

**Systematic review of sensory processing in preterm children reveals abnormal sensory modulation, somatosensory processing and sensory-based motor processing**

**Short title:** Review of sensory processing in preterm-born children

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## **ABSTRACT**

**Aim:** Preterm birth poses concerns in daily functioning and behaviour in childhood, possibly connected to sensory processing disorder. This review aimed to systematically identify assessments, incidence and nature of sensory processing disorder in preterm-born infants and children.

**Methods:** We searched literature through CINAHL-EBSCOhost, Cochrane, Ovid/PsychINFO, PubMed/Medline, Scopus and Google Scholar, published until November 2018. We included electronically available, peer-reviewed studies of preterm-born children that applied standardised sensory processing assessments. We excluded studies of preterm-born children with major neurodevelopmental impairments.

**Results:** We identified 27 studies of premature children, aged from birth to nine years seven months. The assessments represented three versions of Sensory Profile questionnaires and three clinical tests, Test of Sensory Functions in Infants, the Miller Assessment for Preschoolers, and the Sensory Integration and Praxis Test. The studies revealed wide variation of atypical sensory processing: 28–87% in sensory modulation, 9–70% in somatosensory processing, and 20–70% in sensory-based motor processing.

**Conclusion:** Preterm-born children exhibited elevated risk for sensory processing disorder from infancy into school age. Routine screening of sensory processing, intervention intervals and parental consultations should be considered in ameliorating sensory processing and neurocognitive development. Moreover, a larger body of intervention studies is needed.

**Key notes**

- This systematic review explored assessments, incidence and nature of sensory processing disorders in preterm-born population.
- Preterm infants and children exhibited evidence for an elevated risk, compared to their term-born peers, for atypical sensory modulation, somatosensory processing and sensory-based motor processing.
- It is important to follow up and support the development of sensory integration and processing in preterm-born children at least until school age.

Key words: assessment, neurodevelopment, preterm children, sensory integration, sensory processing

## **BACKGROUND**

Premature birth causes substantial risks for impairments in motor (1,2), neurocognitive (3) and behavioural (4) development. The neonatal intensive care, excessive sensory stimuli (5,6) and multiple painful procedures may distress the infant's brain development. The stress may affect the infant's multisensory integration processes, perception of self and environment and lead to adverse consequences in childhood development. (5,7)

The development of adequate sensory integration and processing is an important precursor for learning and behaviour regulation. Sensory integration, as defined by Ayres (1989), is a neurological information process that organises sensations from the body and environment. Somatosensory, vestibular and visual sensations are seen foundational for this process. With somatosensory, the theory refers to body-related tactile and proprioceptive senses. Along with the organisation process, the brain constantly selects, inhibits or enhances sensory information and responses to it. It is through experiences and purposeful activities that the information is further compared, associated and integrated. The brain develops concepts, abilities to plan and to interact within the environment. (8) Dysfunctional sensory integration processing complicates performance and participation in daily activities. The difficulties are heterogenous and depend on the nature of the underlying dysfunctional domains. (8-11)

The conceptual model of sensory processing by Dunn (10) hypothesises a connection between individual neurological threshold and behavioural response strategies. This is described in quadrants where the continua of the neurological threshold and the response strategy form axes. The neurological threshold mirrors the amount of sensory stimulus required for a neuronal excitation response. The brain sensitises to stimuli that require immediate attention and response and habituates to familiar stimuli that do not require constant attention. This is balanced through modulation, the brain's regulation process that facilitates some responses and inhibits others. The behavioural response continuum varies

from passive to active regulation strategies that reflect one's individual temperament or personal traits. A person within low registration quadrant has high neurological threshold and passive behavioural response strategy. Therefore, he or she is unaware of surroundings and remains disengaged. A person within sensory sensitive quadrant has low threshold and passive response strategy. Hence, he or she is easily distracted and has difficulties in busy situations offering sensations. A person within sensation seeking quadrant has high threshold and active response strategy, and therefore, thrives for opportunities with strong sensations. Someone within sensation avoiding quadrant has low threshold and active response strategy, and tends to control sensations, perhaps, with compulsive behaviour. (10,12)

The construct of sensory processing disorder, SPD, by Miller et al (9,13) proposed a nosology, when sensory integration dysfunction impairs daily activities, routines or roles. The construct hypothesised three main categories and six discreet subtypes of disorder that may be present in different combinations. The main categories are sensory modulation disorder, sensory discrimination disorder and sensory-based motor disorder. The first category, sensory modulation disorder, describes a difficulty to grade and regulate sensory stimuli. Three subtypes were proposed, sensory over-responsivity, sensory under-responsivity and sensory seeking/gravating. Sensory over-responsive person responds to weak stimuli or reacts faster, longer and more intensively than what is typical. Someone sensory under-responsive, becomes aware of stimulus only when it is excessively strong, and gives an impression of inactivity, calmness, or numbness. Sensory seeking/gravating person, instead, acts to gain intense sensations through many senses. The second category, sensory discrimination disorder, describes difficulty to interpret sensation, its locations, similarities or differences. It may occur in the visual, auditory, gustatory, olfactory, vestibular, tactile and proprioceptive systems. The third category, sensory-based motor disorder, includes subcategories of postural disorder and dyspraxia. Postural disorder may demonstrate in hypotonic or hypertonic muscle tone, inadequate movement and eye-movement control. It also may occur as difficulty in

automatically adjusting position during activities or in reaching and keeping resistance against gravity. Dyspraxia is defined as difficulty in generating ideas, planning, sequencing or executing novel actions. It typically coincides with tactile, proprioceptive or vestibular under-responsiveness or discrimination deficits. (9,13).

These reference frames highlight the importance of body-related senses, tactile, proprioceptive and vestibular systems, for normal development of body-perception, motor abilities, behaviour and learning. (8-13) In this study, we referred to sensory integration and processing as sensory processing.

Little is known about the nature of sensory processing disorders among preterm population. An effective intervention builds on normative measures and evidence-based knowledge of underlying problems (14,15). We, therefore, conducted a systematic literature review aiming to detect all commercially available, standardised sensory processing assessments represented in preterm studies. We also sought to identify the incidence and nature of sensory processing disorders in preterm-born infants and children.

## **METHODS**

A comprehensive literature search was conducted to identify studies on preterm children with sensory processing assessments. We searched through international databases: CINAHL-EBSCOhost, Cochrane, PubMed/Medline, Science Direct and Scopus. We screened citation indices in Scopus and Google Scholar and manually searched reference lists of identified studies. No grey literature was searched.

The inclusion criteria comprised all electronically available, peer-reviewed observational studies that were published before 8 November 2018. The subjects were children born before 37 weeks of gestation. The summary outcome measures reported on standardised and commercially available sensory processing assessments. The studies were either systematic reviews or original research papers. The sampling was randomised or non randomised, with or without a control group. Publication languages were English, Finnish, French, German, Italian or Swedish.

The exclusion criteria comprised case reports, expert opinions, dissertations, or studies not reporting any summary outcome measures of the standardized sensory processing assessments on preterm-born children. Also excluded were studies of preterm participants with major neurosensory and developmental impairments such as deafness, blindness, cerebral palsy, mental retardation, or chromosomal abnormality and hereditary syndromes that significantly alter sensory processing.

The search question followed the population-intervention-comparison-outcome, PICO, principle (16). The search terms were either medical subject headings, MeSH, or search words with Boolean operators such as OR and AND. The key terms were truncated with a symbol \* or similar, to expand the search options to variations of the words. For example, prematur\* expanded the search to premature, prematurely or prematurity. The key terms were preterm,



prematu\*<sup>r</sup>, sensor\*<sup>r</sup>, sensation, motor skill\*<sup>r</sup>, perceptual skill\*<sup>r</sup>, development\*<sup>r</sup> and the name of the standardized sensory processing assessment identified by Williams and Anzalone (17), Eeles et al (18) and Jorquera-Cabrera et al (19). Manual search of the reference lists of the papers included and of the citation indices supplemented the systematic search. Table S1 illustrates an example of the systematic search retrieval protocol. The first author administered the search. Uncertainties of decisions were resolved through discussion and consensus with members of the research team.

The review applied Equator network recommendations for methodological quality assessments of the studies included: Preferred Reporting Items for Systematic Reviews and Meta-Analyses, PRISMA (20), Consolidated Standards of Reporting Trials, CONSORT for randomised controlled trials (21) and the Strengthening the Reporting of Observational Studies in Epidemiology, STROBE (22) for non-randomised, observational designs.

## RESULTS

The comprehensive literature search (Figure 1) yielded 27 studies, comprising three systematic reviews (Table 1) and 24 original studies (Table S2). All studies were published in English but originated from 11 countries and five continents. In case a study was presented in multiple publications, the review included the most recent paper (23). The review accepted a second publication if the sensory processing outcome measures were different (24-29). Table S3 lists the studies excluded and the reasons behind the rejections.

### Included studies

The included three systematic reviews (Table 1) were published in years 2015 and 2017. Mitchell et al (30) synthesised in their review evidence of sensory processing disorder in multidisciplinary studies with preterm-born children from birth to three years of age. They analysed outcomes from 45 studies with physiological, behavioural, temperament or sensory processing research. Of those 45 studies, one was included in our review (31). The review of Bröring et al (32) reported on sensory modulation problems, their risk factors and associations with neurocognitive and behavioural problems in preterm children. Of the 18 included studies, three concerned general population with prematurity as a risk factor. Of the other 15 studies designed for preterm infants and children, we included 11 in the current review. The review of Machado et al (33) reported on evidence of sensory processing in preterm infants. The review comprised eight studies, which all met the inclusion criteria of the current review.

The included 24 original studies were published between years 1994 and 2018. Of these, 20 were explorative and four were descriptive studies. Among the descriptive studies, two reported on intervention effects of individualised sensory integration therapy (28,34). Another reported on a physiotherapy and parental guidance programme (35) and one on fatty acid supplement effects on sensory processing outcome (36). Table S2 presents a summary of the original studies and outcomes.

### **Methodological appraisal**

The methodological appraisal (20-22) of the studies included (Table S4) varied from low to high risk of bias. The risks of bias concerned missing descriptions such as participant demographic background, participant selection process, allocation and matching of controls, blinding groups and loss of follow-up participants in the cohort studies. Six papers included a flow-diagram of participants during the study process. The risk of bias on the outcome level consisted of a lack of adjustments for confounders and nonexistent demographic comparison between the participants and eventual drop-out groups. Small sample sizes decreased the strength of the studies and possibly led to type II errors.

### **Participants**

The included original studies reported on 2,556 children, of which 1,875 were preterm (56% boys) and 681 term-born control participants (47% boys). In addition, the systematic review of Mitchell et al (30) covered 2,584 participants. The review of Bröring et al (32) covered in population-based studies 2,575 participants and in the preterm studies additional 625 participants that were not included in our review. All studies together, reported on 8,340 participants.

The gestational age in the included original studies varied between 22 and 36 completed weeks for the preterm-born children. The term-born children were born between 37 and 42 weeks of gestation. Extremely low gestational age under 28 weeks or birth weight under 1,000g, were reported in four studies (26,27,37,38). Very low gestational age under 32 weeks or birth weight under 1,500g were reported in 15 studies (24,25,28,29,35,36,39-47). Other five studies reported gestational age under 37 weeks (23,31,34,48,49). The participant age ranged at the time of the studies from four months to nine years and seven months. We identified no studies of preterm-born adolescents or adults. The sample sizes varied from 24 to 243 participants. (Table S2)

### **Sensory processing assessments**

The search identified two types of assessments, clinical measures and caregiver questionnaires. The clinical measures, present in 15 studies, were the Test of Sensory Functions in Infants, the Miller Assessment for Preschoolers and the Sensory Integration and Praxis Test. The caregiver questionnaires, present in 13 studies, were the Infant/Toddler Sensory Profile, the Sensory Profile and the Short Sensory Profile.

### **Clinical measures**

**The Test of Sensory Functions in Infants (TSFI).** The TSFI is designed for infants between four and 18 months of age. The test screens regulatory disorders, developmental delays, risk of learning and sensory processing disorders. The TSFI contains 24 items that are divided in five subdomains: reactivity to deep pressure, adaptive motor functions, visual-tactile integration, ocular-motor control and reactivity to vestibular stimulation. The TSFI scores describe normal, at-risk (6<sup>th</sup>–25<sup>th</sup> percentile), or deficient (<5<sup>th</sup> percentile) sensory processing. The test is most accurate for categorizing sensory dysfunction from 10 to 18 months of age and normal function from four to 18 months of age. The test manual advises combining the results with other standardized comprehensive developmental assessments (50). The TSFI was applied in eight studies (23,28,29,31,34,42,48,49).

**The Miller Assessment for Preschoolers (MAP).** The MAP contains 27 items. It is designed for screening delays in sensory and motor foundations, coordination, cognitive verbal, cognitive non-verbal and combined complex abilities. The test is designed for children between two years nine months and five years eight months of age. The MAP scores describe normal, moderate (6<sup>th</sup>–25<sup>th</sup> percentile) or severe (<5<sup>th</sup> percentile) delays of development (51). The MAP was applied in two studies (38,45).

**The Sensory Integration and Praxis Test (SIPT).** The SIPT contains 17 individual tests that distinguish performance irregularities in tactile, proprioceptive, vestibular and visual

processing, eye-hand coordination, bilateral motor coordination and praxis. The SIPT is designed for children between four years and eight years 11 months of age. Low scores in the individual tests indicate severe (-3.0 to -2.5), definite (-2.5 to -2.0), or mild (-2.0 to -1.0) dysfunction or difficulty. The non-dysfunctional scores are divided into typical (-1.0 to 1.0), above average (1.0 to 2.0) and advanced (2.0 to 3.0) performance. The SIPT offers a profile of test performance but no total scores. (8,52) The SIPT appeared partly in five studies that employed between one and six of the possible 17 individual tests (27,37,41,44,46).

### **Questionnaires**

The Sensory Profile questionnaires identify problems of sensory modulation and self-regulation behaviours. The questions are grouped in tactile, visual, oral, auditory, vestibular and multisensory sections. The responses of the sections are also categorised in four quadrants according to the neurological threshold and behaviour response strategy. High neurological threshold appears as low registration or sensation seeking. Low threshold appears as sensory sensitivity and sensation avoiding. The Infant/Toddler Sensory Profile (ITSP) contains two parts. The infant form is designed until six months of age with 36 items. The toddler form is designed for seven to 36 months of age with 48 items (12). Only the toddler form was identified in the included studies. The Sensory Profile (SP) questionnaire comprises 125 items and its shortened version, the Short Sensory Profile (SSP) covers 38 items. These SP and SSP are designed for children between three and 12 years of age. (53) The questionnaire scores are divided into three categories, normal, possible atypical (>1SD) and atypical (>2SD). (12,53). The ITSP was applied in nine studies (24-26,28,29,31,36,40,47), the SP in three studies (35,41,47) and the SSP in two studies (39,43).

### **General outcome in sensory processing**

The studies of preterm-born infants and children without major neurodevelopmental impairments showed strong elevated risk, compared to term-born children, for atypical sensory processing. The systematic review of Machado et al (33) argued that preterm birth negatively

impacts on sensory processing in childhood. TSFI test total scores indicated at-risk or deficient sensory processing in 37–82% of the preterm infants (23,31,42,48,49) and in 0–22% of the term-born infants (31,48,49). The MAP total scores indicated moderate delay for 34–70% of the preterm-born children and for 14% of the term-born children. Severe delays in the MAP occurred in 20–40% of the preterm-born children and 4% of the term-born children. The SIPT mean scores of the individual tests remained 0.4–1.5 SD lower for the preterm children than for those of the controls or the population norm (27,37,41,44,46).

### **Outcomes in sensory modulation**

Sensory modulation disorder was evidenced in two systematic reviews (30,32) and in nine original studies (24-26,29,31,36,39,43,47). Children born preterm demonstrated significantly more sensory modulation deficits, compared to full-term children. The results were, however, reported in various, non-comparable ways. According to the Sensory Profile caregiver questionnaires, one or more sections or quadrants displayed definitely atypical response (>2SD) in 28–39% (43,47) and possible atypical (1SD) response in 37–87% of the preterm children (26,31,36,39,47). Atypical responses were non-existent among full-term children and possible atypical responses occurred in 12% of the full-term children (39). Among preterm children, the prevalence of possible atypical modulation was 7–65% in auditory, 16–46% in visual, 9–45% in tactile, 18–61% in vestibular, and 18–36% in oral section. Possible atypical quadrants appeared in low registration 23–71%, sensation seeking 14–33%, sensory sensitivity 7–62% and sensation avoiding 18–54%. TSFI outcome for reactivity to vestibular stimulation was deficient in 24–80% and at risk or deficient in 21–30% among preterm infants. TSFI tactile reactivity appeared deficient in 21–93% and at risk or deficient in 17–49% of the preterm infants. (23,29,31,42) The review of Mitchell et al (30) found 44% of positive evidence on sensory processing disorder, mostly sensory modulation disorder (43%) occurring in sensory over-responsivity. The systematic review of Bröring et al (32) found evidence of sensory modulation disorder in 16 of the included 18 studies. The variety and nature varied

widely. Most frequently affected was low registration quadrant and auditory modulation, both found problematic in six studies. Oral, tactile, vestibular and visual modulation was atypical in four studies.

### **Outcomes in sensory discrimination**

Sensory discrimination problems of preterm children were identified in nine papers. According to the TSFI, the prevalence for visual-tactile integration was at-risk or deficient in 18–33% in preterm infants (29,31,42). The MAP outcomes for tactile discrimination and stereognosis are included in the sensory and motor foundations index (51) and are, therefore, reported in the sensory-based motor processing section. The SIPT prevalence for mild to severe dysfunction in somatosensory processing tests was 9–71% in the preterm-born children and 0–30% in the term-born controls (27,37,41,44,46). Poor performance in tactile processing was reported in five studies (27,37,41,44,46) and poor kinaesthetic discrimination in one study (41). The SIPT somatosensory processing scores among preterm children were significantly lower ( $p < 0.001$ ) compared to the population means: in kinaesthesia (-1.1 SD), finger identification (-0.9), manual form perception (-1.5), graphesthesia (-0.85) and localization of tactile stimuli (-0.99) (44).

### **Outcomes in sensory-based motor processing**

Sensory-based motor problems of preterm children were confirmed in a total of 16 studies. The TSFI prevalence of at-risk or deficient outcome in preterm infants was 27–79% in adaptive motor functions (23,29,31,34,42,49) and 3–50% in ocular-motor control (29,31,42). The MAP prevalence of moderate delay was 10–45% in sensory and motor foundations and 44–70% in coordination in preterm children. Prevalence of severe delay was 0–11% in abilities of sensory and motor foundations and 0–11% in coordination. (45) In the SIPT sensory-motor processing, prevalence of mild to moderate dysfunction was 20–40% in preterm children and 3–12% in term-born controls. The prevalence of moderate to severe dysfunction was 8–12% in the preterm-born children. and 6–10%, respectively, in the term-born controls (27,46).

Visual-motor integration deficits were reported in four studies (27,37,46,48). The SIPT prevalence for mild to definite dysfunction in eye-hand motor accuracy was 31%, and definite to severe dysfunction was 10% in preterm children (27,46) and 3% and 0% in term-born controls, respectively (27). Preterm children, who performed in the SIPT motor accuracy and finger identification significantly worse than the controls, were also slower in handwriting ( $p = 0.004$ ) (46).

Motor planning problems (27,38,45) and dyspraxia (37) were addressed in four studies. There were significantly lower scores in the SIPT tests of praxis, visual perception and processing among the preterm children, compared to the matched control children. The extremely preterm children with developmental coordination disorder scored lower in praxis tests than those without coordination disorder.(37) SIPT postural praxis test identified mild dysfunction in 33% and definite to severe dysfunction in 12% of the preterm children and 12% and 0%, respectively, in term-born children (27). MAP combined complex abilities, that also require praxis skills, showed moderate delay in 0–36% and severe delay 11–45% in preterm children. (45)

Associations with other motor tests were reported. TSFI outcomes in visual-tactile integration (48), reactivity to tactile deep pressure (23) and reactivity to vestibular stimuli (23,48) were strongly associated with adverse ( $\leq 5^{\text{th}}$  percentile) motor outcomes in the Alberta Infant Motor Scales. One study identified a strong association between low TSFI total score and Alberta Infant Motor Scales (48), but another study found no such evidence (23). One study found no association of Sensory Profile scores with motor outcome (47).

### **Outcomes on sensory processing intervention studies**

Sensory integration intervention effects were reported in two studies. Pekçetin et al (34) adopted eight weekly sensory integration treatments for a group of preterm infants at the corrected age of seven to nine months. A control group of term-born children had no



treatment. The study applied TSFI before and after the treatment period and compared the outcome with the non-treated group. The intervention showed significant positive effect on sensory processing, more precisely in visual-tactile integration and adaptive motor functions. Lecuona et al (28) implied 10 individualised weekly sensory integration treatment sessions to a group of preterm infants from low socioeconomic level families. The control group of preterm children from low socioeconomic background had no treatment. The study found a significant positive treatment effect on the treatment group, compared to the control group. This was verified with pre- and post-tests by the Bayley Scales of Infant and Toddler Development – third edition (BSID-III), TSFI and ITSP. The mean scores of the treatment group's motor, cognitive and language development and sensory processing improved, whereas the control group had more modest improvement, or poorer performance in all the post-tests. Further two studies applied sensory profiles to obtain results from other interventions: a fatty acid supplementation trial (36) and a parent-guidance and physiotherapy program for preterm infants (35).

### **Associations of sensory processing with neurocognitive development**

A total of 11 original studies reported on associations of sensory processing with neurocognitive development (24-29,39,42,45,47,49). Poor performance in the TSFI total score, visual-tactile integration, adaptive motor function (49) and ocular-motor control (42) was associated with poor cognitive scores in BSID-III. ITSP adverse sensation seeking and low registration, auditory and touch processing were associated with low Mental Developmental Index in BSID second edition (24). Poor TSFI oculomotor control was associated with low motor scores in BSID-III (42). Atypical Short Sensory Profile overall scores were associated with poor executive functions, more specifically working memory and inhibition (39). No association with Sensory Profile and cognitive outcome was reported in one studies of two year-old and another study of one-to-eight-year-old children.(26,47).However, the group of the

two-year-old children were re-examined at six-to-seven years of age. The outcomes of SIPT design copying, postural praxis and manual form perception were significantly associated with their lower full-scale intelligence quotient. (26,27) The review of Bröring et al (32) reported of two studies indicating associations with sensory modulation problems and neurocognitive measures. In five studies, no association was found.

### **Sensory processing deficits and risk factors**

Risk factors associated with sensory processing deficits were reported in 11 studies. The evidence of single factors was diverse and remained weak. Low gestational age correlated with low ITSP, SSP and TSFI scores (31,39,42,43). Low gestational age and length of hospital stay correlated with adverse behavioural and emotional responses in the SP (41) and with low scores in the SIPT finger identification and kinaesthesia (44). Low birth weight (39,43) and days of ventilation were associated with poor SSP scores (43). Low five-minute Apgar score predicted poor SSP and TSFI total scores (43,48). Male sex was associated with adverse reactivity to vestibular stimulation scores (42) and with dysfunctional vestibular, auditory and visual processing scores and low registration (25). Severe white matter injury in brain magnetic resonance imaging correlated with poor ocular-motor control (42). White and grey matter abnormality and surgical closure of the patent ductus arteriosus was associated with sensation seeking behaviour and mildly atypical oral processing (26). Some studies stated no relationship with risk factors such as perinatal conditions (47), gender, brain ultrasound and supplemental oxygen (43).

## DISCUSSION

The amount of research on sensory processing among preterm-born children has increased notably in recent years. Our literature search yielded three systematic reviews and 24 original studies from 11 countries and five continents. Over 50% of the papers were published during the last three years (2016–2018) and 81% during the last six years. The included papers reported on a total of 8,340 participants, age ranging from birth to nine years seven months.

The systematic search identified three clinical assessments and three versions of caregiver questionnaires. The clinical assessments were the Test of Sensory Functions in Infants (TSFI), the Miller Assessment for Preschoolers (MAP) and the Sensory Integration and Praxis Test (SIPT). The caregiver questionnaires were Infant/Toddler Sensory Profile (ITSP), Sensory Profile (SP) and Short Sensory Profile (SSP). The studies of preterm children without major neurodevelopmental impairments showed strong evidence for an elevated risk, compared to their term-born peers, for atypical sensory modulation and sensory-motor processing, and moderate evidence for atypical somatosensory processing. These atypical symptoms persisted at minimum up to nine years of age (27,37,41,44,46,47).

Our systematic review added to the knowledge of sensory processing in preterm children. By identifying two clinical tools, not included in earlier reviews, we gained new evidence of preterm children's performance in somatosensory processing and sensory-based motor problems. The earlier studies have mainly reported on modulation responses of infants and toddlers, observed through the caregiver questionnaire ITSP and the clinical measure TSFI. In addition, the TSFI allowed to observe the ocular-motor control, visual-tactile integration and adaptive motor functions in infants. However, the earlier studies missed assessments that measure direct performance in sensory processing abilities. The identified MAP and SIPT corrected this shortcoming. The MAP that screens performance for the whole range of sensory processing abilities, is only applicable from the age of two years and nine months (51). The

identified SIPT, which is the most comprehensive tool in evaluating of sensory processing abilities (8,19) is applicable from four years of age. Accordingly, there is a need for an assessment of sensory processing performance abilities for infants and toddlers.

Our findings that preterm children have sensory processing problems is in line with previous systematic reviews. Sensory modulation problems were evident in preterm children (30,32). According to review of Mitchell et al (30), modulation problems in multidisciplinary assessments mostly appeared in over-responsivity (30). The review of Bröring et al indicated most problems in low registration (32). Our review identified sensory sensitivity in 7–62% and low registration in 23–71% of the preterm participants. The differences may be explained by the fact that multidisciplinary assessments are not designed to detect sensory processing disorders, especially low registration. Over-responsivity and distractible behaviour may be more easily recognised than non-normal calm appearing behaviour in such studies. Instead, Bröring et al searched modulation problems in studies especially designed for evaluating sensory modulation and, therefore, were able to detect low registration behaviour. Other explanations to the differences may be that individual infants and children naturally differ in sensory processing characteristics and temperament. Moreover, neonatal intensive care procedures, sensory experiences and complications may affect these individuals differently. A suggestion for future research is to study how differences in geographical and cultural influences and impacts of different neonatal intensive care protocols are associated with sensory modulation outcomes.

The research on sensory discrimination in preterm children is scarce. The evidence mainly arose from abnormal visual-tactile integration in infancy (29,31,42) and somatosensory processing at school age, between six and nine years of age (27,37,41,44,46). According to Ayres and Miller et al (8,9,11,13) accurate somatosensory processing is pivotal for

development of motor planning and coordination. Accordingly, behaviours measured by Dunn's Sensory Profile may be related to motor and process skills (54).

Sensory-based motor problems in preterm children appear understudied. Global motor development of preterm children has been widely studied using other measures than those based on sensory processing (1). Studies with the MAP indicated moderate to severe delays in sensory and motor foundations, coordination and combined complex abilities. The study of Leosdottir (38) revealed that the patterns of the MAP scores significantly differed between preterm and term groups. Lane et al (45) compared two groups of preterm children that were allocated according to neurological examination at 18 months of age. At four years of age, the group with neurologically normal scores performed better than the group with lower scores in the neurological examination. However, 25% of children previously allocated as neurologically normal, had severe delays in sensory and motor foundations, coordination and combined complex abilities, using MAP assessment. The risk of delayed abilities was not recognized in the neurological screening at 18 months of age.

Studies with the SIPT revealed visual-motor integration deficits (27,37,46), motor planning deficits (27) and dyspraxia (37). The SIPT, being the most comprehensive tool for detecting sensory integration and praxis difficulties, was only partially applied in the studies. The reason for the sparse use of the SIPT might be its requirement for clinical therapist training in the sensory integration treatment and the SIPT administration. In addition, the scoring of the SIPT is laborious and costly, compared to the other assessments in our review. (8,19)

Regulatory, perceptual, sensory and motor abilities, all crucial aspects of sensory processing, contribute to the development of adequate body schema, motor coordination and motor planning. They further promote adaptive responses in behaviour, facilitate learning and participation. (8,9) Preterm birth, however, may compromise this developmental process. Bart et al (31) investigated differences in participation and sensory modulation between one-year-

old late preterm infants and their term-born controls. The preterm-born toddlers scored worse than the controls on sensory modulation. They indicated significantly more dysfunction in two domains of the ITSP, in auditory and oral processing, and in all subdomains of the TSFI. They showed significantly less involvement in daily activities, play and social participation, compared to term-born children. Moreover, the decreased participation significantly reduced parent satisfaction. Leisure activities and playfulness are positive way to help children with sensory processing deficits to gain perceptual, motor and process skills, and nonetheless, self-confidence. (11,31,55).

Only two studies reported on sensory integration intervention effects. They resulted in significant positive effects on sensory processing. The studies controlled the treatment design with Ayres Sensory Integration Fidelity measure (28,34). It is a recommended tool to conceptualise sensory integration treatments and to strengthen the reliability of the intended outcomes for research (56,57). However, both studies omitted adjustments for possible confounders, which decreases their generalizability (28,34).

The studies indicate that screening and follow-up for sensory processing disorders in preterm-born children should continue after infancy at least up to school age. Early revealing of sensory-based motor performance problems might guide intervention strategies to improve the development of postural control, coordination, motor planning and execution. No longitudinal studies exist in efficiency of sensory integration-based therapy or guidance intervals along childhood years. It is assumable that during years of growing, these children at risk and their families require and benefit from some additional support at some points of challenges in development.

There were some limitations in this systematic review. It mainly reported on positive findings of sensory processing deficits and only some negative findings, which gives rise to a risk of publication bias. No grey literature was searched due to the aim of searching all current, peer-

reviewed and electronically available publications. The included twenty-seven studies might have permitted a meta-analysis, but the variety of study methods and reporting styles made further analyses difficult. The main findings came from the Sensory Profile questionnaires that reflect parent's observations on the child's behaviour, not direct participation of the child. The strength of our review was its discovery of new aspects of sensory processing in studies of preterm children. The MAP and SIPT tests revealed aspects of body-related somatosensory and sensory-based motor processing in preterm children, especially at school age, which have remained unreported in earlier reviews.

The future systematic reviews on the topic are recommended to compare the possible geographical differences of sensory processing disorders and the relationships in the procedures of the neonatal intensive care. However, this would require a growing body of research on sensory processing in preterm infants and children.

## **CONCLUSION**

This systematic review led to strong evidence of preterm infants' elevated risk for dysfunction in sensory modulation and sensory-motor processing. Additionally, the review found moderate evidence for dysfunction in somatosensory processing. The evidence and causality are yet to be specified. A larger body of evidence is needed on the effects of sensory integration treatment for preterm children. Sensory processing deficits negatively affect daily participation and interaction of preterm children and their families, causing disadvantage in mental, social and neurocognitive development. (6,29,31,37,49,58,59). Therefore, routine screening and following of sensory processing development are needed. Treatment intervals and consultations for parents in sensory processing are pivotal in supporting preterm children in their neurocognitive development from infancy through to school age (28,34,38,41-48).

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**ABBREVIATIONS**

BSID = Bayley Scales of Infant and Toddler Development

ITSP = Infant Toddler Sensory Profile

MAP = Miller Assessment for Preschoolers PT = Preterm

SD = Standard deviation

SIPT = Sensory Integration and Praxis Test

SP = Sensory Profile

SSP = Short Sensory Profile

TSFI = Test of Sensory Functions in Infants

## CONFLICT OF INTEREST

The authors have no conflict of interest to declare.

## FINANCE

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## References

- (1) de Kieviet JF, Piek JP, Aarnoudse-Moens CS, Oosterlaan J. Motor development in very preterm and very low-birth-weight children from birth to adolescence: a meta-analysis. *JAMA* 2009 Nov 25;302(20):2235-2242.
- (2) Geldof CJ, van Wassenae AG, de Kieviet JF, Kok JH, Oosterlaan J. Visual perception and visual-motor integration in very preterm and/or very low birth weight children: a meta-analysis. *Res Dev Disabil* 2012 Mar-Apr;33(2):726-736.
- (3) Aarnoudse-Moens CS, Weisglas-Kuperus N, van Goudoever JB, Oosterlaan J. Meta-analysis of neurobehavioral outcomes in very preterm and/or very low birth weight children. *Pediatrics* 2009 Aug;124(2):717-728.
- (4) Arpi E, Ferrari F. Preterm birth and behaviour problems in infants and preschool-age children: a review of the recent literature. *Dev Med Child Neurol* 2013 Sep;55(9):788-796.
- (5) Als H, Duffy FH, McAnulty GB, Rivkin MJ, Vajapeyam S, Mulkern RV, et al. Early experience alters brain function and structure. *Pediatrics* 2004 Apr;113(4):846-857.
- (6) Maitre NL, Key AP, Chorna OD, Slaughter JC, Matusz PJ, Wallace MT, et al. The Dual Nature of Early-Life Experience on Somatosensory Processing in the Human Infant Brain. *Curr Biol* 2017 Apr 3;27(7):1048-1054.
- (7) Brummelte S, Grunau RE, Chau V, Poskitt KJ, Brant R, Vinall J, et al. Procedural pain and brain development in premature newborns. *Ann Neurol* 2012 Mar;71(3):385-396.
- (8) Ayres A.J. *Sensory Integration and Praxis Test (SIPT) Manual*. Los Angeles, USA: Western Psychology Services WPS; 1989.

- (9) Miller LJ, Nielsen DM, Schoen SA, Brett-Green BA. Perspectives on sensory processing disorder: a call for translational research. *Front Integr Neurosci* 2009 Sep 30;3:22.
- (10) Dunn W. The sensations of everyday life: empirical, theoretical, and pragmatic considerations. *Am J Occup Ther* 2001 Nov-Dec;55(6):608-620.
- (11) Ayres AJ. *Sensory Integration and the Child*, 25th anniversary edition. 7th, November 2016 ed. USA: WPS; 2005.
- (12) Dunn W. *Infant/ Toddler Sensory Profile. Manual*. USA: The Psychological Corporation; 2002.
- (13) Miller LJ, Anzalone ME, Lane SJ, Cermak SA, Osten ET. Concept evolution in sensory integration: a proposed nosology for diagnosis. *Am J Occup Ther* 2007 Mar-Apr;61(2):135-140.
- (14) Schaaf RC. Creating Evidence for Practice Using Data-Driven Decision Making. *Am J Occup Ther* 2015 Mar-Apr;69(2):6902360010p1-6.
- (15) Schaaf RC, Burke JP, Cohn E, May-Benson TA, Schoen SA, Roley SS, et al. State of measurement in occupational therapy using sensory integration. *Am J Occup Ther* 2014 Sep-Oct;68(5):e149-53.
- (16) Cochrane. PICO: Formulate an Answerable Question. 2017; Available at: <http://linkeddata.cochrane.org/pico-ontology>. Accessed 6/25, 2017.
- (17) Williamson GG, Anzalone ME. *Sensory integration and self-regulation in infants and toddlers : helping very young children interact with their environment*. Washington, DC: Zero to Three; 2001.
- (18) Eeles AL, Spittle AJ, Anderson PJ, Brown N, Lee KJ, Boyd RN, et al. Assessments of sensory processing in infants: a systematic review. *Dev Med Child Neurol* 2013 Apr;55(4):314-326.

- (19) Jorquera-Cabrera S, Romero-Ayuso D, Rodriguez-Gil G, Trivino-Juarez JM. Assessment of Sensory Processing Characteristics in Children between 3 and 11 Years Old: A Systematic Review. *Front Pediatr* 2017 Mar 30;5:57.
- (20) Stewart LA, Clarke M, Rovers M, Riley RD, Simmonds M, Stewart G, et al. Preferred Reporting Items for Systematic Review and Meta-Analyses of individual participant data: the PRISMA-IPD Statement. *JAMA* 2015 Apr 28;313(16):1657-1665.
- (21) Schulz KF, Altman DG, Moher D, CONSORT Group. CONSORT 2010 statement: updated guidelines for reporting parallel group randomized trials. *Obstet Gynecol* 2010 May;115(5):1063-1070.
- (22) von Elm E, Altman DG, Egger M, Pocock SJ, Gotsche PC, Vandenbroucke JP, et al. The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) Statement: guidelines for reporting observational studies. *Int J Surg* 2014 Dec;12(12):1495-1499.
- (23) Cabral TI, da Silva LG, Martinez CM, Tudella E. Analysis of sensory processing in preterm infants. *Early Hum Dev* 2016 Aug 10;103:77-81.
- (24) Eeles AL, Anderson PJ, Brown NC, Lee KJ, Boyd RN, Spittle AJ, et al. Sensory profiles obtained from parental reports correlate with independent assessments of development in very preterm children at 2 years of age. *Early Hum Dev* 2013 Dec;89(12):1075-1080.
- (25) Eeles AL, Anderson PJ, Brown NC, Lee KJ, Boyd RN, Spittle AJ, et al. Sensory profiles of children born < 30 weeks' gestation at 2 years of age and their environmental and biological predictors. *Early Hum Dev* 2013 Sep;89(9):727-732.
- (26) Rahkonen P, Lano A, Pesonen AK, Heinonen K, Raikkonen K, Vanhatalo S, et al. Atypical sensory processing is common in extremely low gestational age children. *Acta Paediatr* 2015 May;104(5):522-528.

- (27) Lonnerberg P, Niutanen U, Parham LD, Wolford E, Andersson S, Metsaranta M, et al. Sensory-motor performance in seven-year-old children born extremely preterm. *Early Hum Dev* 2018 May;120:10-16.
- (28) Lecuona E, Van Jaarsveld A, Raubenheimer J, Van Heerden R. Sensory integration intervention and the development of the premature infant: A controlled trial. *S Afr Med J* 2017 Oct 31;107(11):976-982.
- (29) Lecuona E, van Jaarsveld A, van Heerden R, Raubenheimer J. The developmental status and prevalence of sensory integration difficulties in premature infants in a tertiary hospital in Bloemfontein, South Africa. *S Afr J Occup Ther* 2016;46(1):15-19.
- (30) Mitchell A, Moore EM, Roberts EJ, Hachtel KW, Brown MS. Sensory Processing Disorder in Children Ages Birth–3 Years Born Prematurely: A Systematic Review. *Am J Occup Ther* 2015 Jan;69(1):1-11 11p.
- (31) Bart O, Shayevits S, Gabis LV, Morag I. Prediction of participation and sensory modulation of late preterm infants at 12 months: a prospective study. *Res Dev Disabil* 2011 Nov-Dec;32(6):2732-2738.
- (32) Broring T, Oostrom KJ, Lafeber HN, Jansma EP, Oosterlaan J. Sensory modulation in preterm children: Theoretical perspective and systematic review. *PLoS One* 2017 Feb 9;12(2):e0170828.
- (33) Machado ACCP, Oliveira SR, Magalhaes LC, Miranda DM, Bouzada MCF. Sensory Processing during Childhood in Preterm Infants: a Systematic Review. *Rev Paul Pediatr* 2017 Jan-Mar;35(1):92-101.
- (34) Pekcetin S, Aki E, Ustunyurt Z, Kayihan H. The Efficiency of Sensory Integration Interventions in Preterm Infants. *Percept Mot Skills* 2016 Aug 10.

- (35) Verkerk G, Jeukens-Visser M, Koldewijn K, van Wassenaer A, Houtzager B, Kok J, et al. Infant behavioral assessment and intervention program in very low birth weight infants improves independency in mobility at preschool age. *J Pediatr* 2011 Dec;159(6):933-8.e1.
- (36) Boone KM, Gracious B, Klebanoff MA, Rogers LK, Rausch J, Coury DL, et al. Omega-3 and -6 fatty acid supplementation and sensory processing in toddlers with ASD symptomology born preterm: A randomized controlled trial. *Early Hum Dev* 2017 Dec;115:64-70.
- (37) Goyen TA, Lui K, Hummell J. Sensorimotor skills associated with motor dysfunction in children born extremely preterm. *Early Hum Dev* 2011 Jul;87(7):489-493.
- (38) Leosdottir T, Egilson ST, Georgsdottir I. Performance on extremely low birthweight children at 5 years of age on the Miller Assessment for Preschoolers. *Phys Occup Ther Pediatr* 2006;25(4):59-72.
- (39) Adams JN, Feldman HM, Huffman LC, Loe IM. Sensory processing in preterm preschoolers and its association with executive function. *Early Hum Dev* 2015 Mar;91(3):227-233.
- (40) Beranova S, Stoklasa J, Dudova I, Markova D, Kasparova M, Zemankova J, et al. A possible role of the Infant/Toddler Sensory Profile in screening for autism. *Neuropsychiatr Dis Treat* 2017 Jan 23;13:191-200.
- (41) Broring T, Konigs M, Oostrom KJ, Lafeber HN, Brugman A, Oosterlaan J. Sensory processing difficulties in school-age children born very preterm: An exploratory study. *Early Hum Dev* 2018 Feb;117:22-31.
- (42) Chorna O, Solomon JE, Slaughter JC, Stark AR, Maitre NL. Abnormal sensory reactivity in preterm infants during the first year correlates with adverse neurodevelopmental outcomes at 2 years of age. *Arch Dis Child Fetal Neonatal Ed* 2014 Nov;99(6):F475-9.



- (43) Crozier SC, Goodson JZ, Mackay ML, Synnes AR, Grunau RE, Miller SP, et al. Sensory Processing Patterns in Children Born Very Preterm. *Am J Occup Ther* 2016 Jan-Feb;70(1):7001220050p1-7.
- (44) DeMaio-Feldman D. Somatosensory processing abilities of very low-birth weight infants at school age. *Am J Occup Ther* 1994 Jul;48(7):639-645.
- (45) Lane SJ, Attanasio CS, Huselid RF. Prediction of preschool sensory and motor performance by 18-month neurologic scores among children born prematurely. *Am J Occup Ther* 1994 May;48(5):391-396.
- (46) Feder KP, Majnemer A, Bourbonnais D, Platt R, Blayney M, Synnes A. Handwriting performance in preterm children compared with term peers at age 6 to 7 years. *Dev Med Child Neurol* 2005;47(3):163-170.
- (47) Wickremasinghe AC, Rogers EE, Johnson BC, Shen A, Barkovich AJ, Marco EJ. Children born prematurely have atypical sensory profiles. *J Perinatol* 2013 Aug;33(8):631-635.
- (48) Celik HI, Elbasan B, Gucuyener K, Kayihan H, Huri M. Investigation of the Relationship Between Sensory Processing and Motor Development in Preterm Infants. *Am J Occup Ther* 2018 Jan/Feb;72(1):7201195020p1-7201195020p7.
- (49) Buffone FRRC, Eickman SH, Lima MdC. Sensory processing and cognitive development of preterm and full term infants. *Cad Ter Ocup UFSCar* 2016 Aug.;24(4):695-703.
- (50) DeGangi GA, Greenspan SI. *Test of Sensory Functions in Infant. Manual.* USA: Western Psychological Services; 1989.
- (51) Miller LJ. *Miller Assessment for Preschoolers Manual. Revised ed.:* Pearson Psychological Corporation; 1988.

- (52) Bundy AC, Lane SJ, Murray EA. Sensory Integration Theory and Practice. 2nd ed. Philadelphia: F. A. Davis Company; 2002.
- (53) Dunn W. Sensory Profile Caregiver Questionnaire. Manual. USA: The Psychological Corporation; 1999.
- (54) White BP, Mulligan S, Merrill K, Wright J. An examination of the relationships between motor and process skills and scores on the sensory profile. *Am J Occup Ther* 2007 Mar-Apr;61(2):154-160.
- (55) Cosbey J, Johnston SS, Dunn ML. Sensory processing disorders and social participation. *Am J Occup Ther* 2010 May-Jun;64(3):462-473.
- (56) Parham LD, Cohn ES, Spitzer S, Koomar JA, Miller LJ, Burke JP, et al. Fidelity in sensory integration intervention research. *Am J Occup Ther* 2007 Mar-Apr;61(2):216-227.
- (57) Schaaf RC, Mailloux Z. Clinician's Guide for Implementing Ayres Sensory Integration: Promoting Participation for Children with Autism. Bethesda, MD: American Occupational Therapy Association; 2015.
- (58) Gourley L, Wind C, Henninger EM, Chinitz S. Sensory Processing Difficulties, Behavioral Problems, and Parental Stress in a Clinical Population of Young Children. *J Child Fam Stud* 2013 Oct 1;22(7):912-921.
- (59) Dar R, Kahn DT, Carmeli R. The relationship between sensory processing, childhood rituals and obsessive-compulsive symptoms. *J Behav Ther Exp Psychiatry* 2012 Mar;43(1):679-684.

## Tables

Table 1. The systematic reviews that were included

Author, Year. Country. Study design, Level of Evidence.	N (Study population) n (studies included); GA weeks (wk), birth weight (g) age at study, years (a), months (mo).	Aim/Objectives	Assess ments	Main results.  Studies in common with the current review.
Mitchell et al 2015. USA.  Systematic review, Level I.	N=2584, n=45 studies;  GA 22–37 or 45– 2865g;  Age birth–3a.	Sensory processing disorder patterns in multiprofessional studies of preterm children.	ITSP, TSFI	295 findings related to SPD:130 (44%) positive; 165 (56%) negative; majority related to sensory modulation disorder, most prevalently to sensory over-responsivity.  One study was in common with the current review.
Bröring et al 2017 (52). Netherlands.	PT N=1259, T N=542, n=18 studies;	Sensory modulation problems in preterm children, associations to	ITSP, SSP, SP,	Sensory modulation problems reported in 15 studies in the preterm population and in three studies of general population samples analysing gestational age. Some relationship evidenced between

Systematic review, Level I.	GA (PT) 5x (22–37w), 7x (<1500g), 1x ≤ 28w;	risk factors, behavioural and neurocognitive problems.	TSFI, Sensory Rating Scale.	sensory modulation disorder and white matter injury and length of stay in neonatal intensive care unit. Nature and severity of SMP differed widely and may affect neurocognitive and behavioural sequelae.  11 studies in common with the current review.
Machado et al 2017 (33). Brazil.  Systematic Review, Level I.	N=N/A, n=8 studies  GA 75% <34;  Age <2a in six studies.	Evidence of grounded and quality evidence concerning the sensory processing in preterm infants during childhood	ITSP, SSP, TSFI,	Preterm birth was considered a risk factor for SPD. Gestational age, male gender and white matter lesions were associated with SPD; negative effects on motor, cognitive and language development. The signals for SPD can be identified early, which is crucial for early treatment and improvement of SPD for the benefit of development.  All eight studies in common with the current review.

Table 1 abbreviations: GA = Gestational age, ITSP = Infant/Toddler Sensory Profile, PT = Preterm-born, SPD = Sensory processing disorder, SRS = Sensory Rating Scale, T = Term-born, TSFI = Test of Sensory Functions in Infant.

**List of figures**

Figure 1. Systematic literature search flow diagram.

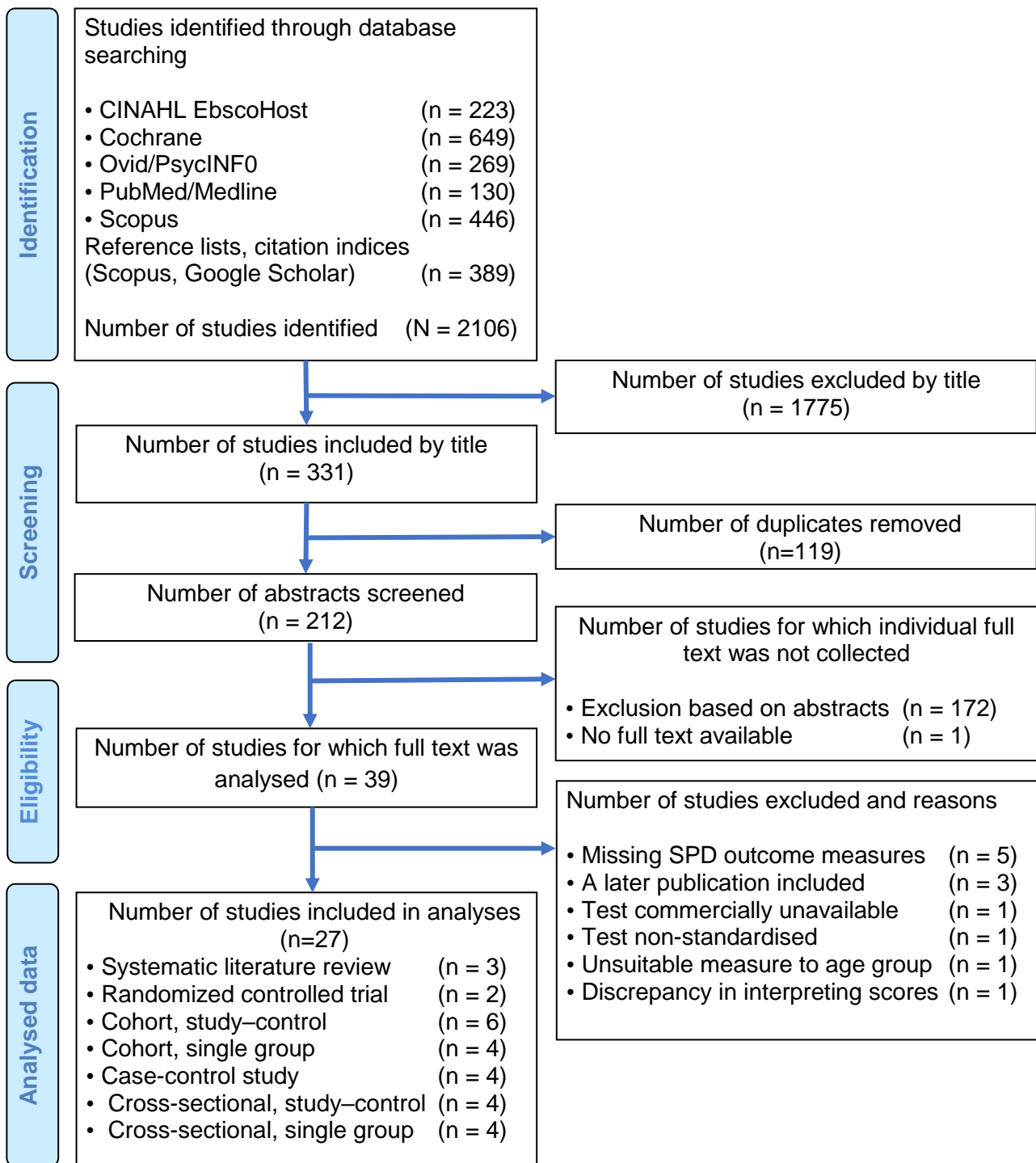


Figure 1. Systematic literature search flow diagram. Reproduced and adapted from PRISMA-IPD Flow Diagram (17). Abbreviation SPD = Sensory Processing Disorder.