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REHABILITATION OF APRAXIA IN ADULTS AND CHILDREN

Caroline M. van Heugten and Chantal Geusgens

Apraxia in adults

Apraxia is one of the more classical neuropsychological deficits. The term apraxia was first used by Steinthal in 1871 and Geschwind proposed the following definition in 1975: 'the apraxias are disorders of the execution of learned movements which cannot be accounted for either by weakness in coordination or sensory loss, nor by incomprehension of or inattention to commands' (Geschwind, 1975, p. 188). Hugo Karl Liepmann (1920) was mainly responsible for the recognition of the errors made by patients and the means by which they were elicited. Since Steinthal much has been written about apraxia, and for clinicians it is one of the most common consequences of stroke. However, it still remains a difficult concept because there is not one accepted taxonomy or classification of the different forms, there is no consensus on assessment and scoring methods for tests and only a few studies have been conducted investigating the effectiveness of apraxia treatment (van Heugten, 2002). Scientific literature generally distinguishes two types of motor apraxia, which are sometimes labelled the two classic forms of motor or limb apraxia: ideational and ideomotor apraxia. Both types of apraxia may cause severe disabilities in the performance of activities of daily living (ADL), resulting in a negative impact on everyday life.

In addition to forms of apraxia associated with the limbs, other body parts can be affected, resulting in disorders such as buccofacial or oral apraxia. And some suggest describing apraxia in terms of the activity that is disturbed, such as walking apraxia or dressing apraxia.

Occurrence of limb apraxia

In adults, apraxia is often apparent after left hemisphere stroke, but it is also seen in patients with Alzheimer's disease (Ochipa, Gonzalez-Rothi and Heilman, 1992), Huntington's disease (Shelton and Knopman, 1991), and it is one of the symptoms of corticobasal degeneration (Jacobs et al., 1999; Leiguarda et al., 1994). Recently, Rapačić and colleagues (2014) showed that limb apraxia can also be present in patients with multiple sclerosis.

Studies into the prevalence of apraxia after stroke show percentages ranging from 28 per cent to 57 per cent in left hemisphere stroke patients, depending on the assessment instrument used, time since injury and setting (van Heugten, 2002). In a cross-sectional cohort study involving 100 patients

with a first stroke admitted to a rehabilitation centre, Zwinkels et al. (2004) found that 25.3 per cent of the total group of stroke patients and 51.3 per cent of the left hemisphere stroke patients scored below the cut-off on an apraxia test in the first six weeks post stroke (34–55 days).

Although no single taxonomy or classification of different forms of apraxia exists, two forms are usually reported as the most typical and most studied forms of apraxia: ideational and ideomotor apraxia (van Heugten, 2002). A patient with ideational apraxia does not know what to do because the idea or concept of the motor act is lacking. The sorts of errors observed in ideational apraxia are behavioural omissions, mislocation or misuse of objects or sequence mistakes. In ideomotor apraxia the idea or plan of action is not impaired (i.e. the patient knows what to do) but the implementation into a proper action is disturbed (i.e. the patient does not know how to do it). The patient may use body parts as objects, show spatial orientation problems, use inappropriate hand postures, or show perseverative and content errors. The most striking error is making mistakes when performing an action on command but performing the same action correctly in a natural setting.

Assessment of limb apraxia

Many different task demands and quantitative and qualitative scoring methods exist (Tate and McDonald, 1995). van Heugten and colleagues (1999; Zwinkels et al., 2004) developed an apraxia test for stroke patients which is often used in clinical practice. Recently the test was validated in patients with mild cognitive impairment and Alzheimer's disease (Smits et al., 2014). This short and simple test consists of two subtests. The first subtest was designed to evaluate the use of objects, thereby assessing ideational apraxia. Three sets of objects are presented to the patient with the same instruction: 'Show me how you would use...'. The second subtest involves the assessment of the ability to imitate gestures, aiming at assessing ideomotor apraxia. This part of the test consists of six gestures, which have to be imitated immediately upon demonstration. The apraxia test has good internal consistency and diagnostic value (van Heugten et al., 1999) and has been shown to be a reliable instrument (Zwinkels et al., 2004). This standardised test can be used in combination with observation techniques in ADL performance (Goldenberg and Hagmann, 1998a; van Heugten et al., 1998).

Recovery and treatment of limb apraxia

Although the incidence of apraxia after acquired brain damage is considerable, the literature on recovery and treatment is minimal. This lack of studies can be attributed to the fact that patients with apraxia often seem to be unaware of their deficit and rarely complain, leading to an underdiagnosis of apraxia. Second, many researchers believe that recovery from apraxia is spontaneous and treatment is not necessary; third, some authors believe that apraxia only occurs when performance is requested of patients in testing situations, and that correct behaviour is displayed in natural settings (Maher and Ochipa, 1997). By now, however, there is agreement that apraxia hinders ADL independence (Goldenberg and Hagmann, 1998b; Hanna-Pladdy, Heilman and Foundas, 2003; Unsal-Delialioglu et al., 2008). These results suggest that treatment of apraxia should be part of the overall neurorehabilitation programme after brain damage.

Donkervoort, Dekker and Deelman (2006) studied the course of apraxia and daily life functioning in left hemisphere stroke patients with apraxia. In a prospective cohort study patients showed small improvements in apraxia and medium-sized improvements in ADL functioning from the start of rehabilitation until four months later. About 88 per cent of the patients were still apraxic at week 20. Less improvement in apraxia was observed in initially less severe apraxic patients. Less improvement in ADL functioning was found to be associated with more severe apraxia, a more independent initial ADL score, higher age, impaired motor functioning and longer time between stroke and first assessment.

A Cochrane review has been published that determined which therapeutic interventions are effective for targeting disabilities due to motor apraxia following stroke (West et al., 2008). The literature search covered up to November 2006 and revealed only three trials with a total of 132 participants (Donkervoort et al., 2001; Edmans, Webster and Lincoln, 2000; Smania et al., 2000). The authors of the review conclude on the basis of these three trials that there is insufficient evidence to support or refute the effectiveness of specific interventions for motor apraxia following stroke. Since the study of Edmans et al. (2000) aimed at evaluating the treatment of perceptual problems, it will not be discussed here.

Smania et al. (2000) conducted a randomised controlled trial (RCT) assessing the effectiveness of a rehabilitative training programme for patients with limb apraxia. Thirteen patients with acquired brain injury and limb apraxia (lasting more than two months) due to lesions in the left cerebral hemisphere participated in the study. The study group underwent experimental training for limb apraxia consisting of a behavioural training programme with gesture-production exercises. The control group received conventional treatment for aphasia. Assessments involved neuropsychological tests of aphasia, verbal comprehension, general intelligence, oral apraxia, constructional apraxia and three tests assessing limb praxic function (ideational and ideomotor apraxia and gesture recognition). In addition, errors made during test performance were scored on the basis of video recordings. The patients in the study group achieved a significant improvement of performance and a significant reduction of errors in both ideational and ideomotor apraxia tests. The change in performance was not significant for the control group. The results show the possible effectiveness of a specific training programme for the treatment of limb apraxia.

In a randomized controlled study Donkervoort et al. (2001) determined the efficacy of strategy training in left hemisphere stroke patients with apraxia. A total of 113 left hemisphere stroke patients with apraxia were randomly assigned to two treatment groups: (1) strategy training integrated into usual occupational therapy; and (2) usual occupational therapy only. The primary outcome measure was a standardised ADL observation by a blinded research assistant. Additional ADL measures were used as secondary outcome measures (Barthel ADL index, ADL judgement by occupational therapists and by patients). After eight weeks of treatment, patients who received strategy training (n=43) improved significantly more than patients in the usual treatment group (n=39) on the ADL observations. This reflects a small-to-medium effect (effect size 0.37) of strategy training on ADL functioning. With respect to the secondary outcome measures a medium effect (effect size 0.47) was found on the Barthel ADL index. No beneficial effects of strategy training were found at follow-up five months later.

Secondary analyses were performed on the data of Donkervoort et al. (2001) to examine the transfer of the effects of cognitive strategy training for stroke patients with apraxia from trained to non-trained tasks. The analyses showed that in both treatment groups, the scores on the ADL observations for non-trained tasks improved significantly after eight weeks of training as compared with the baseline score. Change scores of non-trained activities were larger in the strategy training group as compared with the usual treatment group. These results suggest that transfer of training is possible, although further research should confirm these exploratory findings (Geusgens et al., 2006). A further study was carried out specifically to measure the transfer effects of the cognitive strategy training for apraxia (Geusgens et al., 2007). The study showed that patients performed both trained and non-trained tasks at the same level of independence at the rehabilitation centre as at home, indicating transfer of training effects which remained stable over time.

Dovern, Fink and Weiss (2012) formulated recommendations for clinical practice on the basis of three trials (Donkervoort et al., 2001; Smania et al., 2000, 2006). They advise using the gesture training of Smania et al. (2000, 2006) as lasting effects of the training at two months follow-up were found. Cantagallo, Maini and Rumiati (2012) describe the same trials in their review and make a distinction between restorative training (i.e. the gesture training of Smania et al, 2000, 2006) and

compensatory training (i.e. the strategy training of Donkervoort et al., 2001). They conclude that both types of interventions were found to be effective and the choice of treatment method should be based on the goal of the treatment. VanBellingen and Bohlhalter (2011) suggest that this goal is associated with the form of apraxia; they propose that gesture training is mainly meant for ideomotor apraxia while strategy training is most effective for ideational apraxia.

Buxbaum et al. (2008) present other forms of treatment of limb apraxia in their review, not necessarily based on class 1 evidence. They suggest that multiple cues, six-stage task hierarchy (a method that requires the patient to produce target words and signs in various combinations and in concert with the therapist in response to a therapist model or picture elicitation), conductive education and errorless completion and exploration training can be considered. The errorless learning approach was also recommended by Lindsten-McQueen et al. (2014). New treatment methods and techniques are motor imagery (Buxbaum et al., 2008), transcranial magnetic stimulation (TMS) (Bolognini et al., 2015) and assistive technology specific for patients with apraxia (Jean-Baptiste, Russell and Rothstein, 2014).

The assumption that problems in motor planning can result in apraxia has been put forward in the past by both Luria (1980) and Schwartz et al. (1998). This assumption has recently gained interest again, conceptualising apraxia as an executive dysfunction. ‘Affordances’ (Barde, Buxbaum and Moll, 2007) and ‘technical reasoning’ (Randerath et al., 2011) are two concepts related to motor planning that have been shown to be associated with apraxia. The technical reasoning hypothesis claims that people reinterpret the world in terms of abstract principles (i.e. affordances), combine these principles and translate them into a plan of action in order to solve a problem. Thus far, interventions that have been developed based on this hypothesis have specifically been aimed at the evaluation of the theoretical basis of the hypothesis only. Furthermore, up until now no studies have looked into the technical reasoning hypothesis and its role in explaining deficits in naturalistic actions. Whether the hypothesis can explain these deficits remains to be investigated, whereupon new therapeutic methods for apraxia could be developed.

Apraxia of speech after stroke

Acquired apraxia of speech (AOS) is a communication disorder that also can affect stroke patients and is usually treated by speech and language therapists. Speech sounds are made in an erratic way in the absence of muscle weakness. AOS has been a controversial topic in neurological and neuropsychological literature but there is convergent evidence that the left hemisphere is specialised in the control of skilled motor acts across modalities (Square, Roy and Martin, 1997). AOS can be assessed using the Apraxia Battery for Adults – 2 (ABA-2) which can be offered face-to-face or via the internet (Hill et al., 2009).

Sound production treatment is believed to be the treatment of choice for AOS according to some authors (Wambaugh et al., 2013, 2014b). In a multiple baseline design across participants and behaviours with four participants, different intensities and schedules of practice were evaluated. Positive changes in accuracy of articulation for all participants and all methods were shown (Wambaugh et al., 2013). In another study the same authors investigated the differential effects of blocked versus random practice of sound production treatment in six participants (Wambaugh et al., 2014a). Comparable effects of both practice methods were found in terms of increasing the accuracy of articulation but only the random practice version led to maintenance of effects in two participants. The same group also combined Aphasia and Apraxia of Speech Treatment (CAAST) targeted at language and speech production simultaneously, with treatment techniques derived from Response Elaboration Training and Sound Production Treatment (Wambaugh et al., 2014b). Since no unequivocal results of its effects were found, the authors suggest a modification of CAAST needs to be developed and further evaluated.

In a review on treatment of acquired apraxia of speech Ballard et al. (2015) concluded that both articulatory-kinematic and rate/rhythm approaches to AOS treatment show the strongest effects. Earlier, Knollman-Porter (2008) also recommended the use of augmentative/alternative communication devices, intersystemic facilitation/reorganization, constraint-induced therapy and addressing functional communication in support groups outside the therapeutic environment. In a study with two individuals, constraint-induced aphasia therapy (CIAT) seemed to be effective in increasing word retrieval in patients with chronic aphasia with comorbid AOS (Kurland et al., 2012).

Apraxia in children

Dyspraxia is a developmental disorder seen in children and characterised by an impairment of the organization and planning of movement. The condition is believed to be a result of a delay or disjoint in relaying information between the left and right cerebral hemispheres that would otherwise produce smooth, coordinated motor functions.

Children who have childhood apraxia of speech (CAS) find it very difficult to make sounds in the right order, which makes it hard for others to understand them. Communication can thus break down between speaker and listener.

There is no consensus on the diagnostic criteria and underlying mechanisms of CAS but some agreement exists on the domains in which these children have impairments: non-speech oral motor function, motor speech function, speech sounds and structures (i.e. syllable and word shapes), prosody, language, phonemic awareness/metalinguistic skills and literacy (Morgan and Vogel, 2008). The following features across these domains have been shown to have diagnostic validity: (1) inconsistent error production on both consonants and vowels across repeated productions of syllables or words; (2) lengthened and impaired coarticulatory transitions between sounds and syllables; and (3) inappropriate prosody (ASHA, 2007). CAS is typically diagnosed on the basis of expert judgement of perceptual features, but Murray, McCabe and Ballard (2014) recently developed and evaluated a more objective method to distinguish CAS from other speech disorders in children. They found that polysyllabic production accuracy and an oral motor examination that includes diadochokinesis may be sufficient to reliably identify CAS specifically.

Morgan and Vogel (2008) conducted a systematic Cochrane review into studies evaluating treatment for CAS, but they were unable to include any study upon which to formulate recommendations for interventions.

Recently, Murray et al. (2014) also conducted a review into treatment outcomes for these children and they were able to recommend three treatment methods on the basis of single case experimental research: Integral Stimulation/Dynamic Temporal and Tactile Cueing; Rapid Syllable Transition Treatment (ReST); and Integrated Phonological Awareness Intervention. In clinical practice the Nuffield Dyspraxia Programme (3rd edition, NDP3; Williams, 2009) seems to be used often but, to our knowledge, evidence of its effectiveness is lacking.

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