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Autism Spectrum Disorders – What is the Problem?

The quest for the ‘holy grail’ of developing effective interventions to help people who have Autism Spectrum Disorders (ASD) has brought forth the usual clutch of quacks, charlatans and self-publicists, but mingled with the money-seeking snake-oil sellers are highly committed and professional practitioners and researchers, whose efforts have produced important innovations in the treatment of ASD. The many debates that have been produced in this quest for interventions have generated much heat, some little light, certainly much controversy and a great deal of confusion regarding what interventions are actually effective. The current work aims to untangle some of the mass of data regarding interventions for ASD and to provide some suggestions about the circumstances under which particular approaches may offer some help to the key people involved; that is, those affected by ASD, both directly themselves and through their close connection to somebody who is directly affected.

Autism Spectrum Disorders

The precise definition of ASD is and has been, in flux for 50 or more years. Recently, the diagnostic criteria for labelling the disorder have undergone yet another shift, from those contained in the previous Diagnostic and Statistical Manual (DSM) IV-TR, to those outlined in the new DSM-5. The nature of these criteria and some of the implications of these changes, will be discussed in Chapter 2, but it is safe to assert that ASD presents a range of complex and interacting behaviours and deficits that appear early in a person’s development and persist, sometimes in differing forms, throughout that person’s life impacting a wide range of their functioning. These characteristics mark out ASD from other more specific forms of psychological problems associated with childhood and make complex any intervention.
These issues will be covered later in terms of developing a characterization of the nature of ASD that presents for treatment (see Chapter 3), but, at this point, it is worth noting a few salient generalizations about the condition. For many, it is the types and quality of social and emotional interactions engaged in by people with ASD, as well as behavioural inflexibility and highly focused attention on aspects of their environment, that are central to its characterization. These key aspects of the disorder typically present together, making this a wide-ranging ‘syndrome’, with myriad potential causes – genetic, obstetric, physiological and even parenting. In itself, a disorder with multiple causations is often difficult to treat, but these primary problems are often connected with secondary or co-morbid problems for the individual with ASD, such as high levels of challenging behaviour, social isolation, anxiety and depression. The degree to which such secondary symptoms are present differs between each individual, both in the types of symptom present and also in their severity, making ASD a ‘spectrum’. Of course, these individual symptoms are not unique to ASD and it has been suggested that all share some of these ASD-typical traits – that is, there is a ‘broad autistic phenotype’ in the population. This may be true, but it is the presence of all problems together that characterize the disorder as ASD – a syndrome with a spectrum.

It is certainly the case that ASD cannot be recognized physically (despite some recent claims to contrary) and it is also true to say that ASD is found across the entire range of people in society. This consideration raises another issue: given this broad range of people with ASD and given the potential presence of these characteristics in us all, is it a problem to have ASD? This question touches on another, increasingly important, aspect of the theoretical discussion that it is important to raise early: that is, whether the ‘core’ elements of ASD are always to be regarded as ‘impairments’ that are in need of ‘treatment’. Indeed, some have even queried whether ASD should be ‘treated’ at all. It has been suggested that some of the behavioural and cognitive differences between people with and without ASD can sometimes confer an enhanced ability to the individual with ASD in some areas of performance (e.g. in terms of attention-based tasks requiring focus on detail) – albeit an ability at the expense of flexibility as circumstances change. However, it is equally important to note that there is by no means a consensus on this issue, either in the academic literature or among those individuals with an ASD themselves. The task of answering this question is made doubly difficult given the variation in behaviours and needs of people with ASD.

In framing a focused understanding of the scope of the present work, it may, thus, help to consider two questions related to these issues: (i) is having ASD a problem and (ii) if so, how and for whom? The first question is rather easier to answer from the perspective of a work on intervention, than from the perspective of a more theoretically based tome – quite simply, there are many, perhaps countless, individuals with ASD and their families who desperately need help – therefore, it is a problem for those people with ASD and for those people who their lives they touch, when it is a problem for them. To deny the existence of such a problem is wrongheaded and, to deny the treatment, is cruel, which is worse. As usual, there are very important provisos to add to this straightforward answer: not all people with ASD do require help and not all people with ASD require help with everything. Thus, interventions for ASD must be flexible enough to accommodate these considerations; that is to say, they must recognize that which was noted previously – that ASD is a spectrum and not a unitary disorder and that interventions will need to be tailored to suit different situations with
different individuals. Together, these guiding tenets impact considerably on the focus to be taken regarding understanding the nature of ASD and the development of its interventions.

To answer the second question posed previously – ‘whose problem is ASD?’ – it can be said that it is not just a problem for the person with ASD, but it is a problem for and of, the community in which that person with ASD resides. The current text takes the view that an important contrast is made between the within-person disability model, which attempts to identify immutable causal structures within the person with ASD and a social-environmental approach, which suggest mutable functional connections between the person’s environment and their behaviours (see Chapter 3). Far too much attention and too many resources have been placed on the former view at the expense of the latter, especially when it is seen that almost all scientifically supportable interventions stem from the latter conceptualization of ASD.

Treating ASD

If ASD is to be considered as a problem to be treated, then it is worth saying a few brief words about the nature of ‘treatment’. There are a vast range of interventions now championed as being effective for ASD and the current work surveys those on which significant serious research has been conducted. These will be described throughout the text, but it is taken as a truth that it is possible to say that there are some interventions that are better than others and also that it is not in anybody’s best interests to accept the claim that any intervention is good merely if is offered and conducted well (Jordan et al., 1998). Too many times has it been suggested at conferences and gatherings of professionals that anything goes in this field. A consequence of the assumed need for a clear evidence-base tilts the approach of this book toward adopting a ‘medical model’ of evidence assessment – an approach that has met with considerable success in other areas of treatment – hence, the use of the term ‘treatment’ interchangeably with ‘intervention’ and in preference to currently more trendy re-labelling of ‘treatment’.

With this belief, the current work will take as ‘treatments’ those interventions that have made it out of the laboratory. This focus has the result of acting rather like a filter, determining which, out of the many potential interventions, will be discussed. Many interventions that have been developed purely in the laboratory, on the basis of particular theories, will not be addressed and neither will those that have been developed on the whim of individual practitioners. However, there are still many intervention programmes that pass this filter. Some of the history of interventions is presented in Chapter 3, where it can be seen that interventions for ASD have a longer history than might be expected. However, from the early 1970s onwards, larger numbers of interventions have emerged. For example, although Applied Behaviour Analysis (ABA) programmes (e.g. Lovaas, 1987; see Chapter 4) are often commented upon in the scientific and popular literature, approaches such as the Treatment and Education of Autistic and Related Communication Handicapped Children (Schopler & Reichler, 1971) and the Denver Health Sciences Programme (Rogers et al., 1986), emerged at least as early as ABA and the last few years have seen the development and proliferation of many more interventions for ASD. Many of these treatment types are
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The current filter applied to the selection of particular interventions to be considered also has a number of further components. The current survey addresses only those interventions that accept, as their central focus, the individual affected by an ASD. This sounds like a rather odd or obvious thing to state, but too many times the main focus of an intervention has been to support the underlying theory or philosophical beliefs from which the intervention was developed. To this end, the evidence, as it relates to the impact of interventions on the functioning of people with ASD and not how it supports this or that theoretical view of ASD, will be utilized. This approach reinforces two aspects already introduced into the discussion – in addition to the necessity of obtaining good evidence regarding the effectiveness of interventions, emphasis needs to be placed: firstly, on the functionality of the interventions – they must affect change, rather than adhere to any particular theory and secondly, on the essential individuality of people with ASD – the interventions must be adaptable to one of the most individual group of people. Needless to say, this will not be easy.

One way of summarizing these various aspects of the support to be provided by any good intervention is that they are all concerned with producing a better match mentioned in Figure 1.1, which presents a schematic representation of the interventions that several reviews have suggested as important (Odom et al., 2010; Ospina et al., 2008; Stahmer et al., 2005; Vismara & Rogers, 2010).

Figure 1.1  A schematic representation of a number of individual types of intervention programmes and their broad characterizations (based on Opsina et al., 2008).
between the abilities of people with ASD and the environment in which they are placed. This objective is well reflected in a quote from a person with Asperger’s syndrome reported by Baron-Cohen (2003, p. 180): ‘We are fine if you put us in the right environment. When the person with Asperger’s Syndrome and the environment match, the problem goes away… When they do not match, we seem disabled.’ That is, interventions need to be targeted at adapting the environment to the person in order to more effectively contact that person’s behaviours, develop their skills to their full potential and enhance their chances of success. It is argued in Chapter 3 that this form of treatment philosophy has produced the greatest number of effective interventions for ASD. Thus, there is no tension between such a person-centred psycho-social approach and a hard-core medical model of assessment, as these approaches seem to offer the best way to deal with the size and costs (both human and economic) of the problem posed by ASD.

The Size of the ASD Problem

The importance of developing interventions for ASD can be placed in perspective by a consideration of the scale of the problem (see Williams et al., 2006, for a thorough review). The population prevalence of ASD is generally estimated to be between 0.9 and 1.5% (Baird et al., 2006; Baron-Cohen et al., 2009; Brugha et al., 2009). This sort of estimate has been widely accepted for some time, however, Kim et al. (2011) suggested that a remarkably high number of children may display signs of ASD in diagnostic tests, but do not receive a diagnosis, noting prevalence rates of over 3.5% in males and 1.5% in females. Thus, prevalence estimates vary but, as shall be seen, these estimates depend on a number of factors, such as the definition of the disorder that is employed and the method used to calculate the prevalence.

It is a truism to say that the prevalence of ASD is whatever studies of its prevalence say that it is, but this truism encapsulates an important fact: there is no definitive way of knowing what the prevalence of ASD really is, as all estimates carry with them their own definitions of the disorder. Of course, that is not a particularly helpful thing to point out, as, in developing a view regarding the most appropriate forms of intervention for ASD, of central concern to most service providers are the numbers of individuals who are likely to present with these problems.Crudely put, services can only be delivered if there is the money to pay for those services and those financial calculations will depend on the numbers of people likely to present for treatment. Thus, knowing the prevalence of ASD is important for intervention planning and development.

An ASD epidemic?

As noted previously, most contemporary reports place the prevalence of ASD at about 1% of the population (e.g. Baird et al., 2006; Baron-Cohen et al., 2009). However, an issue that has exercised much current thinking concerning prevalence is that the numbers of people with ASD appear to be rising over time (see Maenner & Durkin, 2010; Williams et al., 2006 and Figure 1.2). Initial estimates, given about 40 years ago, placed ASD prevalence at about 4.5 in every 10 000 (Lotter, 1966), which increased to about 20 in every 10 000 around 30 years ago (Wing & Gould, 1979).
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The prevalence rate was placed at 60 in every 10,000 about 10 years ago (Bertrand et al., 2001; Chakraborti & Fombonne, 2001) and more recent estimates have placed this at around 100 in every 10,000 (Baird et al., 2006; Baron-Cohen et al., 2009). This rise in the estimated rates of ASD has sparked concerns over the causes of the putative rise and concerns about the existence of an ‘epidemic’. However, as shall be seen next, it is unlikely that these figures show anything of the kind (see also Fombonne, 2003; Williams et al., 2006, for similar discussions). Much of the variance in these data can be attributed to differences in the criteria used to define ASD, which have become much less strict and less narrow over time (see Baron-Cohen et al., 2009; Lotter, 1966) and to variation in the methods used to arrive at these prevalence estimates.

**Definitional issues**

The criteria used to define ASD are clearly critical in impacting estimates of prevalence. In extensive reviews of the literature, both Fombonne (2003) and Williams et al. (2006) have noted that this definitional factor shows the highest degree of association with noted prevalence rates and that this factor is also completely confounded with the time at which the rate was calculated. For example, initial prevalence estimates (e.g. Lotter, 1966), using a set of criteria derived from Kanner (1943), noted a rate of only 4.5 in every 10,000; subsequent rates using the DSM-III criteria tended to produce estimates of around 20 in every 10,000 (see Williams et al., 2006) and estimates based on the DSM-IV-TR vary between 60 in every 10,000 (Bertrand et al., 2001) and 116 in every 10,000 (Baird et al., 2006). The overall impact of these changes has been to increase estimates of the rates of ASD in the population (see Figure 1.2).

**Figure 1.2** Estimation of prevalence rates for typical Autism (Autism) and all Autism Spectrum Disorders (ASD) across the last 40 years. The names refer to the first author of each report (see References for full details). It is unlikely that the increase in prevalence in the later studies reflects a real increase in ASD (see text for discussion).
A current concern is the impact of changing the criteria for ASD diagnosis from the DSM-IV-TR to the DSM-5. This change might actually buck the trend of increasing rates of ASD, as many of the behaviours previously included in a definition of an ASD in the DSM-IV-TR would result in a reclassification of the person into having another category of disorder. In fact, one study of the potential impact of the proposed change in diagnostic criteria on prevalence by Worley and Matson (2012), has suggested that the DSM-IV-TR to DSM-% change will reduce prevalence estimates by around 33%. Clearly, the criteria used and differences in professional judgements regarding the diagnosis that exist will impact heavily on this aspect of prevalence estimates.

A further related explanation of the increases in estimated prevalence of ASD concerns the concept of ‘diagnostic shift’: individuals previously diagnosed as falling into one diagnostic category are now placed in another (Shattuck, 2006). The clearest evidence for this suggestion in the context of ASD comes from studies that have noted that as increasing numbers of individuals are diagnosed with ASD, decreasing numbers are diagnosed as having other problems, such as learning difficulties (Croen et al., 2002; King & Bearman, 2009; Matson & Shoemaker, 2009). For example, Croen et al. (2002) noted a 9.1% increase in ASD diagnoses between 1987 and 1994 and a corresponding 9.3% decrease in children diagnosed with mental retardation over the same time period. Although this is the clearest example of diagnostic substitution related to ASD, it is not the only one (see King & Bearman, 2009; Shattuck, 2006). While some have argued that this phenomenon cannot explain the entire increase in ASD prevalence (e.g. Charman et al., 2009; Newschaffer et al., 2005), when coupled with shifts in diagnostic criteria, it is probably sufficient reason to suggest that there is not enough evidence to substantiate the claim that actual levels of ASD are increasing beyond the way that they are calculated and reported.

Methodological issues

Several methods can be employed to estimate the prevalence of ASD, but the two most commonly used are those based on screening particular samples (see Charman et al., 2009; Fombonne, 2003, for discussion) or surveying service and administrative records. Although it is unclear whether one or other of these approaches will systematically produce higher estimates of the numbers of individuals with ASD, emerging evidence has shed some light on this issue. Fombonne (2003) presented data suggesting that during the period 1999–2001, higher rates of prevalence were produced by screening techniques (prevalence rate range: 26.1–62.6 per 10 000), than by survey techniques (prevalence range: 4.8–10.1 per 10 000). This trend is also apparent in some recent estimates of prevalence: Baron-Cohen et al. (2009) suggesting a prevalence of 94 in every 10 000, based on SEN registers in parts of the UK and Baird et al. (2006) suggesting a greater prevalence of 116 in every 10 000, based on a screening procedure.

Screening methods The screening method is considered a more standard technique for prevalence estimates. It involves initially screening a population for cases of actual or suspected ASD and then following-up this initial screen by an intensive
screen conducted on a sample of those individuals identified as having a diagnosis or being at risk of such (e.g. Baird et al., 2006; Chakrabarti & Fombonne, 2001). However, the screening approach presents problems at several points in the procedure that require careful consideration and which have been reviewed (Fombonne, 2003; Williams et al., 2006).

The type of information initially sent out to those identifying the suspected cases of ASD will have an effect on the identification rates, as will the subsequent participation rate (i.e. not all screening letters that are sent out will receive a response). The age of the sample being screened may also play a role. Williams et al. (2006) suggest that greater numbers of individuals are noted in samples that target a younger age group, compared to those samples targeting an older population. The coverage of the initial sample needs consideration in terms of its size relative to the population under consideration. Often these samples are very large: for example, Chakrabarti and Fombonne (2001) covered all 15,500 pre-school children in one UK authority and Baird et al. (2006) sampled over 56,000 individuals in 12 districts in the UK. In some cases, however, the sample is based only on a subset of the entire population to be screened, such as those conducted by Taylor et al. (1999), who sampled around 500 known cases of ASD from a population of around 500,000. In general, prevalence estimates are higher the lower the sample size screened. Similar problems also exist at the intense screening stage of the process, although participation rates typically appear to be relatively high among those who return the initial screening questionnaires (Fombonne, 2003).

Survey methods In terms of the survey approach, typically, the records for an educational authority or service provider are searched in order to note the numbers of children recorded with ASD (see Baron-Cohen et al., 2009). However, it has been noted that there are a number of problems with this, seemingly simple, approach. Such an approach is clearly dependent upon the manner in which an individual’s need is assessed in a particular area, which may depend, not only upon their symptoms, but also on the degree to which help and finances are available in that area (see Charman et al., 2009). An example will serve to illustrate this point. Laidler (2005) examined national administrative data from the USA and concluded that such records may not be suitable for tracking prevalence rates due to a number of peculiarities in this set, such as the flat profile of diagnosis across the age range of children from 1 to 17 years, with a dip between 11 and 12 years old (a transition point in the school system). These data seemed more likely to reflect the needs and constraints of the educational system than reflecting the numbers of children with ASD.

The Economic Cost of ASD

The extent of the economic cost of ASD is, of course, a key issue to service managers with an eye on the budget and is a key issue for interventions in two additional regards. Firstly, such service managers often ask whether any condition is financially important enough to warrant special consideration, over and above the many other conditions that vie for funding. Secondly, if it is an economically important condition, any intervention will not only be assessed against its performance in improving functioning and
prognosis of individuals with ASD, but also against its cost effectiveness. It may be that the intervention that produces the best prognosis and functioning does not produce the best economic return. To answer these questions, it is necessary to turn to health economics.

Unfortunately, health economics is a wretchedly unreliable discipline that relies on assumptions at every level and builds its cost calculations on extremely shaky foundations. Given this, only a short analysis of these findings will be given to offer a flavour of the costs and arguments involved in the treatment of ASD. In a typical analysis of the costs of ASD, assumptions will be made about the prevalence of the disorder, the characteristics of the individuals with ASD and then issues about the associated costs of the lifestyle and services resultant from the disorder, such as residence (supported or not), special education, health and social care, will be made. Additionally, attempts will be made to assess the costs in terms of lost economic productivity for the individual and their families (see Järbrink & Knapp, 2001; Knapp et al., 2009). Once the potential costs have been identified, then figures relating to the costs of these services and so on will be gleaned from existing data or from small samples. Of course, any one of these estimations can be incorrect.

In the UK there have been a number of estimates of the cost of ASD. Järbrink and Knapp (2001) made an assumption that the prevalence of ASD was 5 in every 10 000 and, on this basis, estimated that the annual cost to the country was around £1bn, with a lifetime cost of ASD of around £2.4 mn per individual. However, close to a decade later, with a prevalence assumption that was 20 times higher (100/10 000), Knapp et al. (2009) suggested that the costs to the UK of supporting children with ASD was £2.7 bn per year and £25 bn per year for supporting adults, with a lifetime cost for individuals with ASD of £0.8–1.23 mn if they had associated intellectual problems. An additional cost study, conducted in the USA by Ganz (2007), placed the lifetime cost of ASD at about $3.2 mn per individual. Thus, these later estimates of the cost appear to be fairly consistent, which may just reflect their use of common assumptions that may or may not, in themselves, be correct.

One area of contention in these figures is the associated economic costs to the family. In their estimation of cost, Järbrink and Knapp (2001) suggested that the overall costs of ASD had only a small component that could be related to the family, estimated to be only 2.3% of the total cost. However, the true cost to the family is particularly difficult to assess and Sharpe and Baker (2007) place the cost to the family as being extremely and often ruinously, high. In fact, Arno et al. (1999) attempted to compare the actual cost of care provided to individuals with chronic conditions in the USA and found these costs fell to a substantially greater extent on the family than on the professional services (see Figure 1.3). Although these data were collected in 1999 and in the USA, it is unlikely that the situation is very different in other countries or that subsequent governments have done anything to rectify this situation. Clearly, this is an area in need of further study.

If it is accepted that the cost of ASD is high, then it follows that one outcome of a successful intervention would be to reduce this cost as the individual begins to function more effectively and requires less support. In health economics, this is typically estimated by examining the impact of the treatment on the quality of life of the individual and assessing what the consequent reduction in cost to society will be.
of this improved quality of life for that individual (with the consequent reduction in
need for support). This is usually assessed by reference to the effects of the treatment
on the results obtained from a measurement tool such as the EQ-5D (Gusi et al.,
2010). Unfortunately, there has been very few such quality of life studies conducted
for ASD. Whether or not tools such as the EQ-5D actually measure anything impor-
tant is another question (see Osborne et al., 2014, for a review in different context),
but they are current currency in the field and their relative absence in relation to
ASD makes it difficult to argue for the financial effectiveness of treatment in the
same language as is spoken when discussing the health economics of other disorders
and their treatments. Irrespective of which tools are used to estimate issues such as
quality of life, it is worth keeping in mind when reading the subsequent chapters
that virtually none of the studies of treatment outcome for ASD includes any
measures of this area.

Nevertheless, there are some studies that have attempted to estimate the cost ben-
efits of some interventions for ASD. For example, Jacobson et al. (1998) estimated
that there was a lifetime saving, for an individual with ASD who had undergone an
Applied Behaviour Analytic (ABA) intervention, of between $656 000 and $1 082
000, depending on the effectiveness of the treatment. However, this suggestion was
criticized by Marcus et al. (2000), both on the grounds of being overly speculative
and optimistic in terms of the assumptions made about the costs and outcomes of
ABA programmes and also as no alternative interventions were studied. The latter
problem was remedied, to some extent, by Chasson et al. (2007), who estimated the
savings of employing an ABA approach, compared to ‘treatment as usual’ (in practice
placement in special educational settings), of $208 000 between the ages of 3 and
22 years (i.e. about £11 000 a year). A similar study conducted in Holland by
Peters-Sheffer et al. (2012) estimated a comparative saving of €1.1 mn between the
ages of 3 and 65 years for each individual treated with ABA (i.e. about £14 500 a
year). Again, these estimates are similar to one another, but these figures are based
on assumptions of uncertain validity concerning the impact of such intervention
programmes.

Figure 1.3  Estimated costs of types of care for disabled adults in the USA during 1997
(Arno et al., 1999). ‘Home care’ and ‘Nursing-home care’ refer to professional provided
services, and ‘Informal care’ refers to the cost to families of caring for the individual.
Prognosis for Individuals with ASD

Ultimately, an intervention aims to improve the prognosis for the individual affected and perhaps the best descriptor that can be applied to the findings from the research that has focused on the long-term prognosis for individuals with ASD is ‘variable’ (see Gillberg, 1990; Levy & Parry, 2011). There are a wide range of metrics that can be used in long-term prognosis studies, but these mainly focus on clinically relevant issues (such as independence of functioning and social and economic integration) rather than on the more basic assessments of functioning that are used in outcome-effectiveness studies (such as standardized assessment of intellectual functioning, language skills or adaptive-social behaviours). However, it is difficult to come to any definitive conclusions on this point due to the wide range of outcome measures and techniques used to assess the long-term prognosis for individuals with ASD and also due to the wide range of samples studied, which make it extremely difficult to compare results across reports.

Early reports of the prognosis for individuals with ASD were unremittingly bleak (see Howlin, 1997; Levy & Perry, 2011). For example, Rutter and Lockyer (1967; see also Lockyer & Rutter, 1969; Rutter et al., 1967) reported a series of studies that followed a sample of individuals diagnosed with ASD (using a rather strict definition) and found that over 50% of these individuals were institutionalized 10 years after diagnosis. Similarly, DeMyer et al. (1973) followed individuals with ASD between the ages of 5 and 12 years old and found that 42% were institutionalized at the end of the study. This figure corresponds broadly to that produced by Lotter (1974a; 1974b), who noted that 50% of the somewhat older sample in that study were in hospital and only 1 out of 29 was in employment, at the end of the study period.

However, these rather depressing figures on longer-term prognosis concerning hospitalization have improved over time. In a review of the literature, 20 years after these initial studies were published, Howlin (1997) noted that rates of hospitalization or institutionalization, in studies published after 1980, had dropped to less than 10%. Howlin (1997) relates this improvement to the increase in the number of interventions that had become available to those with ASD in this period. This may well be true (see current Chapter 3), but it has to be noted that this figure may not represent anything as simple or hopeful as intervention-promoted improved prognosis. The change in this figure also corresponds with a closure of many institutions for people with mental problems (see DHSS, 1981, The Harding Report, ‘Care in the Community’) – there were fewer hospitals in which to place people with ASD. Consistent with this view, Gillberg (1990) noted that, while not institutionalized or hospitalized, 66% of the sample with ASD studied was still dependent upon others (i.e. the care cost had just been out-sourced by the government to the family!). Moreover, the decline in institutionalization also corresponds with the introduction of a broader definition of ASD, meaning those with less typical symptoms (and perhaps less severe symptoms) than employed in Kanner’s definition, were also studied in those later reports and for whom prognosis may be better.

In order to give a comparison that has a chance of examining trends in time and to see if the increase in available interventions may have had an impact on prognosis, the notion of the ‘summative’ outcome can be employed (Lockyer & Rutter, 1970; see Levy & Perry, 2011, for a discussion). Most studies will assess whether the individuals have had a ‘good’, ‘fair’ or ‘poor’, outcome. These categories will be based on a range of indices...
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including: independence, employment, functioning and so on, although these may differ from study to study. Pre-1980, a number of reviews have noted that the average of individuals with ASD with a ‘good’ prognosis typically was around 10% (see Howlin, 1997; Levy & Parry, 2011), whereas an average of 60% of outcomes could be said to be ‘poor’ (Howlin, 1997). For example, DeMyer et al. (1973) found that up to 15% of the individuals assessed displayed ‘good’ or ‘borderline’ levels of ‘recovery’ (where functioning was rated as similar to individuals without ASD), 16–25% showed ‘fair’ recovery and 60–75% had ‘poor’ recovery. Inspection of these data post-1980 (i.e. Billstedt et al., 2005; Cederlund et al., 2008; Howlin et al., 2004; Lovaas, 1987), suggests that about 20% of outcomes can now be regarded as ‘good’ (or better) and 50% as ‘poor’ or worse.

Of course, these figures are based on quite different data across studies and also are subject to changes in expectations over time of how an individual with ASD should be able to function in society. When the more positive outcomes (good and fair) are grouped to provide a less fine-tuned distinction, these differences are somewhat less pronounced than when the more subtle distinctions between ‘good’ and ‘fair’ imply. Figure 1.4 shows the percentage of individuals with a fair or good outcome for a number of studies organized across time, where such summative outcomes can be determined and these data make for some salutary reading.

Thus, although there has been some improvement in the prognosis from the initial reports, including fewer hospitalizations and a doubling of ‘good’ prognosis outcomes, this latter figure suggesting improvement only takes into account about 20% of all ASD cases. Indeed, beyond the broad categorization of prognosis into ‘good’ or ‘poor’, there are still concerning issues for ASD prognosis. For instance, Hofvander et al. (2009) found that 60% of individuals with ASDs were unemployed and 84% of their sample had no significant social relationships. These figures are mirrored by those presented by Mordre et al. (2012), who noted that 80% of their sample was unemployed and 92% had no relationship. These poor economic and social integration outcomes are also consistent with the findings of UK Adult Psychiatric Morbidity Survey conducted for people with ASD (Brugha et al., 2009). In fact, the data from
Mordre et al. (2012) show little difference in these figures from those presented nearly half a century earlier by Rutter and Lockyer (1967).

There are two caveats to these rather unsatisfying conclusions on the change in prognosis for people with ASD over the years. The previous figures show the collective outcomes for all cases of ASD studied in these reports. There are, of course, differences in prognosis depending on the severity of ASD and the level of intellectual functioning (see Levy & Perry, 2011), although the literature on this is again mixed. For instance, Mordre et al. (2012) found prognosis was better for those with less severe forms of ASD (i.e. non‐typical autism compared to Autistic Disorder according to the DSM‐IV); whereas Billstedt et al. (2005), using a similar sample, failed to find a difference between individuals with atypical ASD and Autism Disorder in terms of prognosis, although it was noted that prognosis was better for those with Asperger’s Syndrome relative to Autistic Disorder. In terms of IQ, Farley et al. (2009) noted that 50% of individuals with ASD with IQ scores of 100 or greater achieve independence (a level of intellectual functioning that often corresponds to those with Asperger’s Syndrome under the DSM‐IV criteria). Carbone et al. (2010) also suggest prognosis for higher intellectually functioning individuals with ASD is better than for more severe cases – although these positive results, in terms of better social and economic prognoses, should be weighed against the higher prevalence of depression in higher functioning individuals with ASD (see Ghaziuddin et al., 1998; Chapter 2).

It should also be borne in mind that it is actually quite unclear what experiences and interventions the individuals in these prognosis studies have undergone. Carbone et al. (2010) notes that prognosis, especially among the less high functioning, can be improved by intervention and, as noted earlier, this is the point made by Howlin (1997) in examining the change in prognosis figures over time. In fact, it is the role of intervention in improving prognosis that sparked one of the most important debates in this field and, in some ways can be considered pivotal in the development of treatment for ASD. Lovaas (1987; see Figure 1.4) published data from a study of an ABA intervention conducted on young children that showed 50% of the sample having ‘recovered’ (i.e. being indistinguishable from their peers without ASD). These claims were made largely on the basis of the impact of the intervention on the children’s IQ and on their school placement. While this claim has been disputed by subsequent reports (see Chapter 4), it does point to the importance of intervention in developing functioning and potentially highlights one route to improvement of prognosis, in terms of impacting on one area of functioning (such as IQ) and hoping for a knock‐on effect to the more clinically relevant variables.

**Treatment Approaches to be Included**

The preceding three sections have highlighted the size of the problem facing those who would develop interventions for ASD. If these human and economic costs are to be addressed, then the discussion must revert to that of effective treatment programmes and which treatments should be given consideration. As noted previously, there are huge numbers of intervention strategies that have been used in an attempt to help individuals with ASD and it is impossible to detail within one text the evidence for all of these strategies.
To focus the discussion, some selection criteria were outlined in a previous section of this chapter, but the following sections give more detail about the approach to be taken to the selection and assessment of treatments. In particular, the current survey will focus on treatments that are ‘comprehensive’ in nature. There are many views on exactly what a ‘comprehensive’ approach should look like (see Odom et al., 2010; Vismara & Rogers, 2010), but there are certainly three features that need to be present: (i) the intervention must potentially cover the full range of problems associated with ASD; (ii) the approach must be based on some underlying philosophy of treatment that unites the intervention’s strategies and (iii) there must be sufficient evidence from good quality studies on which the assess the impact of the approach.

Wide-ranging nature

A comprehensive approach must attempt to deal with the full range of problems that can present with ASD and they certainly must cover the core problems of ASD. Moreover, any intervention programme that lays claim to the term ‘comprehensive’ should also be capable of being the only from of treatment provided for the individual. That is not to say that such an intervention could not be offered alongside other interventions, if that is desired, but, theoretically, to be termed ‘comprehensive’ it has the potential to be used on its own to provide an encompassing treatment package.

This definition would differentiate a comprehensive intervention approach from a component strategy of that intervention. Although the component strategy might contribute to the operation and success of the overall intervention, it would not, in itself, be used as a treatment. For example, many intervention packages for ASD employ a visual prompt (see Dettmer et al., 2000; Hayes et al., 2010) to help guide the learning of a person with ASD, but, while that visual prompt is clearly part of the intervention, it would not be considered itself as an entire intervention programme.

This criterion also differentiates a comprehensive intervention from approaches that are designed to improve just one aspect of functioning for people with ASD – such as social functioning (e.g. Social StoriesTM by Karkhaneh et al., 2010) or communication abilities (e.g. Picture Exchange Communication System by Frost & Bondy, 2002). These domain-focused interventions may employ only a single intervention strategy, focused on one aspect of functioning or they may employ multiple strategies in dealing with this aspect of the individual’s functioning, but, either way, these focused interventions do not attempt to cover the whole range of problems that a person with ASD may exhibit. For example, the ‘Circle of Friends’ approach (e.g. Whitaker et al., 1998) is a rather complex intervention, involving a number of elements that has been employed for individuals with ASD (James, 2011), but it focuses only on developing the social functioning of the individual and not the restricted and repetitive behaviours or sensory problems, of the individual, so does not qualify as a ‘comprehensive’ intervention.

Cohesive principles of treatment

In addition to providing treatment strategies that impact on a wide range of ASD symptoms, in order to be truly comprehensive an intervention must be based on a cohesive set of principles. In considering what such unifying principles could be, it is
important to understand that these principles can be quite different in form from one another. They may be based on psychological assumptions regarding the controlling variables of behaviour (as in the behavioural approaches to be discussed in Chapter 4 to 6) or the developmental needs of the individual (as in the developmental approaches, to be discussed in Chapters 8 and 9). For example, the model presented by Ospina et al. (2008) places ‘behavioural’ approaches or treatments based around the impact of learning about the environment at one end of a spectrum and ‘developmental’ approaches or treatments organized around within-person characteristics at the other end of this spectrum.

However, it should not be assumed that these principles must fall along some kind of psychological continuum, rather they could relate to other important treatment considerations. Other sets of principles could be established around the core problems that are believed to be important in ASD. Such views suggest that by targeting the ‘core’ deficit of ASD, the symptoms resulting from this central deficit will be remedied (as in some sensory-physical treatments, discussed in Chapter 10). For other approaches, the unifying principle is that the interventions are professionally driven – based on a set of beliefs concerning the best ways to optimize the coherence of the delivery system involved (e.g. the school-parent partnership). These professionally driven programmes focus on a ‘systems-based’ approach to the intervention delivery – and they have a cohesive and coherent view about how the elements in the intervention fit together with one another and have a developed philosophy of education (see Chapter 7). Yet other comprehensive approaches comprise component elements that may have little in common other with one another, other than they are included due to the belief that together they will have a positive impact on the individual with ASD (often termed ‘eclectic’ models and analysed in Chapter 11).

Strength of evidence

Any book focused on assessing the evidence for ASD interventions needs to consider whether there is enough strength of evidence to get these interventions to the wicket in the evaluation game. Thus, the final criterion is that there must have been a reasonable number of good quality studies conducted on the intervention to allow conclusions to be reached or, at least, tentatively suggested. That does not mean that the other approaches than those assessed are ineffective – absence of evidence, is not evidence of absence – but it does mean that these interventions cannot draw on evaluation evidence in their support. If these interventions were drugs, of course, they would not have been released on the market.

It has to be noted that there are many respectable arguments in favour of assessing an intervention from practice; that is, a particular strategy has been used by practitioners and they anecdotally report its success (see Baranek, 2002). These reports are very useful aids to professionals and certainly can be just as helpful as outcome-effectiveness studies in furthering the field. Moreover, this practice-led argument should not be confused with the view that ‘anything should go’ or that, somehow, evaluation in the field is unnecessary (Jordan et al., 1998) or even impossible (see Lord & McGee, 2001, for a discussion of many assessment problems). However, for others to be able to ultimately assess whether a particular intervention should be used, especially one with potentially large resource implications, more
than anecdotal reports or even case reports, are needed. This book will mention some such studies, but only to illuminate the stronger evidence regarding the intervention. Interpretation of the evidence provided by any study is dependent upon its scientific quality.

There are now a substantial number of studies that have assessed the impact of interventions for ASD, however, these studies vary considerably in their quality and hence, in the extent to which any weight can be placed on their conclusions. In considering this issue, Eikeseth (2009; see also Makrygianni & Reed, 2010a; Reichow & Wolery, 2009) outlined four general levels of scientific merit and these are shown in Table 1.1. Levels 1–3 would allow some degree of evidence to be derived from the results and Level 4 would not and only studies with the first three levels of quality have been included in the current book. Thus, these studies have used appropriate research strategies (e.g. randomized control trials, controlled studies and observational studies), assessment across a range of ASD-related behaviours (e.g. cognition, language and communication, social abilities and behavioural problems) and well-validated tools (e.g. standardized intelligence/language tests, Vineland Adaptive Behaviour Scales etc.). Unfortunately, many interventions have not really adhered to these basic needs – often presenting anecdotal case reports or using idiosyncratic assessment tools. These problems reduce the numbers of interventions that can be reasonably assessed quite dramatically.

It should be noted that the treatment approaches that are most often assessed are not necessarily the interventions that are most often employed. Figure 1.5 shows the relative levels of employment of interventions for ASD, as documented by Stahmer et al. (2005) and Green et al. (2006). Apart from illustrating the difficulty in accurately assessing the actual levels of usage of interventions (the results from the two studies

| Table 1.1 Categorization of the scientific merit of outcome-effectiveness studies for ASD interventions, based on Eikeseth (2009). |
|---|---|---|---|
| **Diagnosis** | **Design** | **Measures** | **Fidelity** |
| **Level 4** | Insufficient scientific value | | | |
are quite dissimilar to one another), these data also serve to show that, although many of these approaches are listed also in Figure 1.1 they have not been assessed by quality studies to any large degree. This contrast is presented not only to illustrate that the adoption of evidence-led practice still has some way to go, but also to suggest that outcome-effectiveness studies are not the only driver of the perceived usefulness of an intervention strategy.

The relatively high prevalence of behavioural approaches (54% of all interventions adopted by parents of children with ASD, as noted by Green et al., 2006), is reflected in the high numbers of outcome-effectiveness studies conducted regarding this approach. In the most wide-ranging of the meta-analyses, Ospina et al. (2008) recorded 101 outcome-effectiveness studies and categorized 53% of them as targeting behavioural aspects of ASD. In contrast, Green et al. (2006) note that almost 10% of parents report using facilitated communication procedures, but this technique does not register high in terms of number of studies conducted.

There is one final caveat that the data reported by Green et al. (2006) also highlight and which is important to keep in mind when translating the findings reported here to real world situations. Although 54% of parents use a behavioural intervention of some description, they will also use a variety of other approaches in addition. In fact, Green et al. (2006) report that the average number of interventions employed by parents was seven. This number differed across the type and severity of the ASD noted, with a mean of 4.5 interventions being used for individuals with Asperger Syndrome, 7.2 interventions being employed for mild ASD severities and 8.7 approaches being used for severe ASD. Thus, while much of the research effort recorded in this text has been directed as isolating the impact of comprehensive approaches when performed in isolation, this does not necessarily correspond to what actually happens in most cases.
Structure of the Present Chapters

Based on these considerations, the current book assesses comprehensive treatments for ASD that share the characteristics of addressing the full range of ASD problems, being able to be applied on their own, having a coherent underlying treatment approach and having sufficient quality research to allow conclusions to be drawn. It is organized to describe and evaluate five categories of comprehensive interventions, although it should be noted that these are only broad groupings and that they do have some overlap with one another: (i) behavioural approaches (Chapters 4–6); (ii) environmental-systems approaches (Chapter 7); (iii) developmental approaches (Chapters 8 and 9); (iv) core-deficit treatments (Chapter 10) and (v) eclectic treatments (Chapter 11).

Each set of chapters sets out and discusses the conclusions that might be drawn from these studies – examining the evidence for the intervention’s success and the range of situations where it might reasonably be applied. In developing such an evidence base, each chapter is based on reports that have adopted certain study designs: either various forms of outcome-effectiveness studies or descriptive and meta-analytic reviews of these outcome-effectiveness studies.

Outcome-effectiveness studies

There are three broad categories of outcome-effectiveness studies that can be considered as providing evidence of, at least, Level 3 standard (see Table 1.1): Randomized Control Trials; Controlled Studies and Observational Studies. Each of these types of study concentrates on examining the impact of an intervention for ASD on the participants’ functioning over a period of time and they do so by taking both a baseline measure prior to the introduction of the intervention (i.e. a pre-intervention measure) and at least one follow-up (post-intervention) measure after a period of application of the intervention. However, each form of design has its own strengths and weaknesses that need to be borne in mind while assessing these data.

Randomized control trials

This form of evaluation evidence is often thought to be of major importance in the assessment of outcomes for any intervention. Participants in a randomized control trial (RCT) are randomly allocated to either the treatment group (i.e. the targeted intervention for ASD) or to a control/comparison group (which could be an alternative treatment, a waiting list control or a no-treatment group). The RCT aims to overcome any potential selection bias in the samples at the start of the programme through such a random allocation. If participants are assigned to a group for non-random reasons, then the groups may well differ from one another at baseline along some critical dimension and the difference along this dimension, rather than the treatment itself, may be the critical factor. The groups in the RCT are then given a pre- and post-test measure and the relative change between them is taken to indicate the impact of the intervention in the absence of any other confounding variable.

Although sometimes thought of as a ‘gold standard’ of evaluation, the RCT study does have some drawbacks. A practical limitation is that it can be very difficult to obtain high numbers of individuals for such studies as they require recruitment
**de nova**, rather than utilizing existing programmes. Also, it may not be ethical to randomly allocate people to treatment alternatives, especially if one possible alternative is to receive no treatment. Finally and importantly, individuals do not choose treatments randomly and violation of this aspect of the process can reduce both the external validity of such studies and the estimation of the effectiveness of the treatment: if a treatment relies to some extent on patient compliance, as most psychological treatments do, then denial of patient choice may reduce this factor and the efficacy of the programme.

**Controlled studies** Controlled studies offer an alternative to the RCT and they attempt to compare two or more interventions across pre- and post-intervention tests. The major difference from an RCT is that there is no randomization of participants into groups prior to the start of the intervention. As noted previously, this could introduce a number of confounds into the study, such as baseline differences between the groups of participants. Although most of these studies do attempt to examine whether there are any baseline differences in the participants in the various groups, the critical differences may not be measured. However, while this consideration does reflect a limitation of the controlled study, it also produces an opportunity to study the impact of the ‘real world’ effectiveness of the interventions and this may well offer external-validity advantages over the RCT, as discussed earlier. The other key advantage of a controlled study is that it can utilize pre-existing interventions more readily than a RCT and, consequently, it may be able to recruit greater numbers of participants in each study.

**Observational studies** Both the RCT and the controlled study offer opportunities to compare the effectiveness of two or more interventions over a period of time. This allows both an assessment of the impact of the target programme over time and of its relative impact compared to an alternative programme. However, it may not always be possible to compare two programmes (or indeed ethically desirable to do so) and the final form of outcome-effectiveness study to be discussed is the observational study. In this type of study, participants are measured at baseline and at a follow-up point and the change in their functioning is calculated, but no control group is included. The outcome data from observational studies are almost always collected on participants undergoing an existing intervention, rather than an experimental approach and, as such, represent an accurate real world effectiveness of a treatment.

The observational study allows an idea of the change over time as a result of exposure to an intervention, but a major problem with the interpretation of such change is that it may have occurred anyway with time in the absence of the intervention. However, as with the controlled study, an observational study can utilize existing interventions and can recruit significant numbers of participants. This recruitment can be even greater than in a controlled study, as there is no requirement for a comparison group and this presents another opportunity; with sufficient numbers of participants, the potential impact of those baseline characteristics on the treatment outcomes can be more easily established. That is not to say that this cannot be done with these two sorts of study design, but it is easier with more participants.
Assessing the collective evidence

The conclusions that can be drawn on the bases of these individual outcome-effectiveness studies are obviously limited by the particular characteristics of the study and the sample involved. For this reason, two additional forms of analysis of the impact of interventions for ASD often have been employed, which combine the results of individual studies and the current book draws on both techniques.

One method of combining and integrating the results of individual studies is through the traditional descriptive review of the literature. This provides an opportunity to describe and characterize the area. However, a problem with such descriptive reviews is that they tend to be somewhat selective and potentially idiosyncratic, in their inclusion of studies. Nevertheless, where there are relatively few studies of a particular intervention and where these studies each measure quite different areas of functioning from one another, this narrative technique will be employed.

However, in an attempt to become more systematic in the analysis of outcome-effectiveness studies and to overcome some of the problems of a narrative review, the meta-analysis (see Wilson & Lipsey, 2001) has been developed. The meta-analysis attempts to give specific criteria for inclusion of studies and gives quantitative aggregate summaries of the outcomes of the individual studies. The current book will perform its own meta-analyses on each sort of intervention included where the data permit. These will be conducted to assess the overall effectiveness of an intervention and also to allow potential predictors of its success to be established.

A critical aspect of any meta-analysis is the calculation of effect sizes and these metrics will form a large part of the subsequent evaluation of the interventions. ‘Effect size’ is the name given to an index of the magnitude of a treatment effect and there are many ways in which these can be calculated (see Wilson & Lipsey, 2001). The current text will employ Cohen’s $d$ as the effect size statistic of analysis (see Cohen, 1988), as it is relatively straightforward to understand and can be calculated from basic data contained in many articles or estimated from the results of reported statistical tests or from reports of other effect sizes.

In general, for the interpretation of effect sizes, Cohen’s (1988) suggestions are followed here: an effect size of 0.30 or less is taken to be small (and clinically insignificant); an effect size of between 0.31 and 0.69 is of medium size (and clinically relevant) and an effect size of above 0.70 is taken to be large (and clinically important). However, one important proviso should be given in the interpretation of these effect sizes: not all studies give adequate descriptions of the data to allow such calculations and, in some instances, the effect sizes are only approximations. However, since the goal is not to establish the exact effect size, but to give indications of effectiveness and predictors, this approach seemed satisfactory.

It is possible to produce two expressions of the effectiveness of interventions using effect size as a metric (Wilson & Lipsey, 2001). Firstly, the change between the pre- and post-treatment performances of the individuals can be calculated and this is the only type of effect size that can be performed for an observational study. This effect size is typically expressed by the change in the group mean produced by the intervention divided by the standard deviation of the scores at baseline. The method used to calculate this metric throughout the current text is shown in Table 1.2, but also may be illustrated by a simple example: if an IQ measure had changed by 9 points at the
post-test compared to the pre-test and the standard deviation of the samples’ IQ at baseline was 15, then this would give an effect size of 0.66 (i.e. 9/15).

Secondly, the performances of individuals in an intervention programme and in a control group can be compared with one another and this analysis can be performed for controlled studies and RCTs. The calculation for this metric may also be illustrated by an example: if IQ improved by 9 points over treatment for the intervention group and by 3 points for the control group, then the difference would be 6 points in favour for the intervention group. If the pooled standard deviation of the two groups at baseline was 15, then this effect size would be 0.4 (i.e. 6/15). This is a simple example and the calculation gets somewhat more complicated if the standard deviations of the two groups are different at baseline or the sample sizes are different (see Wilson & Lipsey, 2001; Makrygianni & Reed, 2010a). Nevertheless, the basic calculation is quite clear.

Both of these types of effect size will be employed in the current assessments. However, in terms of comparing one intervention against another, effect sizes derived from the first type of analysis are often most useful. While the effectiveness of an individual programme against a control needs the second type of effect size – when combining across different studies, for different interventions, the control conditions might show radical differences from one another. Differences in the control groups used would impact on the relative sizes of the second effect size, in a way that it will not impact the first.