerged between them in a written operations task, with the LD group outperforming the BIF group.

By comparison with their peers with LD and a normal IQ, the children with BIF thus revealed greater difficulties in tasks demanding more complex cognitive abilities (such as reading comprehension vis-à-vis reading decoding), and in understanding and using procedures to complete written math operations. The differences between the two groups could be attributable to their different IQ levels.

Unexpectedly, the children with BIF who had been diagnosed with SLD did not differ from those who had not: both had a worse performance in reading comprehension and written math operations than children with a normal IQ.

Overall, tasks that involved more complex cognitive abilities, rather than automatisms, seemed to differentiate children with BIF from children with SLD and average intelligence, and this could be explained by the former's impaired intellectual functioning.

⁶ The majority had a clinical diagnosis of SLD. In a few cases, the clinical judgment was "suspended" because of the children's young age.

2. LIMITATIONS OF THE STUDIES IN THE PRESENT DISSERTATION

Some limitations of the studies discussed in this dissertation need to be acknowledged, which regard both the methods used and the participant selection process.

Some of these limitations relate to the object of the studies, BIF. It is difficult to find cases of children identified as having BIF alone, due largely to the paucity of information available on this condition and a consequent lack of clinical attention paid to it. This situation makes it more difficult to conduct research on BIF. When considering school-age children, for example, it is hard to start with a clinical diagnosis if no results of tests are available. BIF is not a separate diagnostic category and the R41.83 code (present in the U.S. version of ICD-10-CM) is rarely used for diagnostic purposes. It is occasionally mentioned in a clinical diagnosis, associated with other primary diagnoses. This problem clearly emerged from the clinical records analyzed for the present research project. In several cases, a child's intellectual functioning was described using vague expressions like "at the lower end of the norm". This could be due to a lack of consensus on BIF, and to the cut-off that clinicians use to describe intellectual functioning as normal (i.e. one or two standard deviations below average). These issues are probably direct consequences of the lack of a diagnostic category for BIF.

Apart from the study limitations associated with the condition per se, there are others that relate to the method used (i.e. data collection from clinical records). Different instruments are used to assess children for diagnostic or screening purposes, depending partly on the reason for their referral, but also on the choice of tools made by

the different clinicians and mental health services. This means that children are not always administered the WISC-IV, or not in its complete version. This issue was particularly relevant for the first two studies (chapters 2 and 3), which analyzed WISC-IV scores to identify the BIF profile and any subtypes.

There are similar issues concerning how learning skills are measured. Specifically, not all learning skills of interest to our research (i.e. reading and math) are always assessed, and the same tools are not always used. Several measures of learning skills are available to Italian professionals. To contain the variability generated by the use of different tests, the present research only considered children measured with certain instruments - the most often used in Italy to assess reading and math skills. This inevitably resulted in a reduction of the sample's size.

The data collection method adopted, and the recruitment process in particular, also prevented us from administering further tasks to the children to make up for missing information. For the same reason, it was also impossible to obtain any standardized measures of adaptive functioning (that were missing in the great majority of medical records), which are important for the purpose of defining BIF, and distinguishing it from typical development and intellectual disability (e.g. Di Nuovo & Buono, 2009b).

Finally, a further limitation of the studies concerns the lack of a control group of typically-developing children. Although using standardized tools enabled us to draw comparisons with the typical population (based on normative data), a control group of typically-developing children matched for chronological age might have helped us to better analyze the profiles of BIF, given the variability characteristic of typically-developing children.

3. FUTURE DIRECTIONS

From the research perspective, the results described in the previous chapters are worth considering when planning future studies. BIF is a relatively unexplored topic, and the lack of consensus among authors and professionals, makes it difficult to interpret the results of studies and implement them in clinical and educational practices.

Based on the studies discussed in this dissertation, it would be useful to explore the evolution of BIF profiles longitudinally, particularly bearing the subtypes in mind. As different profiles may be identified, it would be interesting to examine them from a developmental perspective, to clarify their prognostic implications, and see if changes occur throughout a BIF individual's lifespan. The developmental trajectories and outcomes might not be the same for the different subtypes.

Future studies should also analyze the different profiles in more depth to clarify whether these subtypes are homogeneously characterized by different behavioral features or learning characteristics. The study described in chapter 3 partially investigated whether the subtypes were associated with other variables (clinically diagnosed comorbidities and socio-economic status). Future research could examine this issue more deeply, by considering larger samples of participants.

Paying attention to BIF subtypes would be useful for the purpose of designing adequate support measures to help children develop adequately and improve their outcomes across the lifespan. Further investigating the effects of socio-economic disadvantage seems to be particularly relevant.

The effects of BIF on academic achievement deserve more attention too. The study described in chapter 4 investigated reading and math skills, which are undoubtedly important at school. Their impairment can negatively affect the learning of

other academic subjects too, with a fallout on all academic results. It would be interesting to explore how learning skills relate to cognitive skills other than IQ in more detail, in order to devise scientifically valid intervention to support children with BIF.

4. IMPLICATIONS FOR CLINICAL, EDUCATIONAL AND RESEARCH PRACTICE

The present dissertation addressed some interesting issues, mainly in an attempt to consider BIF as a specific clinical and diagnostic category, distinguishable from mild intellectual disability and from other clinical conditions.

There are obvious educational and clinical implications in the findings of the studies described here, which can be interpreted in the light of the current debate on the clinical definition and diagnosis of BIF. The issue raised by many experts is whether or not BIF needs to be associated with a specific diagnostic category. The studies described in the previous chapters identified specific characteristics in school-age children with BIF that differentiate this condition from other clinical conditions, such as intellectual disability and SLD, and from typical development.

Taken together, our results support the conviction that it would be useful to have a specific clinical category for BIF alone, not only as a possible comorbidity associated with other disorders. A specific diagnostic category should prompt more attention to individuals with BIF, and their strengths and weaknesses, with the aim of improving their prognosis. To date, without its own diagnostic category, BIF has often been diagnosed as part of other conditions on the grounds of the most evident symptoms (e.g. SLD when learning difficulties are the main reason why the child is brought to the

clinician's attention) in order to ensure the child receives extra support at school. This solution seems arbitrary, however, and does not always guarantee that the support will meet the child's specific needs. The findings described in chapter 4 of this dissertation underscore this point, particularly when we look at children with BIF with versus without an associated diagnosis of SLD: both groups were impaired on reading comprehension and written math operations compared with a LD group. This should make us reflect on which clinical diagnosis is most appropriate in children with an IQ in the borderline range: we would need to ascertain whether their difficulties are due to their lower IQ or the consequences of other neurodevelopmental disorders, or whether a comorbidity exists.

In the light of DSM-5, which removed the discrepancy between an average intelligence and poor academic performance as a criterion for diagnosing SLD, the present findings would justify a dual diagnosis (i.e. BIF and SLD in comorbidity), given the differences that emerged in some of the abilities analyzed.

Whether individuals with BIF are considered as a population with particular characteristics, they might need different types of support and treatment from those devised for children with intellectual disability or other clinical disorders, such as SLD. From an educational perspective, support measures for children with SLD or intellectual disability might not be very suitable for children with BIF. For example, reading comprehension training in children with SLD and a normal intelligence may be not effective in children with BIF because of their limited intellectual abilities. Again, the teaching approach and level of simplification of school subjects should be adapted to their level of intellectual functioning.

Accepting a specific cognitive profile for children with BIF may facilitate the diagnostic process and the design of support or treatment plans, focusing on their

strengths and weaknesses. Though individual differences must be borne in mind, it could be used to orient both the diagnosis and the subsequent treatment of BIF. The discrepancy identified in our study between the scores on the GAI and CPI (as seen in children with SLD) points to the need to consider the FSIQ or the four factor indexes, rather than the GAI and CPI, when assessing intellectual functioning in cases of BIF. In the diagnostic process, considering only the discrepancy between the GAI and CPI could lead to BIF going unrecognized, especially when both indexes are in or near the borderline range.

The existence of subtypes of BIF has an important influence on educational practice and should be better investigated. Paying attention to the different subtypes would help clinicians and all professionals working with children with BIF to offer better support to promote the most positive prognosis. The different subtypes are likely to be associated with different developmental trajectories, especially in terms of outcomes in daily abilities.

Finally, the present findings could be useful in research practice. According to Contena and Taddei (2017), and several other published studies, BIF often overlaps with mild intellectual disability, and it has been studied by enrolling individuals with a mild-to-borderline intellectual disability, as a single group. The findings of the present dissertation, and particularly those described in chapter 2, suggest that BIF differs from intellectual disability. Researchers should consider this aspect in the selection of study participants.

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