Longitudinal associations between language difficulties and internalizing problems

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Summary

A growing body of research over the past decades has revealed associations between early language difficulties and later emotional problems. Results show that there is an association, but less is known about the nature of this association: the timing, what types of language difficulties and internalizing problems that are associated, possible gender differences, and the mechanisms behind the association (Yew & O'Kearney, 2013).

In the three papers of the current thesis, we have used different models to investigate different aspects of the longitudinal association between language difficulties and internalizing problems in a population based sample. In Paper I, we developed a four wave cross-lagged model from 18 months to eight years to investigate the bidirectional associations between language difficulties and internalizing problems. In this paper, we also created gender specific dichotomized variables to be able to study this association separately for each gender. In Paper II, we tested a mediation model with four indirect pathways between language difficulties and internalizing problems. In Paper III, we used sibling data to investigate if there is a common underlying factor explaining comorbid language difficulties and internalizing problems at five and eight years and the longitudinal stability of this factor. All papers were based on data from the Norwegian Mother and Child Cohort Study (MoBa). In Paper I we used the full MoBa dataset, in Paper II, a subsample over-selected on children with language difficulties and in Paper III, we used siblings in MoBa.

The results showed that there were bidirectional associations between language difficulties and internalizing problems, especially for girls and especially during the years of transition to school. Further analyses of this vulnerable period showed that language difficulties the last year of preschool were associated with Fist grade internalizing problems. We found support for a model including two indirect pathways from language difficulties to internalizing problems in this period, one through emotion regulation and one through social engagement. Finally, the results showed that the longitudinal association between language difficulties and internalizing problems from five to eight years, was best explained by a common underlying factor shared by siblings.

The results in the current thesis fill a knowledge gap about longitudinal associations between language difficulties and internalizing problems. Much of the

previous longitudinal research have investigated early language difficulties and later internalizing problems in clinical samples. Furthermore, research in this field has been dominated by an underlying assumption that problems in one area affect problems in another area through mechanisms of exclusion or withdrawal from social play. We add to current knowledge by showing longitudinal predictions in both directions, showing associated areas in a mediation analyses, and finally, showing a common underlying factor predicting comorbid language difficulties and internalizing problems.

List of Papers

Paper I

Helland, S. S., Røysamb, E., Wang, M. V., & Gustavson, K. (2017). Language difficulties and internalizing problems: Bidirectional associations from 18 months to 8 years among boys and girls. *Development and Psychopathology*, 1-14. https://doi.org/10.1017/S0954579417001559.

Paper II

Helland, S.S., Røysamb, E., Schjølberg, S., & Gustavson, K. Pathways from preschool language difficulties to school-age internalizing problems. *Under review in Journal of Abnormal Child Psychology.*

Paper III

Helland, S.S., Røysamb, E., Brandlistuen, R.E., Melby-Lervåg, M., & Gustavson, K. A common family factor underlying language difficulties and internalizing problems: Findings from a population-based sibling study. *Under review in Journal of Child Psychology and Psychiatry.*

1. Introduction

Language is an essential part of children's social and emotional development (Morgan, Farkas, Hillemeier, Hammer, & Maczuga, 2015). Children use language to build friendships, to play, to learn how the world works, and enables children to learn about things that are not there (Salmon, O'Kearney, Reese, & Fortune, 2016). Using words, a completely new imaginary universe can be built: "*If you were the donkey and I was a horse – we both lived in this castle*". Language is also a tool for learning to handle emotions for children (Beitchman & Brownlie, 2013). Young children learn early on to use their words when they are experiencing difficult emotions. If a child is angry or frustrated, she may be told to "*use her words*" to explain why she is angry, instead of hitting her best friend (Vallotton & Ayoub, 2011). A sad and withdrawn child may be told to "*put words*" on his feelings or to "*explain what is wrong*", so that he can be helped.

Language skills are not commonly regarded as part of a child's mental health, but language disorder is a formal diagnosis in the Diagnostic and statistical manual for mental disorders (DSM-5) (American Psychiatric Association, 2013). One reason for this may be that language difficulties have historically been the field of pedagogical research where language has been studied as an important ingredient for school success. In a Norwegian context, it is most common for pedagogic professionals to assess and diagnose problems with language and for psychologists to assess and diagnose emotional and behavioural problems. In the current thesis, we aim to show the relevance of language to child mental health. Knowledge about the role of language disorders in mental health is important, given that one of the most essential tools of clinical psychologists and psychiatrists aiming at helping people who experience difficult emotions, is language (Palumbo, Mody, Klykylo, McDougle, & Guenther, 2015).

The relation between language difficulties and social and emotional outcomes has been shown in previous research. Whilst the prevalence of language impairment in childhood varies from 7% to 9% (Norbury & Sonuga-Barke, 2017), there are reports of prevalence of language impairment among young people in prison ranging from 60% to 90% (Hughes, 2012). Similarly, it has been estimated that between 50% and 70% of children having language impairment also experience social, behavioral or emotional problems or vice versa (Benner, Nelson, & Epstein, 2002).

Even though the relation between language difficulties and internalizing problems has been established, we know little about why there is such a link between problems in each area. Furthermore, to entangle these mechanisms, we also need knowledge about what

specific areas of language and social, behavioral and emotional problems are linked. To answer these questions it is necessary with longitudinal data that enables us to investigate the role of developmental age in the association, taking stability and change within each area into account.

1.1. Language difficulties

Is estimated that around 7% to 9% of preschool and school aged children experience language difficulties (Hollung-Møllerhaug, 2010; Norbury et al., 2016; Tomblin et al., 1997). Language difficulties are commonly reported as more prevalent for boys than girls (Zubrick, Taylor, Rice, & Slegers, 2007). Bornstein and colleagues found language development to be stable for both boys and girls in the preschool years from two to six years, with girls consistently scoring higher on multiple language measures (Bornstein, Hahn, & Haynes, 2004).

Studies show a strong familial component of language difficulties (Stromswold, 2001), estimating that 50% to 75% of the variation in language difficulties for school-aged children is explained by variation in genetics (Bishop, 2002). Family characteristics such as parenting style, maternal education or family socioeconomic status may also affect the development of language difficulties (Bornstein, Hahn, & Suwalsky, 2013; Durkin & Conti-Ramsden, 2010). Levels of language difficulties tend to be stable from five years, even for children with additional cognitive, social, emotional, and behavioural problems (Bornstein, Hahn, & Putnick, 2016; Norbury & Sonuga-Barke, 2017).

Definition of language difficulties

Language difficulties concerns problems with producing and understanding language. In the DSM-5 classification (American Psychiatric Association, 2013), language disorder refers to persistent difficulties in the acquisition and use of language across modalities (spoken, written, sign language, or other) due to deficits in comprehension of production that include the following: 1) reduced vocabulary (word knowledge and use); 2) limited sentence structure (ability to put words and word endings together to form sentences based on the rules of grammar and morphology); and 3) impairments in disclosure (ability to use vocabulary and connect sentences to explain or describe a topic or series of events or have a conversation). These difficulties need to be more than expected for their age, resulting in social, academic or other impairments. Furthermore, it is a requirement that the language difficulties are not attributable to hearing impairment or other neurological disabilities that may explain the difficulty.

In clinical practice, language difficulties are often assessed and identified by pedagogic staff or speech language therapists. In these professions, there is not a tradition for relying on medical diagnoses (Bishop, Snowling, Thompson, & Greenhalgh, 2016a). In Norway, assessment of language difficulties may include scores from scales such as *Språk 6-16 screening test* (Ottem & Frost, 2005), discrepancies between verbal and non-verbal intelligence measured by the *Wechsler Intelligence Scale for Children (WISC)* (Wechsler, 2003), and clinical anamneses and observations.

To place our research in the context of previous findings, it is important to have some clarity around the concepts used. Most clinical studies have examined children who have been assessed by a professional and concluded to have Specific language impairments (SLI). Other terminology, based on scale measures has been used interchangeably, such as language difficulties, language disorders, language impairment, and language delay. In a recent consensus study (Bishop et al., 2016a; Bishop, Snowling, Thompson, & Greenhalgh, 2016b), it was agreed to use the term *language disorder* to refer to language difficulties that affect the communication or learning in everyday life, showed by research unlikely to catch up spontaneously. They also agreed to use the term *Developmental language disorder* (DLD), replacing the former SLI, when referring to language disorder not associated with other known biomedical conditions.

In the current thesis, the measures are not based on clinical evaluations, therefore we decided to use the broader term *language difficulties* throughout the thesis, referring to mother report on a scale measuring levels of language difficulties. Additionally, although the measures we used are continuous, they are not appropriate measures of language *skills*. The reason for this is that all items are describing possible difficulties and not positive functions and we assume that skills are more than the absence of problems.

Different types of language difficulties
Language is a multidimensional system consisting of different linguistic domains, such as phonology, syntax, morphology, semantics, pragmatics, vocabulary and grammar across different modalities, such as production and comprehension (Justice et al., 2015). Evidence has been found for an emerging dimensionality, suggesting early unidimensionality of vocabulary and grammar and emerging mulitdimensionality into school age (Tomblin & Zhang, 2006). Some have theorized that vocabulary and grammar may be regarded as lower level language functions, whereas the ability to understand narratives and infer meaning, or in other words semantic and pragmatic language, are higher order aspects of language (Justice et

al., 2015). Justice and colleagues found that in preschool age, grammar, vocabulary and discourse formed one single construct, and by First grade discourse was separated as a single dimension whereas grammar and vocabulary were separated by eight years.

Semantic language refers to using words to refer to mental states or emotions, or the meaning behind the words (Salmon et al., 2016). Some findings have indicated that semantic language difficulties are especially related to internalizing behavior (Van Daal, Verhoeven, & Van Balkom, 2007). In addition, as semantic language may be easier to separate from internalizing behavior, than for example expressive language difficulties (Ottem, 2009), the association between semantic language difficulties and internalizing problems will be a focus in the present study. As language is a skill that develops over time, possible language difficulties will also have different manifestations depending on the age of the child. At the younger ages, we use a unidimensional construct including both expressive and receptive language at 18 months and three years, and at five years and upwards we use semantic language measures in all papers.

1.2.   Internalizing problems

Internalizing problems consist of symptoms of anxiety, depression, and withdrawal (Zahn–Waxler, Klimes–Dougan, & Slattery, 2000). Research on internalizing problems has lagged behind research in other areas of child disorders (Tandon, Cardeli, & Luby, 2009). There may be several reasons for this; one is that internalizing problems are less problematic for their surroundings, such as teachers or parents. Another is that internalizing problems refer to internal states that are more difficult to measure, especially in younger children. There has also been debate as to whether internalizing problems should be viewed as one dimension, or divided into the diagnostic categories of anxiety and depression (Tandon et al., 2009).

In the DSM-5, childhood depression is characterized by symptoms of sadness, irritability, loss of pleasure, concentration difficulties, negative self-evaluation, fatigue, and change in appetite (American Psychiatric Association, 2013). The DSM-5 criteria for childhood depression are the same as for adults, but recent research has suggested that some developmental adjustments should be made, such as a preoccupation with play themes of death in childhood (Luby et al., 2002). Preschool depression is estimated to around 2% (Egger & Angold, 2006; Wichstrøm et al., 2012). There is evidence from a longitudinal study that boys high on depressive symptoms in preschool showed an increase in symptoms into school age and a later decline, whereas girls high on depressive symptoms in preschool showed a stable high level of symptoms from two to 11 years (Sterba, Prinstein, & Cox, 2007).

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Anxiety disorders are defined according to the DSM-5 as excessive fear and anxiety and related behavioural disorders (American Psychiatric Association, 2013). There are eleven different types of anxiety disorders described in the DSM-5, the most common are: separation anxiety, social phobia, generalized anxiety and specific phobia (Whalen, Sylvester, & Luby, 2017). Most studies show a prevalence of preschool anxiety disorders from 10% to 20%, and there is little evidence of gender differences in preschool anxiety disorders (Whalen et al., 2017).

Children with internalizing problems have a stronger family history of internalizing problems than controls and internalizing problems have been found to be transmitted both through genetic and environmental mechanisms (Tandon et al., 2009). Twin studies have shown heritability estimates of 30% to 80% for parent-rated depressive symptoms in children (Rice, Harold, & Thapar, 2002). Risk factors for developing internalizing problems are negative family environments, parenting style, parental mental health, child temperament, problematic peer relationships, and stressful life events (Tandon et al., 2009; Whalen, Sylvester, & Luby, 2017).

1.3.   Previous findings

Associations between language difficulties and internalizing problems have been found in previous studies (Beitchman et al., 2001; Brownlie, Bao, & Beitchman, 2015; Clegg, Law, Rush, Peters, & Roulstone, 2015; Yew & O'Kearney, 2013). There is evidence for increased prevalence of internalizing problems in groups of children with language disorders (Beitchman et al., 1996) and evidence for increased prevalence of language difficulties among children with already identified socioemotional problems (Cohen & Horodezky, 1998). The association has been investigated for different types of language difficulties (Conti-Ramsden & Botting, 2008; Van Daal et al., 2007) and at different ages (Beitchman, Brownlie, & Bao, 2014; Morgan et al., 2015). The association has especially been found for receptive language difficulties (Beitchman et al., 1996; Conti-Ramsden & Botting, 2008) and semantic language difficulties (Van Daal et al., 2007), but there is still a need for investigating these differences in diverse samples (Yew & O'Kearney, 2013). Research on different age groups has indicated that the association may be weaker at younger ages (Henrichs et al., 2013), and then peaking in preschool years, when social and emotional skills are rapidly developing. As children grow older, the association may decrease again, as their language improves and new emotion regulation and compensatory skills develop (Beitchman et al., 2014). There are however few studies investigating all of these ages in one model (Bornstein et al., 2013).

Results on gender differences in the association between language difficulties and internalizing problems are mixed. In an early study, Beitchman and colleagues (1990) found girls to be at greater risk of developing co-occurring problems, whilst Conti-Ramsden and Botting (2008) did not find any gender differences. Some of the associations are however weak and some studies have failed to find the association (Lindsay & Dockrell, 2000; Whitehouse, Robinson, & Zubrick, 2011).

Associations have between language difficulties and internalizing problems have been found in cross-sectional data (Rescorla, Ross, & McClure, 2007; Zubrick et al., 2007). In a meta-analysis of these associations in longitudinal studies, it was found that children with SLI were almost twice as likely to develop internalizing problems than their peers with typical language development (Yew & O'Kearney, 2013). Most studies investigate the association for children with SLI, either in clinical samples (Conti-Ramsden, Mok, Pickles, & Durkin, 2013) or community samples (Beitchman et al., 1996). Some studies have included children from population-based studies and used continuous language measures, showing less strong associations (Clegg et al., 2015; Whitehouse et al., 2011).

One limitation in earlier studies in the area is that they often include early language difficulties and later internalizing problems and do not take into account changes in each construct. It has been showed that children move in an out of categories of language difficulties (Zambrana, Pons, Eadie, & Ystrom, 2014) and internalizing problems (Sterba et al., 2007), and only a few studies have included these developmental changes in their design (Bornstein et al., 2013). Furthermore, it has been noted that the association between language difficulties and internalizing problems is not necessarily direct, and may involve other areas of functioning such as emotion regulation and social skills (Fujiki, Brinton, & Clarke, 2002). Finally, many research suggest that problems in each area may be caused by a common underlying factor, but this has not been the direct focus of study (Yew & O'Kearney, 2013).

1.4.   Theories on developmental associations

Research on the association between language difficulties and internalizing problems has not mainly been driven by an explicit theoretical framework, but rather by clinical observations of the co-occurrence of problems in these areas. One early theory was however proposed by Redmond and Rice (1998), who suggested two conceptual models to explain the relationship between socioemotional behavior and verbal abilities. The social adaption model proposes that the socioemotional behaviors of children with language difficulties are *a result* of social adaptation to their language impairments. These adjustments include less social initiation, less

assertiveness and higher rates of peer rejection. In the social deviance model it is assumed that children with language difficulties are inherently more withdrawn than other children, either due to bidirectional effects or a common underlying factor, with an unknown direction between problems in the two areas. In their original study, Redmond and Rice (1998) found support for the social adaptation model, and much of the research in the area has followed this model.

Developmental cascades

A more recent perspective that is applicable to research in this area is the concept of developmental cascades. The theoretical concept of developmental cascades describes how function in one domain influences function in another domain over time (Masten & Cicchetti, 2010; Masten et al., 2005). Developmental cascades involve the relation between developments in several areas over time. The models commonly include stability in each construct and covariation among constructs to be able to determine longitudinal distinct pathways. If concurrent covariance is not accounted for, it is argued that it is difficult to determine whether there is a unique cascade effect from one area to another, or if this covariance was already there at the first assessment (Masten et al., 2005).

There is an underlying assumption of causal mechanisms in the theoretical description of developmental cascades in that it is assumed that function in one area *influences* function in another over time (Masten & Cicchetti, 2010). It is however also acknowledged that methodological requirements such as controlling for covariates, is necessary to approach an understanding of causality, but there may always be factors not accounted for (Masten & Cicchetti, 2010). It is important to note that the framework of developmental cascades also could be understood as problems in one area *predicting* problems in another area, which does not rely on causal association, but still provides valuable information for intervention purposes. When concurrent correlations and stability of each construct is accounted for, this framework also allows the investigation of how problems in one area predict *change* in problems in the other area.

Mechanisms

In addition to describing how problems in one area predict problems in another, understanding the mechanisms behind the association has been a focus. One aspect of the developmental cascade model is that the association between areas does not need to be direct; there may be indirect pathways.

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It has been suggested in previous literature that the association between language difficulties and internalizing problems is not straightforward (Conti-Ramsden et al., 2013; Fujiki et al., 2002). There are several lines of explanations including indirect pathways in the literature on associations between language difficulties and internalizing problems. It has commonly been suggested that language difficulties may lead a child to be excluded or withdraw from play (Liiva & Cleave, 2005), and result in internalizing problems. Others have suggested that language difficulties lead to problems learning emotion regulation strategies from the caregivers (Fujiki et al., 2002), or have difficulties in the development and use of inner speech (Lidstone, Meins, & Fernyhough, 2012) which again may lead to internalizing problems.

Mediation models are useful for theory development and testing as well as for the identification of possible points of intervention in applied work (Shrout & Bolger, 2002). It has been suggested that researchers commonly focus on finding associations between variables, and less attention is paid to understand the underlying processes (Spector & Brannick, 2011). The mediation models share the same requirements as the developmental cascade models as it is important to control for continuity and stability over time (Masten et al., 2005). A theoretical mediation model was proposed by Salmon and colleagues (2016), in a review on the mechanisms behind the association between language difficulties and emotional and behavioural difficulties. Their model was based on knowledge from longitudinal studies and they suggest that the association is mediated by self-regulation and emotional knowledge. Furthermore, they see parent-child conversations as vital in all of these areas, suggesting that this is an important target for intervention (Salmon et al., 2016).

Comorbidity

When investigating associations between areas of difficulties, we may draw upon knowledge from outside the developmental theories traditionally used. Medical theories of comorbidity encompass all the above perspectives, and provide a framework for further exploration. Comorbidity commonly refers to the presence of more than one distinct condition in an individual (Valderas, Starfield, Sibbald, Salisbury, & Roland, 2009). There has been disagreement among developmental theorists on the extent to which these conditions could be reflected by continuous measures (Hankin et al., 2016), but support has been given to using continuous measures to describe comorbidity, for example within the externalizing spectrum (Krueger & Markon, 2006). Given the present move of emphasis from dichotomous to continuous measures in developmental psychology (Baron-Cohen, 2017), we examined

comorbidity in the present thesis using continuous measures to place our research in the frame of previous studies. A further benefit is that dimensional measures are well suited to capture subclinical problems that may play an essential role in comorbidity. Subclinical problems in one area may trigger the development of problems in another area or they may develop in tandem due to common underlying genetic or environmental influences.

Wilcutt (2014) proposed four different ways to explain comorbidity. Firstly, a causal model suggests that problems in one area directly cause problems in another area, either though causing weakness for developing problems in the other area or directly. Secondly, the correlated liability model suggests that comorbidity is due to share etiological influences. Thirdly, the three independent disorders model suggests that comorbidity is a third disorder with an etiology distinct from each of the disorders alone. Finally, phenocopy or artefactual comorbidity models assume that problems in one area leads to the manifestation of problems in the other area, without the underlying dysfunction.

Common factor

In research on the association between language difficulties and internalizing problems, it has been suggested that this association could be due to a common underlying factor, but this has rarely been the direct focus of study (Irwin, Carter, & Briggs-Gowan, 2002; Yew & O'Kearney, 2013).

In psychopathology, comorbidity is the rule rather than exception, with estimates of 50% having one diagnosis also having another (Kessler et al., 2005). The high comorbidity between different areas of psychopathology has given rise to debate about the accuracy of the constructs (Krueger & Markon, 2006). The concept of a common underlying factor could be understood as the correlated liability model, proposed by Wilcutt (2014).

Recent developments propose that psychopathology can be understood on more than one level, and that there may be a higher-order etiology structure accounting for some of the correlation between the constructs (Caspi et al., 2014; Lahey, Van Hulle, Singh, Waldman, & Rathouz, 2011). Support for a common underlying factor for internalizing problems has been found in previous studies, including in a latent structure analysis of a broad set of DSM diagnoses (Røysamb et al., 2011). These findings have received further support from studies identifying a largely genetic basis of this factor (Kendler et al., 2011). There was some environmental explanation, but this was not shared in the family. Most studies on common underlying factors for psychopathology include adult data, but some have found similar factors for children and adolescents (Lahey et al., 2011; Tackett et al., 2013). Thus, there is

increased knowledge about a general factor for psychopathology, but there is less knowledge about the role of cognitive or neurological aspect of such a factor.

1.5. Literature gaps

Although associations between language difficulties have been found in previous studies, there are still some mixed results and literature gaps that warrant further investigation. In particular:

- Internalizing problems have received far less attention than externalizing problems in the literature (Tandon et al., 2009), also in relation to language difficulties.
- The majority of research uses clinical samples. In clinical groups there may be other clinical conditions that account for some of the association (Brownlie et al., 2015).
- Most studies look at cross-sectional associations. In the longitudinal studies that exist, few studies include baseline adjustment (Yew & O'Kearney, 2013).
- Most studies are on early language difficulties and later emotional problems. There is less knowledge about bidirectional associations over time (Bornstein et al., 2013).
- There is a need for more research on mechanisms behind the association (Conti-Ramsden, 2013).
- Many models are based on an assumption that there is a causal relationship between language difficulties and internalizing problems, and little is known about common underlying factors (Yew & O'Kearney, 2013).

2. Research objectives

The main objective of the current thesis was to investigate the longitudinal associations between language difficulties and internalizing problems, and to more fully understand the underlying mechanisms of these associations.

Paper I

The aim of the first paper was to examine the longitudinal association between language difficulties and internalizing problems to determine a developmental pattern from 18 months to eight years. A second aim was to investigate the role of gender and types of language difficulties in this association.

Paper II

The aim of the second paper was to explore indirect pathways between preschool language difficulties at five years and school-age internalizing problems at six years. We investigated the importance of four theory-derived mediators: empathy, assertiveness, engagement, and emotion regulation.

Paper III

The aim of the third paper was to investigate if the association between language difficulties and internalizing problems could be explained by a common underlying family factor shared by siblings affecting both areas. The stability of this common factor from five to eight years was investigated.

3. Methods

3.1. Participants

3.1.1. The Norwegian Mother and Child Cohort Study

The thesis is based on data from the Norwegian Mother and Child Cohort Study (MoBa; (Magnus et al., 2016; Magnus et al., 2006). MoBa is a prospective population-based pregnancy cohort study conducted by the Norwegian Institute of Public Health. The objective of MoBa is to provide knowledge about the etiology of diseases to enable prevention. The eligible sample consisted of all pregnant women attending routine ultrasound examination during pregnancy weeks 13 to17 in Norway during 1999 to 2008. One restriction in recruitment was the ability to read Norwegian, as the primary questionnaires were in Norwegian only. Questionnaire data were gathered at gestational week 17 from both parents, and from mothers at gestational week 30, and when the child was 6 and 18 months, and 3, 5, and 8 years. The participation rate was 41%, and the cohort now includes 114 479 children, 95 244 mothers and 75 500 fathers.

3.1.2. A MoBa sub-study on language, emotion regulation and social skills

In a MoBa sub-study, we collected new data on emotional, social and cognitive functioning, not available in the full MoBa dataset. The aim of the sub-study was to include in depth measures on emotional and social functioning and to validate existing short-scale developmental measures in MoBa. In the sub-study, 2000 mothers who had returned the questionnaire for their 5-year-old child were invited to participate. There was an over-sampling of children with language difficulties, to ensure sufficient variation in language skills (Hauner, Zinbarg, & Revelle, 2014). We invited 400 mothers of children with probable language impairment and 1600 random controls to fill out a questionnaire. Children with probable language impairment were selected based on mothers' answers on any three language related questions in the 5-year-questionnaire: 1) My child has delayed or deviant language development; 2) Has your child been assessed by a professional for language delay or other difficulties with language/speech or communication? (Only delay in spoken language, good language comprehension or delay in both using spoken language and ability to understand spoken language); and 3) Child scoring -1 standard deviation or lower on mean scale score across six language scales. The final sample consisted of 1176 children whose mothers returned the sub-study questionnaire, equaling 58.8% of the invited mothers, of which 260 had children with probable language difficulties.

3.1.3. The Medical Birth Registry of Norway

The Medical Birth Registry of Norway (MBRN) contains data on all births in Norway since 1967, registered by medical personnel (Irgens, 2000). In the current thesis, we used information from MBRN on serious malformations or syndromes to exclude cases, and information such mother's age at birth and parity, used as control variables.

3.1.4. Samples in the papers

In the ongoing data collection of MoBa, quality assured data files with an updated sample are released at regular intervals. In the current study, we used MoBa data version 8 (Paper I) and version 9 (Paper II and III), see available sample in Table 1. We used all available data in Paper I, and subsamples in Paper II (sub-study) and Paper III (siblings). Thus, some of the missing data at eight years in Paper I compared to five years is due to attrition and some is due to the continuous nature of the data collection process.

Table 1. MoBa samples available at each time point for version 8 and 9

MoBa questionnaires	v8	v9
MBRN	114 275	114 247
Pregnancy	102 265	102 241
18 months	76 432	76 417
3 years	58 844	58 841
5 years	32 841	41 609
6 years (sub-study)		1 176
8 years	19 946	32 105

In all three papers, we excluded children based on reports from the MBRN on reduced hearing, cerebral palsy, suspected syndromes, or other malformations. The reason for this was that all of these developmental problems include a physical element that is likely to affect language development, as motor skills are involved in language production.

In Paper I, we used all available MoBa data on children at 18 months, 3, 5, and 8 years. We excluded 8 366 participants from the original sample based on report from MBRN, resulting in a sample consisting of 18 months (n=76 432), 3 years (n=58 844), 5 years (n=32 841), and 8 years (n=19 946). We then used list-wise deletion in the analyses, resulting in a final sample in each cross-lagged analyses (language to internalizing/internalizing to

language) from 18 months to three years (n=45 384/45 539), from three years to five years (n=20 939/20 993), and from five to eight years (n=7 602/7 565).

In Paper II, the sample consisted of all children participating in the MoBa sub-study (n=1 176). We excluded 158 participants from the original sample based on report from MBRN. We also excluded children who were still in preschool (n=39), children who had started 2nd grade (n=30) and children without information about age at return of questionnaire (n=20) or gender (n=1), resulting in a final sample of 928 children.

In Paper III, we used data from children at five (n= 41 609) and eight (n=32 105) years. We excluded children based on reports from MBRN (n=5 081 at five years), and multiple births (n=1 140 at five years). We only included children whose mothers had returned questionnaires for at least two siblings at five (n=5 568) or eight years (n=3 654). For longitudinal analyses we included only children whose mothers had returned questionnaires for two siblings at five *and* eight years (n=1 208). The reason for the large reduction of sample size from full MoBa sample to sibling sample is that children were excluded from the study if they did not have a MoBa sibling, if the sibling had not turned five or eight years respectively, or if their mother had not returned a questionnaire for one of the siblings. Additionally, the exclusion based on MBRN results in two siblings being excluded if only one fulfilled the criteria.

3.2. Measures

3.2.1. Internalizing problems
Child Behavior Checklist (CBCL) (Papers I, II & III)
The CBCL (Achenbach & Rescorla, 2000) was used to measure internalizing problems at 18 months, three, five, and six years. Due to limited space in the MoBa questionnaires, only selected items from the internalizing dimension were included (see Table 2). Mothers were asked to rate statements about their child's functioning as 1=*Not true*, 2=*Somewhat or sometimes true* or 3=*Very true or often true*. Different items from the internalizing dimension were used in the three papers, based on how we best could answer the aim of the different studies. In Paper I, we investigated change and stability over time, thus using the items that were most repeated at the three time points. We used four items at 18 months (α =.41), four items at three years (α=.53) and eight items at five years (α=.68). In Paper II, we also chose to use the items that were repeated from five to six years, which was the anxiety/depression subscale. At five years, we included seven items (α=.67) and at six years we additionally

Table 2. Available CBCL internalizing items in MoBa

	18 months	3 years	5 years
Anxious/depressed	Clings to adults or too dependent (I)	Clings to adults or too dependent (I)	Clings to adults or too dependent (I, II, III)
	Gets upset when separated from parents (I)	Gets upset when separated from parents (I)	Gets upset when separated from parents (I, II, III)
	Too fearful or anxious (I)	Too fearful or anxious (I)	Too fearful or anxious (I, II, III)
			Feelings are easily hurt (I, II, III)
			Nervous, high-strung, or tense (I, II, III)
			Self-conscious or easily embarrassed (I, II, III)
			Unhappy, sad, or depressed (II, III)
Emotionally reactive	Disturbed by any change in the routine (I)	Disturbed by any change in the routine (I)	Disturbed by any change in the routine (I, III)
		Sudden changes in mood or feelings	
Somatic complaints	Does not eat well	Does not eat well	Does not eat well (III)
		Stomach aces or cramps (without medical cause)	Stomach aces or cramps (without medical cause) (III)
		Vomiting/throwing up (without medical cause)	Vomiting/throwing up (without medical cause) (III)
		Constipated, doesn't move bowls	

Note. Numbers in brackets indicate if items are used in Paper I, II, and III.

included one item (α=.65), as the full scale was available at six years. In Paper III, we only used the CBCL at five years and as repeated items was not an issue, we included all the available internalizing items at five years for greater variance to be explained (α=.65) (see Table A1 in the appendix for description of full scale). Correlations between the internalizing scales with items available in MoBa and the full internalizing CBCL scales were calculated in the sub-study described above. These correlations are presented in Table A2 in the appendix.

Short Mood and Feelings Questionnaire (SMFQ)
Screen for Child Anxiety Related Disorders (SCARED) (Papers I & III)
At eight years, a composite score of SMFQ and SCARED was used to measure internalizing problems. SMFQ is a measure based on the DSM-III-R criteria of depression (Angold & Costello, 1987). A 13-item subscale is used in the MoBa study (α=.79). Mothers are asked to rate how true items are for their child during the last two weeks as 1=*"Not true"*, 2=*"Sometimes true"* or 3=*"True"*. Examples of items are "Felt miserable or unhappy" and "Felt so tired that s/he just sat around and did nothing". SCARED is designed to measure DSM defined anxiety symptoms (Birmaher et al., 1997). A 5-item short scale developed by Birmaher and colleagues (1999) is used in the MoBa study (α=.44). Mothers are asked to rate statements about their child's functioning as "1=*"Not true"*, 2=*"Somewhat or sometimes true"* or 3=*"Very true or often true"*. Examples of statements are "My child gets really frightened for no reason at all" and "My child is afraid to be alone in the house". At eight years, we combined 13 items from the SMFQ and five items from the SCARED into an internalizing scale (α=.75). Mean score of each scale was created and standardized scores were included in the composite score to ensure equal weight of anxiety and depression symptoms.

3.2.2. Language difficulties
20 Questions about language difficulties (Language20Q) (Paper I, II, III)
Language difficulties at five and eight years were measured by Language20Q, developed by Ottem (2009) to identify children with risk of language difficulties. The Language20Q scale includes three subscales: expressive, receptive and semantic language impairment. Expressive impairment involves having problems with being understood by others. Receptive impairment refers to problems with understanding others and storing information. Semantic impairment involves problems with the meaning of words, which may be impaired with regard to both

understanding and producing language. The Language20Q is validated in a Norwegian sample, in a study of 250 children with typical language development and 48 language impaired children against Language6-16, an established Norwegian scale (Ottem, 2009). Mothers are asked to rate statements from 1="*Does not fit the child/absolutely wrong*" to 5 = "*Fits well with the child, absolutely right*". To examine the structural validity in the current sample, we performed a confirmatory factor analysis in Mplus including all 20 items at five years and a three-factor model with one second-order factor showed good fit (RMSEA=.049, CFI=.962 and TLI=.957).

In Paper I, we used all three subscales at five years; semantic (e.g. "Forgets words she/he knows the meaning of", eight items, α =.85), receptive (e.g. "Is quickly getting tired in tasks demanding attention to language", six items, α =.85) and expressive language (e.g. "Is difficult to understand", six items, α =.82). The polychoric correlation between the dichotomized semantic and receptive scales was .81, between the semantic and the expressive scale was.76, and between the receptive and the expressive scale was .76. At eight years we used the semantic subscale only (α =.84). In Paper II, we used the semantic subscale at five years (α =.83) and in Paper II at five (α =.85) and eight years (α =.84).

The Ages and Stages Questionnaire (ASQ) (Paper I)

The ASQ (Bricker et al., 1999) was used to measure language difficulties at 18 months (three items, α=.59) and three years (six items, α=.56) in Paper I. Mothers rated how true statements were for their child from 1= *"Yes"*, 2= *"Sometimes"* to 3= *"Not yet"*. Example of expressive language statement is "Can the child tell you at least two things about a familiar object? If you for example say "Tell me about the ball" can the child answer something like "It is round and I can throw it and it is big?"." Example of receptive language statement is "Without giving your child help by pointing or repeating directions, does your child follow three directions that are unrelated to one another? Give all three directions before the child starts. For example, you may ask your child to "Clap your hands, walk to the door and sit down" or "Give me the pen, open the book and stand up". The ASQ has shown good test-retest reliability, inter-rater reliability and concurrent validity when compared to standardized tests (Janson & Squires, 2004). The Norwegian ASQ has shown good construct validity (Richter & Janson, 2007).

3.2.3. Mediation variables

Social skills (Paper II)

The Social Skills Improvement System (SSIS) Rating Scale (Gresham & Elliott, 2008) was used in Paper II to measure social skills. The scale is developed to screen children suspected for having significant social skills deficits. Three subscales were used in the present study: The Empathy subscale consists of six items (α=.81), e.g. "Tries to understand how you feel" and "Tries to make others feel better". The Assertiveness subscale contains seven items (α=.69), e.g. "Expresses feelings when wronged" and "Asks for help from adults". The Engagement subscale includes seven items (α=.82): "Joins activities that have already started" and "Starts conversations with peers". Mothers were asked to rate how common the items are for their child on a scale from 1= Never to 4 = Almost always. The SSiS is validated based on test content, internal structure, inter-correlations among scales and subscales, item-total correlations, and relations with other variables (Gresham & Elliott, 2008).

Emotion regulation (Paper II)

The Emotion Regulation Checklist (ERC) (Shields & Cicchetti, 1997) was used to measure emotion regulation in Paper II. It consists of the two subscales Liability/Negativity and Emotion Regulation. The latter was used in the present study with six items (α=.71), e.g. "Is a cheerful child" and "Responds positively to neutral or friendly overtures by adults". Mothers were asked to rate these items on a 4-point scale from 1= Never to 4= Almost always. Good interrater reliability, construct validity and discriminant validity has been shown for the ERC (Shields & Cicchetti, 1997). The scale has been used in several studies (Blandon, Calkins, Keane, & O'brien, 2008; Fujiki, Spackman, Brinton, & Hall, 2004). Mother report of regulation has been found to significantly correlate with teacher report (r = .17 to .25) (Blandon et al., 2008).

3.2.4. Covariates

There are other variables that may affect a child's language development, internalizing problems, and potentially the relationship between the two areas. Socioeconomic factors and family environment have commonly been reported to affect both internalizing problems (Ashford, Smit, Van Lier, Cuijpers, & Koot, 2008) and language development (Schjølberg, Eadie, Zachrisson, Øyen, & Prior, 2011). Some studies have also found that the associations between language delay and internalizing problems have disappeared when results are

adjusted for covariates (Whitehouse et al., 2011). In Paper I and II we perform the analyses adjusted for covariates, and in Paper II we only adjusted for gender as adjusting for background variables would remove the variance in the common factor we were studying.

3.3. Statistical analyses

3.3.1. Variables and correlations
In Paper I, we created dichotomized variables for language difficulties and internalizing problems, based on the approximate prevalence in the population. For language difficulties, we included a group of children with 5% highest scores on language difficulties, and for internalizing problems, the highest 10% scores. In Paper II, we logtransformed the two main study variables, and in Paper III, we created cut variables. These cut variables consisted of five categories: 1= *no problems*, 2 = *between score 1 and the mean score*, 3= *between the mean score and one standard deviation above the mean score*, 4 = *above one standard deviation to two standard deviations above the mean score*, and 5= *above two standard deviations above the mean score*. Correlations were estimated in all three papers. Pearson's r was used with continuous measures (Paper II), and polychoric correlations were used when variables were categorized as dichotomous variables in Paper I and with five categories in Paper III.

3.3.2. Logistic regression analyses
In Paper I, we used logistic regression to estimate the association between two dichotomous variables: language difficulties/not language difficulties and internalizing problems/no internalizing problems. In a simple logistic analysis, we estimated to what degree belonging to one category (e.g. language difficulties) is associated with belonging to another category (e.g. internalizing problems), through a report of odds-ratio (OR). In our analyses, we also adjusted for baseline of the outcome variable, thus investigating to what degree having language difficulties at one time point predicted *developing* internalizing problems between two time points. The OR indicates the odds of an outcome given the exposure, compared to the odds of the outcome without the exposure (Szumilas, 2010).

3.3.3. Structural equation modelling

In Paper II and III, we used structural equation modelling (SEM) in Mplus (Muthén, 2010). SEM is useful to determine if there is support for a given theory in a dataset and includes regression, correlation, and factor models in one framework. SEM allows comparison of different competing models, for example models with and without gender differences in estimates. There are several indexes used to determine model fit in SEM analyses. Chi-square (χ^2) indicates how well the model captures the observed covariance and is interpreted in relation to the number of degrees of freedom. The Comparative Fit Index (CFI) and Tucker-Lewis Index (TLI) compares the model fit to the null hypothesis, with a score greater than .90 indicating good fit. The Root-Mean-Square Error of Approximation (RMSEA) measures to what extent the model fits well in the population and a good fit is indicated by a value of .06 or less. SEM allows comparison of the fit of different competing models. Models with fewer unknown parameters are generally considered as standing a better chance of being scientifically replicable and explainable (Bentler & Mooijaart, 1989). Hence, if two models have similar fit, the simplest model will be preferred (Kline, 2011). If the simplest model has significantly poorer fit than the more complex mode, the latter will be preferred. In our papers, we used SEM to test the fit of mediation pathways for each gender in a mediation model (Paper II) and to test the fit of a latent common factor, including longitudinal pathways (Paper III).

In Paper II, we modelled the direct association between language difficulties at five years and internalizing problems at six years, adjusted for internalizing problems at five years. We then tested a mediation model with indirect pathways through empathy, assertiveness, engagement, and emotion regulation. In the mediation model, we estimated the confidence intervals using 5000 bootstrap samples.

In Paper III, we tested a latent factor with equal loadings on language difficulties and internalizing problems for two siblings. Secondly, we tested this model longitudinally, and compared the fit of different models including versus not including direct pathways between the observed variables.

3.3.4. Sibling analyses

Siblings share family environment and 50% of their genes and correlations between siblings provide valuable knowledge on common factors for the family. In sibling-designs, information about siblings is often used to exclude environmental and genetic factors shared

in the family, to minimize the effect of confounders and approach knowledge of causal mechanisms (D'Onofrio, Lahey, Turkheimer, & Lichtenstein, 2013). For example, in sibling control studies, it is possible to investigate pregnancy exposure by comparing associations between siblings exposed and not exposed for example to smoking during pregnancy, thus adjusting for the effect of shared genes and environment (Gustavson et al., 2017).

In Paper III, we developed a model where we, instead of excluding information on shared genes and environment, included this information in the model to investigate a common family factor for comorbid language and internalizing difficulties. We developed a latent factor with equal loadings from language difficulties and internalizing problems for siblings, and investigated if this model had better fit when including direct associations from one difficulty to the other.

4. Main findings

Paper I

In Paper I, we investigated bidirectional longitudinal associations between language difficulties and internalizing problems. The main results were from logistic regression analyses of a four-wave cross-lagged model adjusted for covariates. Due to multiple analyses in the model, only results on a .001 level were highlighted. The results showed that language difficulties at 18 months and three years ages predicted change in internalizing problems for girls. In the opposite direction, internalizing problems at three years predicted change in language difficulties for both boys and girls, whilst internalizing problems at five years predicted change in language difficulties only for girls. Additional analyses revealed that expressive language difficulties at five years predicted change in internalizing problems from five to eight years for girls.

Paper II

In Paper II, we investigated a mediation model of the association between semantic language difficulties at five years and change in internalizing problems from five to six years. The initial direct effect from language to internalizing problems was reduced, as expected, when baseline internalizing problems were controlled for, but the association was still significant. When entering mediators into the model, the association between language and internalizing problems almost disappeared, which indicated a partial mediation. The indirect association was accounted for by two significant mediators: emotion regulation and social engagement. Finally, we tested gender effects by allowing paths to be different for each gender, one path at the time. These results showed that the best fitting model included separate pathways for boys and girls for the concurrent association at five years, and no other gender differences.

Paper III

In Paper III, we used sibling data to investigate a common underlying factor for language difficulties and internalizing problems. We modelled a latent factor loading equally on language difficulties and internalizing problems for both siblings. When the common factor was included in the model, the correlation between language difficulties and internalizing problems within each sibling was reduced to a minimum. Longitudinal analyses of the model from five to eight years showed that the common factor also accounted for the majority of the stability in comorbidity. The best fitting longitudinal mode included no pathways from one

difficulty to the other within each sibling, only individual stability pathways for language difficulties and internalizing problem within each sibling.

5. Discussion

In the current thesis, we investigated the longitudinal association between language difficulties and internalizing problems. We found that problems in each area predicted each other longitudinally, especially during the years of starting school, from five to eight years. We then investigated this period of transmission to school more in depth, by looking at different pathways from preschool language difficulties to school age internalizing problems. Here we found support for two of the four mediation pathways proposed in a theoretical model. Finally, we turned to the question of whether there is a common underlying factor explaining both language difficulties and internalizing problems. Here we found that a common family factor shared by siblings explained the majority of the variation in comorbid difficulties at five and eight years, and that this factor was highly stabile over time.

5.1. Methodological considerations

A main strength of the current thesis is the use of a longitudinal population-based data set in an area of research mainly dominated by clinical studies. A further strength is the diverse use of the MoBa data material in all three articles: 1) Full use of the large longitudinal data set including all participants at all available ages, 2) the use of the full population-based data set to select out possible cases, and 3) the use of MoBa to select siblings. There were however some methodological challenges we must consider before turning to interpreting the findings.

5.1.1. Measurement reliability and validity

The reliability and validty of the results rely on the reliability and validity of the measures. Reliability refers to the consistency or precision of a measure, and may be defined as the degree to which a measurment is a showing consistent results across time and informants. The most commonly used measurment of reliability or internal consistency is Cronbach's alpha, as it is easier to use compared to other estimates, such as test-retest estimates (Tavakol & Dennick, 2011). Cronbach's alpha is an indicator of the extent to which all the items in a test measure the same construct and concerns the inter-relatedness of the items in the test. If the items are correlated to each other, the alpha increases, but the alpha also increases with many items in a test (Tavakol & Dennick, 2011). Some of the aphas in the current study are low, especially for internalizing problems measured by CBCL at 18 months (α=.41), three years (α=.53) and five years (α=.68). One explanation for this is that the scales we used included few items. Cronbach's alpha is based on Pearson's correlations, which holds an assumption of

normally distributed continuous variables. The CBCL items are on an ordinal scale with few response categories and a skewed distribution, and a calculation of alpha based on Pearson's correlation may lead to underestimation of true association. An alternative is to caclculate the reliability using polychoric ordinal alphas (Gadermann, Guhn, & Zumbo, 2012). This gives $\alpha=.68$ at 18 months, $\alpha=.78$ at three years, and $\alpha=.86$ at five years.

Validity refers to the degree to which an instrument measures what it is intended to measure. There were different concerns regarding the validity of the internalizing measure and the language measure used in the current study. Internalizing problems are commonly measured with scales that are validated in diverse samples, and CBCL is a well-validated measure (Achenbach, 2001). The challenge in the current study was that only selected items were available in the MoBa dataset. To meet this challenge and validate the items that were available, we included the full CBCL in the MoBa sub-study. Table A2 in the appendix shows correlations between items used in the MoBa and the full scale. We see that there are slightly stronger correlations between the MoBa items as the child grows older, with items in the anxious/depressed subscale at 18 months correlating .78 with the full anxious/depressed scale and .99 at five years.

The measure of language difficulties represented a different challenge. In the literature, it is more common to use clinical evaluations of language difficulties as opposed to continuous measures as used in the current study. Thus, there was a need to investigate if our measures appropriately captured children who indeed were struggling with language. In a validation analyses in Paper I, the dichotomous measures of language difficulties were compared to mother's report of whether the child had been assessed by a professional for a language disorder (see Paper I for details on the validation). We have also performed phone interviews with 100 mothers of children in the MoBa sub study. Analysis of these interviews was beyond the scope of the current thesis, but will in the future provide valuable information about the validity of the questionnaire language measures in MoBa.

A general challenge in the study of child development, is that younger children express themselves in different ways than older children. One way we have met this is to use different measures of language difficulties for the younger ages – 18 months and three years, and more differential measures of language difficulties at five and eight years. For measuring language it is essential that the measures are adequate for the age of the child, as language developes and becomes more advanced for all children (M. LRice, 2013). For internalizing problems, we also used more differentiating measures at eight years, where separate measures of anxiety and depression were available.

5.1.2. Mother report

One challenge in the current thesis was the use of a single informant. It could be argued that more informants would improve the reliability of the results, on the other hand, it is likely that different informants would report on different aspects or that children's behaviors are context dependent. Some researchers have suggested that the concordance of parent and child raters on symptoms of internalizing problems is low to moderate (Rice, Harold, & Thapar, 2002). Modest agreement among informants is a known problem in psychological research. Previous studies have shown that parents have the highest agreement, parents and teacher somewhat lower and children and parents have the lowest agreement (Achenbach, McConaughy, & Howell, 1987). It has also been found lower interrater-agreement for internalizing problems than for externalizing (Martel et al., 2017).

It is possible that estimates of the association between language difficulties and internalizing problems are affected when both are reported by the same informant. It has been suggested that when parents are worried about how their child functions in one area, this may affect the reporting in another area, especially when the development in this area is particularity age dependent, such as language difficulties (Aro, Laakso, Määttä, Tolvanen, & Poikkeus, 2014). In one previous study, it was found that parent report of child behaviour accounted for a small proportion of the variance of a professional assessment of language skills (Chaffee, Cunningham, Secord-Gilbert, Elbard, & Richards, 1991). To further improve the validity of the measures, we could have included other informants, such as father report or teacher report.

5.1.3. Measurement overlap

Another potential problem regards measurement overlap. It has for example been found that shy children score lower on expressive language tests than on receptive tests (Coplan, Wichmann, & Lagacé-Séguin, 2001). In the current theses, there are some of the items in the language scale that may tap internalizing problems, such as "Uses short sentences when answering questions". It is conceivable that a child who is feeling socially anxious may also use shorter sentences. Indeed, additional analyses showed highest correlations between the language items "Uses short sentences when answering questions" ($r= .18$) and "Has difficulties retelling a story s/he has hears" ($r=.19$) with the CBCL anxiety/depression subscale. Whilst the internalizing items that showed the highest correlation with the full semantic language scale were "Feelings are easily hurt" ($r=.17$) and "Nervous, high-strung or

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tense" (r=.14). These analyses showed that no single item was responsible for the association found in the current study, thus ruling out phenocopy as a major explanation of the findings.

5.1.4. Population based and clinical samples

Most studies on longitudinal associations between language difficulties and internalizing problems have investigated children with SLI in clinical samples (Yew & O'Kearney, 2013). Our results need to be interpreted in relation to the use of categorical measures and clinical samples used in previous studies. Recently, there has been a move towards using continuous measures in research on psychopathology (Hankin et al., 2016). It has been argued that due to the high comorbidity between categorically defined disorders, different types of psychopathology may best be described as dimensional models on multiple levels (Hankin et al., 2016). This has also been found for other neurodevelopmental problems such as autism (Baron-Cohen, 2017). In the current thesis we have used both dichotomous and dimensional measures.

A commonly cited literature review has reported an estimated comorbidity between language impairment and emotional and behavioral disorders of 50% to 70% (Benner et al., 2002). One reason for this high estimate is referral bias. Only studies including children with formal diagnosis of either language impairment or emotional or behavioral diagnoses were included in the review. This excludes all who have not been referred for assessment, and it has been showed that children with comorbid difficulties are more often referred which results in an overestimate of comorbidity (Brownlie et al., 2015). A second reason this estimate may be too high is that the review only included studies that had reported both language impairment and emotional or behavioral problems, thus excluding a population with problems in one area and unassessed problems in the other. Results from population based studies, may balance this estimate. In the current study, we found a co-occurrence of language difficulties and internalizing problems of .16 for boys and .30 for girls at five years (Paper II) and the concurrent polychoric correlation for both genders to be .23 at five years and .27 at eight years in Paper III, which may be a more realistic estimate of the association in a general population.

It is important to emphasize that the results in the current thesis are for most children, and that the associations may be different and probably higher in a sample including only clinical cases. Our results do however complement results from previous clinical studies, in that we include a wider variation of problems including subclinical problems.

5.1.5. Attrition

Attrition is a major concern in most longitudinal studies, and attrition rates up to 70% are often reported (Gustavson, von Soest, Karevold, & Røysamb, 2012). If those who drop out of the study differ from those who stay, estimates may be biased. Previous studies have shown that estimates of associations between variables tend to be more robust against selective attrition and non-response than estimates of means and frequencies (Gustavson & Borren, 2014; Nilsen et al., 2009). Findings from the Avon Longitudinal Study of Parents and Children (ALSPAC) showed that despite some bias in association estimates, valid conclusions could be made about the direction of the associations and their approximate magnitude (Howe, Tilling, Galobardes, & Lawlor, 2013). Findings from the Danish national Birth Cohort suggest that most association estimates were relatively unbiased, while others were more clearly biased (Greene, Greenland, Olsen, & Nohr, 2011). Monte Carlo simulations have shown that association estimates may be clearly biased when non-response is related quite strongly to both the exposure and the outcome variables (Gustavson & Borren, 2014). In attrition analyses in Paper I in the current study, we found that mothers who had returned the eight-year questionnaire, had slightly better mental health, than mothers who did not return the eight-year questionnaire (see Table 1 in Paper I). The differences between the two groups on the child functioning scores (Cohen's d) were all less than 0.1, which is considered a weak effect.

5.2. Interpretation of the findings

In the current thesis, we have tested different theoretical perspectives on the association between language difficulties and internalizing problems. In Paper I, we investigated developmental cascades, in terms of how problems in one area predicted problems in another. In Paper II, we turned to an in-depth study of a developmental cascade, when investigating mechanisms in terms of indirect pathways between language difficulties and internalizing problems in a mediation model. In Paper III, we used theories on comorbidity to investigate if an underlying common family factor could explain problems in both areas. The results from all three papers could be understood in terms of Willcutt's (2014) different types of comorbidity. We will especially focus on how our results may be understood in terms of causality, developmental cascades and a common underlying factor.

5.2.1. Causality

In previous research, it has commonly been assumed that there is a causal relationship between language difficulties and internalizing problems. This has been seen in wordings like "impact on" and "effect of" (Masten & Cicchetti, 2010). The large literature on mechanisms also indicates an underlying assumption that problems in one area lead to problems in the other. Explanations have been proposed such as language difficulties leading to exclusion from play, again leading to sadness and withdrawal (Carpenter & Drabick, 2011; Liiva & Cleave, 2005; Troesch, Keller, & Grob, 2016). It has been suggested that assumptions of causal mechanisms in developmental research are often supported when associations are well-replicated and when associations remains after controlling for confounders (D'Onofrio et al., 2013).

We do not know if the associations found in the current thesis are causal. For an association to be regarded as causal, 1) the cause needs to precede the effect, 2) the cause and effect need to be related, and 3) there needs to be no other plausible explanations for the effect (Cook, Campbell, & Shadish, 2002). In the current thesis, we used longitudinal data that enabled us to determine associations in a time sequential manner, and found associations both between early language difficulties and later internalizing problems (Papers I and II) and between early internalizing problems and later language difficulties (Paper I). In both studies, we adjusted for baseline, so that we investigated how problems in one area predicted change in problems in the other area over time. Thus, Paper I and II fulfilled the first two criteria. To determine if there are other plausible explanations for the association, it is common to adjust for covariates. In Paper I and II, we controlled for possible confounders that could explain problems in both areas, such as mother's age, educational level and parental language other than Norwegian. It has be argued that a mediation model, such as presented in Paper II, by definition is a cause-effect process (Hayes & Rockwood, 2017). In future studies, different mediation models could be tested against each other, with clear a priori hypotheses about mediators and confounders (Spector & Brannick, 2011).

Researchers have however pointed out that only to include covariates to control for selection factors is not sufficient to determine causality and a suggestion has been made to a priori identify alternative explanations and then use diverse designs to test competing causal hypotheses (D'Onofrio et al., 2013). It has been warned that uncritical use of control variables does not necessarily purify the association of study and it has been recommended to be explicit about the hypothesized role for all variables (Spector & Brannick, 2011).

In the current thesis we do not claim the association between language difficulties and internalizing problems to be causal, but we still highlight the importance of research illustrating that problems in one area may predict problems in another area. Although we could not exclude that there were other confounding factors causing problems in both areas, the results in Paper I and II provided knowledge that problems in one area *predict change* in the other area. This has been called for, as very few studies control for initial level of internalizing problems (Yew & O'Kearney, 2013). Even though we cannot determine causality in the current thesis, neither confirm nor disconfirm, it is valuable to identify markers for developing comorbidity and to early identify risk groups. This information may be used to target intervention at particular developmental ages.

A causal explanation does not take into account that other environmental or genetic factors may explain problems in both areas. Randomized controlled trials have been noted as the gold standard in terms of finding causal mechanisms. As this is impractical or unethical to carry out in many areas of psychology, alternative methods have developed to let us approach findings of causality. Several quasi-experimental designs have been developed to test causal hypotheses by excluding possible alternative explanations, such as sibling designs (Lahey & D'Onofrio, 2010).

5.2.3. Bidirectional associations and a common underlying factor

In the current thesis, we have found support both for developmental cascade models where problems in one area predicts problems in the other, and for a model where a common underlying factor predicts problems in both areas.

The advantage of using developmental cascade models is that the design enables the study of multiple areas in the same model at multiple time points, which gives a picture of what associations are particularly salient at different ages. In Paper I, we found that the associations between language difficulties and internalizing problems were especially salient from five to eight years during the transition to school, and we found associations across different types of language difficulties, such as expressive and receptive difficulties. Developmental cascade models are also flexible in describing pathways of development across several domains. In Paper II, we used this framework to describe possible indirect pathways between the two areas, and found that emotion regulation and social engagement were possible pathways, while we did not find any pathways thought empathy and assertiveness.

However, in both of these models, the associations were predictive and not necessarily causal. That is, we could describe patterns, but we do not know *why* the associations developed. To approach the question of why these associations develop, we used a different method in Paper III. Here we found that comorbid language difficulties and internalizing problems at five and eight years could be explained by a common underlying factor shared by siblings. The logic of this model was that if one problem *causes* change in the other, associations between language difficulties and internalizing problems should be stronger within than between siblings. The common factor captured covariance that was equal within as between siblings and was thus not likely to reflect causal mechanisms. Residual covariance between internalizing problems and language difficulties not explained by the common factor, was also modelled.

In other studies on common underlying factors, it has been argued that even though there is strong evidence of a general psychopathology factor, it is difficult to rule out that these findings may be due to causal associations between the constructs (Lahey, Krueger, Rathouz, Waldman, & Zald, 2017). In Paper III, we made an attempt to investigate this through including longitudinal direct pathways between language difficulties and internalizing problems within siblings. We found that including direct pathways between difficulties did not give better fit to the model, than only including a common underlying factor. In a similar study, the authors found that the association between language skills and self-control was explained both by a common factor and causal associations (Beaver, DeLisi, Vaughn, Wright, & Boutwell, 2008).

It has been suggested that if there is a causal relationship between language difficulties and internalizing problems, it is more likely that this effect is visible in a school setting, where there are different social an academic demands. Indeed, Redmond and Rice (1998) suggested that teachers are more likely to regard children with language difficulties as socially immature compared to other children, whilst parents know the child in more detail and also know the communication strengths in a less socially and academically demanding environment. It would be interesting to replicate the model in Paper III with teacher report measures.

In sum, the existence of a common factor is also compatible with results from Paper I and II, in that an underlying factor may explain the development of problems in both areas. We were, however, not able to determine the content of the common factor with the current design, but will discuss possible explanations based on results from Paper II, and previous findings.

Emotion regulation and social skills

In the mediation model in Paper II, we were not able to determine the direction of the associations between the mediators and the internalizing outcome as they were measured at the same time point. Nevertheless, the analyses provided us with information about areas of child functioning that are important in the association between language difficulties and internalizing problems. In Paper II, we found that social engagement and emotion regulation mediated the association between language difficulties and internalizing problems.

It has been found in previous research that the temperament trait negative emotionality is related to psychopathology (Tackett et al., 2013). Furthermore it has been found that negative emotionality is more associated with a general psychopathology factor than with specific disorders in children and adolescents. In the same study, they found a stronger correlation between negative emotionality and a general psychopathology factor than for other temperament traits such as prosociality, and that this correlation was accounted for by genetic influences (Tackett et al., 2013). There is less knowledge about the role of negative emotionality in language difficulties, but one study showed increased levels of difficult temperament in a children with language impairment compared to typical language development, and no difference on sociability (Prior, Bavin, Cini, Eadie, & Reilly, 2011).

Emotion regulation is commonly thought to develop as a combination of a neurobiological basis in combination with environmental parent–child interactions (Maughan & Cicchetti, 2002). We could hypothesize that emotional and social climate in the family is associated with language and internalizing both in terms of genetic and environmental transmission, and these areas are again important aspects of temperament.

Neurobiological explanations

It has been suggested in previous research that the association between language difficulties and internalizing problems could be explained by common underlying neurobiological vulnerabilities (Yew & O'Kearney, 2013) or neurodevelopmental immaturity (Snowling, Bishop, Stothard, Chipchase, & Kaplan, 2006), which could include several areas of functioning.

Firstly, emotion regulation has a neurobiological basis, as mentioned above. Another aspect of neurodevelopment that may be more associated with social skills, is theory of mind, and studies have found evidence of increased deficits in theory of mind in a group of adults with a history of SLI (Clegg, Hollis, Mawhood, & Rutter, 2005). Similarly, others have

suggested that it is possible that social cognition may account for problems in both areas, suggesting continuities with autism-spectrum disorders (Snowling et al., 2006)

Another neurological explanations that could be underlying difficulties in both areas is executive functions. Executive functions involves cognitive regulation such as shifting between tasks, effortful control, and working memory. There is evidence that executive functions are associated both with a general internalizing factor (Hankin et al., 2016) and with a general psychopathology factor (Caspi et al., 2014). As language difficulties also have been found to be associated with executive functions in terms of phonological short-term memory (Bishop, Laws, Adams, & Norbury, 2006), a common underlying factor may involve this area. Finally, attention could also be associated with a common underlying factor as this area has been seen as relevant for both internalizing problems (Hankin et al., 2016) and language difficulties (Cohen et al., 2000). It is however likely that it is not deficits in only one area that composite a common underlying factor, but across several areas (Pennington, 2006).

Shared environment explanations

It is possible that factors in the environment shared by siblings could explain the comorbid difficulties. Children do not live in isolation, and internalizing problems measured by a scale could reflect withdrawal and anxiousness as reactions to a dysfunctional environment, and not a problem in the child per se. Examples of this could be parental conflict, economic problems, or illness. In clinical diagnostic assessment, these factors are often taken into account which we were unable to do in the current study.

In a large population based study, several early predictors of comorbid language difficulties and socioemotional and behavioral problems at school entry, were identified (Hughes, Sciberras, & Goldfeld, 2016). They found that four factors predicted comorbidity in children: witnessing violence, a history of parent mental illness, living in a deprived community, and parental education levels. Others have also suggested social adversity to be a possible common factor (Snowling et al., 2006). A limitation in studies like these, is that they only provide knowledge about factors that are studied. Furthermore, all of the environmental factors they found could also be explained though heritability of traits or neurobiological factors. The results of two meta-studies showed that parenting explained 8% of the variation in childhood depression and less than 4% of the variation in childhood anxiety (McLeod, Weisz, & Wood, 2007; McLeod, Wood, & Weisz, 2007). It is however important to note that even though parenting may not be an important cause of developing comorbid language

difficulties and internalizing problems in children, parents may play an essential part in potentially resolving these problems (Salmon et al., 2016).

Across all of these areas, whether the common factor involves emotion regulation, executive functions or social skills, the question remains if the common factor is transmitted from parents to children through genes, the environment or a combination.

Heritability

The design of the current study, did not allow us to determine if the common family factor was genetic or environmental. To do so requires designs that can separate these factors, such as twin studies. In twin studies, it is possible to separate influences of additive genetic effects (A), shared or common environmental effects (C) and non-shred environmental effects (E) (Røysamb & Tambs, 2016). The common factor found in Paper III in the current study includes A and C, but excludes E. We could speculate based on previous findings on heritability of language difficulties and internalizing problems separately.

Studies including children with SLI have found estimates of genetic heritability as high as 50% to 75% for school age children (Bishop, 2006). Twin studies on internalizing problems have suggested that genetic factors account for 50% of the variation in internalizing problems in toddlers and 30% is accounted for by shared environment factors (Tandon et al., 2009). Longitudinally, twin studies may also provide information about how genes and the environment affect the stability and change of development over time. Genetic factors commonly have high influence on stability, but have also been found to be particularly important at certain ages (Røysamb & Tambs, 2016). In a large meta-analysis on environmental contribution to child psychopathology, it was concluded that shared environmental influences accounted for 12% to 16% of the variance in internalizing problems (Burt, 2009). They also found that the shared environmental influence decreased over time and explained less of the variation in internalizing problems in adolescence than in childhood. In the current study we found that the common underlying factor was highly stable over time, but as our highest measurement point was eight years, we did not explore this into adolescence.

Even though we cannot separate genetic and shared environmental contributions in the development of comorbid language difficulties and internalizing problems, our results do maintain that the importance of non-shared environment plays a lesser role than anticipated in previous studies. Non-shared environment, such as friends, teacher, and class, appear

important for each of the conditions examined, but less for their comorbidity and co-development.

5.2.4. Gender differences

In Paper I, we found that the bidirectional associations between semantic language difficulties and internalizing problems from five to eight years were significant and strong for girls, but not for boys. This is in line with previous findings on associations between reading disabilities and internalizing problems found for girls, but not for boys (Willcutt & Pennington, 2000). In Paper I, we dichotomized the study variables so that the children with the most difficulties relative to their gender were compared to other children of the same gender. In Paper II, we did not find the same gender difference from five to six years. This could be partly due to a shorter time span, the use of continuous measures, or a different sample. There is a possibility that the boys in the sample overshadowed the girls. We did not use gender specific categories when sampling on children with probable language difficulties in Paper II. As girls in general have better language skills than boys, the sample consisted of higher levels of language difficulties for boys than girls. We did however find gender difference in the concurrent association between semantic language difficulties and internalizing problems at five years in Paper II, indicating that a stronger association for girls had already developed by this age. In Paper III we were unable to investigate gender differences as this would lead to too small groups. Instead, all observed variables were adjusted for gender. Previous research has indicated that comorbid associations are stronger for girls than boys, for example between ADHD and depression and anxiety (Solberg et al., 2018) and between conduct disorder and depression (Zahn-Waxler, Shirtcliff, & Marceau, 2008). This would be an important focus to pursue in future research on a common factor for language difficulties and internalizing problems.

5.2.5. Clinical implications

Knowledge about predictive associations and common underlying factors does not indicate what type of intervention that would be effective, but rather targets for intervention. It is especially important to be aware of possible neurodevelopmental disabilities in interventions based on verbal communication. It is possible that a child does not understand or is unable to explain what they mean due to language difficulties. It is also important to be aware that problems in one area may cover up problems in the other area. For example, from a clinical

viewpoint, a withdrawn child may be interpreted as unmotivated at school, but the behaviour may reflect an underlying problem with understanding or producing language. In general, there is a need for increased awareness for these problems, as studies have showed that children with language difficulties and internalizing problems may be difficult to detect in a classroom setting (Stowe, Arnold, & Ortiz, 1999) and have limited access to specialist services (Heiervang et al., 2007). Regardless of whether the association between language difficulties and internalizing problems is causal or there is an underlying common factor, a child who has difficulties in one area is also at risk of needing help in the other area. Given that symptoms might fluctuate over time in childhood, it may be important to also consider more than one area of functioning, to best guide intervention.

5.2.6. Future research

In future studies it would be essential to further explore the shared family factor, and family designs could shed light on the heritability mechanisms (D'Onofrio et al., 2013). To further investigate transmission through shared environment, parenting style would also be interesting to explore. Children do not develop in isolation and for children 18 months to eight years, as in the current study, parents contribute important surroundings. In the MoBa sub-study, we have collected information about how parents react to children's emotions, such as anger or sadness though the *Coping with children's negative emotions scale* (Fabes, Poulin, Eisenberg, & Madden-Derdich, 2002). It could be possible to explore if adaptive emotion regulation is transmitted through parenting styles. It is already some evidence that developmentally targeted parent-child conversations may be beneficial for this group (Salmon et al., 2016), but this is an area that warrants further investigation.

Similarly, it would be interesting to further explore other cognitive correlates of the common factor, such as social cognition and executive functions, on other psychopathology correlates such as externalizing problems, eating disorders or thought disorders. Finally, it would be interesting to test the role of language difficulties and cognitive correlates in three levels of psychopathology, from a general psychopathology factor, to an internalizing factor and specifically for symptoms of anxiety and depression.

5.3.   Conclusion

The results from the current thesis show the relevance of language difficulties in the understanding of mental health in children. We have found bidirectional associations between language difficulties and internalizing problems, explored potential mechanisms and finally, showed that there is an underlying family factor that may account for the association between difficulties in these two areas. Previous research on the association between language difficulties and internalizing problems has been dominated by explanations of causal mechanisms, such as exclusion or withdrawal from play. The reason for this may be that the study of language difficulties has been the field of pedagogues who are more tuned in to peer play and social mechanisms, as opposed to mechanisms in the families. Future research should be more explicit about the theoretical framework and academic discipline it stems from, to make the results more comparable. In order to more fully understand child functioning, we need to incorporate this knowledge into a broader picture. The results from the current thesis indicate that there should be an increased awareness of cognitive correlates to emotional problems, as well as knowledge about common family factors.

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