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Annual Research Review: Child and adolescent mental health interventions: a review of progress in economic studies across different disorders

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Background: Resources for supporting children and adolescents with psychiatric disorders continue to be scarce. Economics research can identify current patterns of expenditure, and help inform allocation of treatment and support resources between competing needs or uses. Scope and methods: The aim was to identify the costs of supporting children and adolescents, the economic impacts of childhood psychiatric disorders in adulthood and any new evidence on the cost-effectiveness of interventions. An electronic search of databases (including PubMed, Medline and Psychinfo) identified peer-reviewed journal articles published between 2005 and 2012. Findings: Sixty-seven papers provided data on support and treatment costs now or in the future, or cost-effectiveness analyses of services. Half the articles came from the United States. Most articles focussed on autism spectrum disorder (ASD; 23 articles), attention deficit hyperactivity disorder (ADHD; n = 15), conduct disorder (CD; n = 7), and anxiety or depression (n = 8). **Conclusion:** Only 14 studies used a cost perspective wider than health care; most included education costs (n = 11), but only five included costs to the justice system. The number of studies estimating costs to the family has increased, particularly for children with autism spectrum disorder (ASD). In the United Kingdom, support costs for children and adolescents with conduct disorder (CD) appear to be lower than for those with attention deficit hyperactivity disorder (ADHD), although for the United States, the opposite may be true. Support costs for children and adolescents with ASD may be higher than both CD and ADHD. However, there were many differences between the samples and the methods employed making comparisons between studies difficult. Outcomes in adulthood include negative impacts on (mental) health, quality of life, public sector services, employment status and income. The evidence base is improving for child and adolescent psychiatric disorders, although only one full cost-effectiveness analysis was identified since the previous review published in 2012. However, we still do not know enough about the economic implications of support and treatment for specific disorders. Keywords: Economic evaluations, childhood psychiatric disorders, treatment costs, support, costs, outcomes, resources.

Introduction

Economic evaluation is increasingly becoming part of the research landscape and integral to the evidence base that supports services and interventions. In the United Kingdom, for example, NICE produces guidance for care providers to promote good health and prevent ill health that has weighed up both the costs and the benefits of treatments (www.nice.org.uk/ guidance). While economic evaluation of interventions for child and adolescent psychiatric disorders has lagged some way behind its adult counterpart, there have been three recent reviews (Kilian, Losert, McDaid, Park, & Knapp, 2010; Romeo, Byford, & Knapp, 2005; Zechmeister, Kilian, & McDaid, 2008). Each of these reviews aimed to identify and summarise evaluations that explore both the costs and the outcomes of interventions to treat or prevent mental health problems.

This review aims to do something slightly different, not least because the production rate for full economic evaluations is low, and take a wider look at the recent cost-related literature for child and adolescent psychiatric disorders. The first aim is to identify the costs of supporting those with psychiatric disorders. To match the breadth of consequences that childhood psychiatric disorders can have, the main interest is in studies that take a broad cost perspective. Health services can provide an important contribution to young people's health and mental health, but education plays a large part in children's lives and those with psychiatric disorders often require additional supports to help them attain. The justice system may bear considerable costs for some adolescents and, of course, families bear costs in terms of (co-) payments for services, other out-of-pocket expenditures, lost employment, and additional time spent caring. A narrow health care perspective may miss a large part of the full cost burden.

The costs of supporting children and adolescents with psychiatric disorders can be far higher than for their peers without these problems, but such disorders can also lead to problems in adulthood. They can lead to continued implications for public sector services, peer and family relationships, and reduced participation in the labour market. The second aim of this article, therefore, is to explore the extent to which these 'downstream' costs have been tracked or predicted. Bernfort, Nordfeldt, and Persson (2008) outline the data domains required to estimate societal

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costs for childhood psychiatric disorders, illustrated with ADHD: psychosocial and psychiatric problems, (lower) educational achievement, substance abuse, risky behaviour, production loss, health care and material costs, criminality, life years lost and intangibles such as impaired quality of life for themselves and their family. The authors found that the lack of data on longer term consequences of the disorder was a particular challenge to such estimates.

The final question to be addressed here links closely with the earlier reviews. Is there new evidence from full economic evaluations of interventions for childhood or adolescence psychiatric disorders? This includes evidence from cost-effectiveness and cost-consequences analyses that, alongside costs, use in their analysis 'natural units' of outcome such as a change score from clinical scales, or depression-free days. It also includes cost-utility analyses, which use a health-related quality-of-life outcome such as quality-adjusted or disability-adjusted life years (QALYs, DALYs). These studies are generally undertaken within a randomised controlled trial, or other comparative design, and measure shorter term *incremental* costs and outcomes.

Box 1: Types of economic studies **Cost analyses** Cost of illness studies Sums the costs incurred for treating or supporting people with similar problems. Cost offset studies Compare costs incurred with costs saved with no reference to outcomes (individual changes in clinical status or quality of life). Cost minimisation analysis Compares alternatives to find the treatment option with the lowest cost. This is justifiable where the analysis has already shown no difference in outcomes between the options. Cost and outcome analyses Cost-effectiveness analysis Measures outcome using clinical assessments or a single outcome such as the number of symptom-free days. The net-benefit approach, linked to the construction of cost-effectiveness acceptability curves (CEACs), can show the probability that an

intervention is cost-effective for different valuations of a unit of outcomes (improvement). Cost-consequences

Cost and outcome results are presented for change on each of the domains assessed in the study. Cost-utility analysis

Values the impact of the intervention in terms of improvements in preference-weighted health-related quality of life, such as the Quality Adjusted Life Year (QALY). Has the potential to allow comparisons across diagnostic or clinical groups.

Cost-benefit analysis

Costs and outcomes are valued in the same (commonly monetary) unit. The difficulties associated with valuing individual outcomes in monetary terms mean that this type of analysis is rarely undertaken.

Source: Romeo et al. (2005)

Methods

Calculating and categorising costs

Costs arise as the result of resources being used over a specified period: perhaps, quarterly outpatient appointments, a number of days as an inpatient, weekly social work contacts, or attendance at a special unit in school. Consistency in unit cost estimations (cost per hour, per attendance, etc.) to attach to these resource use details is important as it allows comparisons between studies. Both the United Kingdom and the Netherlands have 'standardised' methods to estimate unit costs based on economic theory (Curtis, 2012; Oostenbrink, Koopmanschap, & Rutten, 2004).

Two cost initiatives were specifically aimed at helping policy makers and providers understand cost information. For South Africa, researchers developed a spreadsheet, attaching costs to province-level information about population and age, epidemiologic data, service utilisation and associated staff requirements (Lund, Boyce, Flisher, & Kafaar, 2009). The analysis estimated that the costs for minimum and full coverage of public sector child and adolescent mental health services were 5.99 and 21.50 per child per annum (US\$PPP, 2005). The authors report that full coverage would amount to 20% of the Department of Health's budget. Identifying the links between population needs, resources (buildings and staff) and costs can provide invaluable help in developing services. In Australia, the National Mental Health Benchmarking project aims to help child and adolescent mental health services understand why their performance and costs per treatment hour and per episode might vary (Brann, Walter, & Coombs, 2011; Furber, Brann, Skene, & Allison, 2011). Economic theory suggests eight reasons why costs might vary: prices paid for labour, service location or provider sector, organisational factors, service outputs, quality of care, patient characteristics or outcomes (Beecham, 2006; Beecham, Green, Jacobs, & Dunn, 2009).

Once costs have been calculated, they are often categorised in different ways. Direct and indirect costs are commonly used terms but are not always applied consistently across research analyses. In a pan-European cost of illness study, the authors defined 'direct health care costs' as those goods and services related to the prevention, diagnosis and treatment of a disorder (Gustavsson et al., 2011; Olesen, Gustavsson, Svensson, Wittchene, & Jönsson, 2012). 'Direct non-medical costs' were defined as other goods and services related to the disorder but provided outside the health care system, and 'indirect costs' included lost production due to work absence or early retirement. Combining costs resulting from resources used in these domains allows calculation of the costs of support for a child or for a specified group of children. When these terms are used below, they follow these definitions.

Public sector costs include services funded through the state financing system and can cover both direct health and nonmedical costs. Costs can also arise where parents must pay (or part-pay) for services, where adaptations or additional repairs have to be made in the home, or where carers lose time from work to help the child. In the pan-European study, costs associated with informal care (time spent by parents and other family members) was included with direct nonmedical costs as informal care was assumed to replace formal services. More commonly, researchers identify family-borne costs separately.

Identifying the literature

Medline, Pubmed and Psychinfo were used to search for peer-review journal articles published in English between 2005 and 2012. Search terms (including wildcard and truncation symbols) were employed that would identify a wide range of cost-related papers (for example, economic, cost, cost-effective) that focussed on children and adolescents (terms such as child, adolescent, youth, young person/people

and by age group). The terms mental health/illness and psychiatric disorder formed part of the search parameters, as well as individual disorder terms. Exclusion criteria included infants and adults, children whose parents had mental health problems, prevention services and disorders usually seen in adulthood. Evaluations of psychiatric medication were excluded. Most such evaluations have a narrow payer-only perspective (Wu et al., 2012); the majority focus on medication for ADHD (Schlander, 2010).

An initial screen of titles and abstracts led to the removal of 886 citations that were duplicates, only considered medication or which did not include cost data. The remaining 121 papers were scrutinised in more detail and 64 were found to provide data on the costs of support, now or in the future, or included a cost-effectiveness analysis. Separate searches using ASSIA, EMBASE, ERIC and the Cochrane Library identified a further three papers, and 'early view' papers were identified in conversation with colleagues.

Results

Most articles focussed on a single disorder, commonly autism (23 articles) or attention deficit hyperactivity disorder (n = 15; see also Bishop, 2010; López-Muños, Alamo, Quintero-Gutiérrez, & García-García, 2008). A further seven papers estimated the costs associated with conduct disorder, and eight focussed on anxiety or depression. Following an overarching section, the studies for each of these broad disorder groups are described below. The remaining seven articles are considered in the subsequent section, five of which relate to the costs of eating disorders. Low numbers of papers mean that these sections are more discursive than analytic, but the final section provides an overarching discussion of the cost-related literature on child and adolescent psychiatric disorders.

Where the journal articles report at least direct health and direct nonmedical costs, results are presented in Tables 1-4. The Tables also show these costs recalculated to their value at US\$ PPP 2012. Following the procedure used in the pan-European study on the costs of brain disorders (Gustavsson et al., 2011), each country's consumer price index was employed and the resulting figure then converted to US\$ using purchasing power parity values (PPP) for total consumption (GDP). Although these costs are presented at comparable price levels, some caution is needed when making direct comparisons between the studies. The samples in each study may not have the same degree of disorder severity, may be more (or less) representative of the full population of these children or adolescents, may include a slightly different array of services and, where the estimates come from different countries, will be derived from different service and financing systems.

The costs of child and adolescent mental health problems

Few studies provided a national picture of the costs of all child and adolescent psychiatric disorders. The recent pan-European cost of brain disorders study included only three childhood psychiatric disorders. Total costs were estimated to be €21.3 billion per year (€PPP, 2010) for an estimated 5,932,112 children with attention deficit hyperactivity disorder (age 6–17 years), conduct disorder (5–17 years) or autism spectrum disorders (2-17 years; Gustavsson et al., 2011; Olesen et al., 2012). Indirect costs were excluded, but direct health care costs accounted for just 12% of the total with direct nonmedical costs (including informal care) absorbing the major part (88%). However, while 11 studies supported the prevalence estimates, the authors judged just four to be of sufficient quality to inform the cost estimates (p758; Hakkaart-van Roijen et al., 2007; Järbrink, 2007; Knapp, Romeo, & Beecham, 2009; Romeo, Knapp, & Scott, 2006). Average annual costs per child were €3,595. Annual costs were lowest for ADHD (\notin 781), and highest for autism (\notin 27,261); costs per case for conduct disorder were €1,735 (Olesen et al., 2012).

Recent estimates for the costs of all childhood psychiatric disorders in the United States are based on an earlier calculation (Ringel & Sturm, 2001). Uprated from 1995 to 2007 prices, mental, emotional and behaviour problems in children were estimated to cost \$247 billion per annum (O'Connell, Boat, & Warner, 2009). To account for treatment provided in non-(mental) health settings, health care costs were multiplied by two based on a study by Costello, Copeland, Cowell, and Keeler (2007).

Recent analysis of the British Child and Adolescent Mental Health (BCAMH) Surveys provides some of the most up-to-date information on the costs of supporting and treating mental health problems in the United Kingdom (Snell et al., 2013; 2007–2008 prices). The BCAMH surveys are designed to have a nationally representative sample of all British children aged 5-15. The total mean cost for all children with a psychiatric disorder was £1,803 per child per annum, amounting to £1.47 billion nationally, more than three-quarters of which was absorbed by additional education services. In contrast to the pan-European study, mean costs per child were highest for those with hyperkinetic disorders at £3,108 per annum, followed by conduct disorders (£1,856), and the lowest costs accrued for emotional disorders at £1,165 per annum. Costs falling to the justice system were excluded due to poor quality data, but can be high, particularly for those with externalising disorders.

Both UK and US research has shown that children and adolescents with psychiatric disorders can have health care costs at least twice as high as their peers with other disorders or without any disorder. These costs derive from increased contacts with services as well as parental absences from work (see, for example, Ford, Hamilton, Goodman, & Meltzer, 2005; Jones & Foster, 2009; Petrou et al., 2010). Children who have not received any specialised treatment for

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Table 1 Autism cost studies

Authors	Children & circumstances	Cost categories included	Country & year for costs Reported mean costs per child	Costs per annum US\$ PPP 2012
Knapp et al. (2009)	Literature & new analyses of existing data sets Age 0–17 years All levels of intellectual ability	Accommodation (excl. parental home), health & mental health care, education, social care and family expenses (5%)	UK, p.a. 2005–2006 £25,399	\$46,020
	·		United Kingdom, 6 months, 2006–2007	
Barrett et al. (2012)	Baseline data from an RCT for communication. Data from parental interview	Accommodation (excl. parental home), health & mental health care, education, social care	£2,581	\$9,120
	N = 152, age 2–5 years	Parents' out-of pocket expenses	£227	\$802
		Parents' absence from work	£275	\$970
			United States, life-time, 2003 prices	
Gantz (2007)	Literature review, analysis of	Direct medical costs	\$305,956	\$387,950
	survey & claims data	Direct nonmedical costs	\$978,761	\$1,241,069
	Life-time costs	Indirect costs	\$1,875,667	\$3,478,246

Table 2 ADHD cost studies

Authors	Children & methods	Cost categories included	Country & year for costs Reported mean costs per child	Costs per annum US\$ PPP 2012
			United States, p.a., 2005	
Pelham et al.,	Review of literature	Health & mental health care	\$2,636	\$3,140
(2007)	Diagnosed school-age children	Education	\$4,900	\$5,830
		Crime & delinquency	\$7,040	\$8,380
			United States, p.a., 2010	
Doshi et al. (2012)	Review of literature Children & adolescents	Health & mental health care – child	\$621-\$2,720	\$660-\$2,880
		Health & mental health care – family	\$1088-\$1,658	\$1,150-\$1,760
		Family lost productivity	\$142-\$339	\$150-\$360
		Education	\$2,222-\$12,447	\$2,360-\$13,190
		Justice system (adolescents)	\$267	\$280
			United Kingdom, p.a.,	
			2010	
Telford et al.	Longitudinal ADHD Survey	Health & mental health care	£1,310	\$2,170
(2013)	Age 12–18 years with	Education	£4,155	\$4,450
	childhood ADHD	Social services	£28	\$45
			United Kingdom, p.a., 2007–2008	
Snell et al.	BCAMH survey; $n = 445$	Health & mental health care	£269	\$460
(2013)	age 5–15; subsample with	Education	£2,725	\$4,650
	hyperkinetic disorders	Social services	£114	\$190
			Netherlands, p.a., 2004,	
Hakkaart-van	Data from parental interviews	Health care	€728	\$1,010
Roijen et al.	70 children treated for ADHD	Mothers' health care	€2,243	\$3,120
(2007)	by a paediatrician.	Absence from & reduced productivity at work	€552	\$770
			Belgium, p.a. (adjusted), 2002	
De Ridder & Graeve (2006)	Survey of parents of children with ADHD.	Health care, social care, other nonmedical resources	€779	\$1,130
		Parents' health care and out-of-pocket expenses	€588	\$850

a psychiatric disorder can have nonpsychiatric health care costs almost twice as high as their peers without a psychiatric disorder (Wilkes, Guyn, Li, & Cawthorpe, 2012).

For the United Kingdom, recent analysis of the 1970 British Cohort Survey has begun to identify the longer term (age 30) economic outcomes of childhood mental health problems (Knapp, King,

Authors	Children & circumstances	Cost categories included	Country & year for costs Reported mean costs per child	Costs per annum US\$ PPP 2012
			United States, p.a., 2000	
Foster et al. (2005)	Fast Track multicentre study. Interviews and administrative data. N = 80 with conduct disorder	Health & mental health (52%), education (34%), juvenile justice (20%)	(Av. for school years 7–13) \$10,970	\$15,580
			United Kingdom, p.a., 2002–2003	
Romeo et al. (2006)	Baseline data from a controlled trial. Interviews with parents	Health & mental health care, education, social care	£1,277	\$2,450
、	of children with persistent	Parental service use	£45	\$90
	antisocial behaviour.	Parental expenses	£32	\$60
	N = 80, age 3–8 years	Additional parental time on household tasks	£4,526	\$8,680
		Absence from work	£79	\$150
			United Kingdom,	
			p.a., 2000–2001	
Clark et al.	Children identified as 'most	Health & mental health care	£55	\$110
(2005)	concerning'. Interview data	Education	£394	\$800
	from primary carers	Social care/voluntary sector	£532	\$1,080
	N = 56, age 8–19 years, 53% with conduct disorder	Criminal justice system	£35	\$70
			United Kingdom,	
			p.a., 2007–2008	
Snell et al.	BCAMH survey; $n = 445$	Health & mental health care	£128	\$220
(2013)	age 5–15; subsample with	Education	£1,632	\$2,780
	conduct disorders	Social care	£96	\$160

Table 3 Conduct disorder cost studies

Table 4 Anxiety and depression cost studies

Authors	Children & circumstances	Cost categories included	Country & year for costs Reported mean costs per child	Costs per annum US\$ PPP 2012
			Netherlands, p.a., 2003	
Bodden, Dirksen, Bögels, Nauta et al. (2008)	Baseline data for an RCT testing CBT. Data from cost diaries over 2 weeks.	Health & mental health care Housekeeper and friends & family care	€1,396 €65	\$1,970 \$90
(2000)	Clinically anxious youth $N = 118$, age 8–18 years	Absence from & reduced productivity at work, household tasks and school absence	€1,191	\$1,680
		Parental out-of pocket expenses	€95 United Kingdom,	\$130
			p.a, 2007–2008	
Snell et al. (2013)	BCAMH survey; $n = 445$	Health & mental health care	£89	\$150
	age 5–15; subsample with	Education	£1,048	\$1,790
	emotional disorders	Social care	£28 United States, 3 months, 2003	\$49
Domino, Burns, Mario et al., 2009 and Domino, Foster	Baseline data from an RCT $N = 433$, age 12–17 years with depression	Health & mental health care, education, social care, justice system	\$220	\$1,120
et al., 2009	•	Indirect costs (caregiver time & travel)	\$61	\$310

Healey, & Thomas, 2011). Men who had childhood antisocial conduct problems were less likely than their peers without these childhood problems to be economically active, but if they were working, were likely to earn more money. Poor outcomes at ages 36–53 following adolescent conduct problems were also found in the 1946 birth cohort (Colman et al., 2009). Poor employment outcomes at age 30 were also found for adults who had childhood attention deficit disorder (ADD), and if they were working, they

were likely to be in less-skilled, lower paid jobs than those without ADD. Compared with their peers with no externalising behaviours, those with severe or mild externalising behaviour in adolescence were more likely to have symptoms of depression and anxiety or abuse alcohol, and more likely to become parents in their teenage years, to have left school with no qualifications, and to experience adversity (see also, Healey, Knapp, & Farrington, 2004). Analysis of the WHO World Mental Health Survey found that childhood mental health problems were associated with early termination of education in low- and middle-income countries, but not with reduced household income (Kawakami et al., 2012; Lee et al., 2009).

These large-scale analyses are rare, not least because they require large amounts of data, ideally longitudinal, on a wide range of topics. Their usefulness lies in providing a broad estimate of the longer term health and economic outcomes and the resources absorbed, pointing to areas where there is a need to find ways of improving outcomes (and thus reduce these costs), perhaps by more effective or more easily available treatments, which will have an impact both today and in the future.

Autistic spectrum disorder (ASD)

Introduction. Commonly diagnosed around the time of entry to school (Mandell et al., 2010), Autistic Spectrum Disorder (ASD) can have lifelong quality-of-life impacts on children and their families, as well as costly support implications. Children with autism can have severe problems with communication and social interaction and display patterns of ritualistic and stereotypical behaviour. Intellectual disabilities can occur in half of those with autism, and around 30% of UK children with autism have a clinically recognisable emotional or anxiety disorder or an additional diagnosis of conduct disorder (Green, McGinnity, Meltzer, Ford, & Goodman, 2005; Knapp et al., 2009). There are similarly high rates of psychiatric comorbidity in the United States. Analysis of the US National Survey of Children's Health showed that 39% of parents of children with autism (n = 324,000) reported depression or anxiety problems for their child, compared with just over 4% for children without autism (Gurney, McPheeters, & Davis, 2008; see also Peacock, Amendah, Ouyang, & Grosse, 2012). The combination of all these factors can lead to high costs associated with service use, as well as for the family.

Twenty-three papers were found that explored the costs of supporting children and adolescents with ASD, including three that take a wide cost perspective (see Table 1) and six that identify the scope and level of family-borne costs.

Results. Just two studies identifying the costs of supporting children and young people with autism

were found for the United Kingdom. Using a prevalence of at least 1% across all ages, one estimate suggests that there are 107,016 children and adolescents age 0-17 years living with ASD in the United Kingdom (Knapp et al., 2009). This study found no nationally representative samples of UK children with ASD, so estimated the costs from existing literature and reanalysis of other research data. Costs included housing (an estimated 1,333 children live in residential or foster care placements, commonly those with additional intellectual disabilities), as well as health, social care and education services, and families' out-of-pocket expenditures (2005-2006 prices). Children were grouped by age (0-3 years, 4–11 years and 12–17 years), with or without intellectual disabilities (ID), and place of residence (at home, in residential or foster care) and national costs estimated for these groups.

- Children with ASD and ID living with families: 0-3 years, £0.86 m; 4-11 years, £739.49 m; 12-17 years, £914.69 m
- Children with ASD and ID in residential/foster care: 0-3 years, £0.86 m; 4-11 years, £16.23 m; 12-17 years, £55.03 m
- Children with ASD but no ID living with families: 0-3 years, £1.51 m; 4-11 years, £541.49 m; 12-17 years, £447.93 m

UK data for preschool children were rare. A subsequent estimate for 152 children aged 2– 5 years recruited to take part in a randomised trial of a communication intervention found total support costs of between £317 and £66,980 over 6 months (mean = £2,580; 2005–2006 prices; Barrett et al., 2012). Education and child care absorbed 45% of the costs, 41% were for community health and social care services (80% of the children had seen a speech and language therapist) and hospital services accounted for just 12%. All but one child lived at home and mean family costs and productivity losses were estimated to be around £3,000 over the 6-month period (range £556– £9,611).

One US study used a similarly wide cost perspective and drew on the published literature to create a US-based prevalence-based cohort from which *lifetime* costs could be calculated (Gantz, 2007). The costs included all hospital and health care services and equipment, child care, adult care, respite care, home care and modifications, special education and supported employment (2003 prices, discounted at 3%). Over a person's lifetime, direct nonmedical costs were three times greater than direct health care costs, and indirect costs made up more than half of the total. The total lifetime incremental costs per capita were as follows:

- age 3–7, \$446,203;
- age 8–12, \$314,297;
- age 13–17, \$285,082.

Other US studies focused on health care costs, commonly analysing large administrative or insurance claims' databases. Originally estimated at 2003 and 2004 prices, three of the four studies show health care costs for children with ASD of around \$6,000 (Leslie & Martin, 2007; Liptak, Stuart, & Auinger, 2006; Shimabukuro, Grosse, & Rice, 2008). The fourth study, with data on children taken from one health plan, shows a much lower cost per annum of \$2,757 (Croen, Najjar, Ray, Lotspeich, & Bernal, 2006). Co-occuring intellectual disabilities can increase costs by a factor of 2.7 (Peacock et al., 2012), but the rising health care costs for children with autism are likely to be

because more children are treated rather than because children receive more intensive treatment (Flanders, Engelhart, Pandina, & McCracken, 2007; Wang & Leslie, 2010). Health care costs have been found to be higher for

children with ASD than for those without ASD (Croen et al., 2006; Liptak et al., 2006; Peacock et al., 2012; Peng, Hatlestad, Klug, Kerbeshian, & Burd, 2009; Ray et al., 2006; Shimabukuro et al., 2008) with significantly higher numbers of visits to all types of physicians (Gurney et al., 2008). Health care costs have also been found to be higher for children with ASD than for those with asthma, diabetes or other psychiatric disorders (see, for example, Flanders et al., 2006; Flanders et al., 2007; Mandell, Cao, Ittenbach, & Pinto-Martin, 2006). A recent review confirmed these findings, although not for children with depression, and also concluded that families of children with ASD had greater difficulties accessing services and more unmet needs (Trenago & Cheak-zamora, 2012). There is also evidence of a fall in the level of support young people with ASD receive once they leave school and should be supported by adult services (Clarke et al., 2011; Shattuck, Wagner, Narendorf, Sterzing, & Hensley, 2011; Singh, 2009).

Family-borne expenses are rarely estimated for different groups of children with special needs or disabilities (see, for example, Busch & Barry, 2007), although six US studies have looked at these costs for children with ASD. Family-borne costs include those accruing for time spent in informal care activities (although there are few data on this), impact on employment and income, (part-) payment for services and medications, special foods, equipment and other additional household items.

Co-payments for medical care can be costly in the United States. Liptak et al. (2006) estimated the mean for children with ASD was \$613, slightly lower than for children with depression, but higher than for other psychiatric disorders by a factor of seven (1999–2000 prices). More recent US studies have found that while 60% of parents do not make copayments for health care, on average, these costs are around \$700 (Young, Ruble, & McGrew, 2009) and that copayments may absorb more than 3% of family income (Parish, Thomas, Rose, Kilany, & McConville, 2012). Mothers of children with ASD may also be relatively disadvantaged in the workplace. One US study found them to be 6% less likely to be employed and earn 56% less than mothers of children with other health limitations (Cidav, Marcus, & Mandell, 2012). There may also be a higher overall impact on family income where there is a child with ASD; 14% annual income compared to 2% for other special needs children (Montes & Halterman, 2008).

In noninsurance-based systems, such as the United Kingdom, copayments for services are rarer. Family-borne costs, therefore, may be much lower. Among their preschool sample, Barrett et al. (2012) found that families spent an additional £227 over 6 months on adaptations and repairs to the home, large and small equipment items, training, special diets, travel to receive services and assessments in abroad. Half the families in this study reported taking time off work (on average 22 hr over 6 months).

Conclusion. There were more papers related to ASD found in this literature search than for any other psychiatric disorder. Although the costs in Table 1 are all presented in comparable prices, the three studies cannot easily be compared. The data sources are different for each study, as is the scope of costs. The two UK studies show data over 6 to 12 months and the US study presents lifetime costs. The net effect of the literature is that there is a reasonably good set of data about the scope of health care costs in the United States, particularly hospital costs, but less about the implications of ASD for other provider sectors, such as education. For the United Kingdom, the research evidence on the costs of support for children with ASD is very patchy. For other countries, evidence is even rarer with just one study from Sweden of children with high functioning autism (Järbrink, McCrone, Fombonne, Zande'n, & Knapp, 2005). It is, however, very encouraging to see studies beginning to explore the various costs to families.

Early diagnosis and early intensive treatment are recommended for children with ASD, but we know little about how best to use resources to intervene to reduce some of the consequences of ASD. There is evidence of the effectiveness of early intervention ABA programmes (Peters-Scheffer, Didden, Korzilius, & Sturmey, 2011). Potential cost-savings, if these relatively high-cost intensive behavioural interventions were more widely available, have been estimated for Ontario, Canada (Motiwala, Gupta, Lilly, Ungar, & Coyte, 2006), Texas in the United States (Chasson, Harris, & Neely, 2007) and for the Netherlands (Peters-Scheffer, Didden, Korzilius, & Matson, 2012).

There are no individual-level cost-effectiveness evaluations of ASD interventions, although a 51-cen-

tre European study of children with pervasive developmental disorder may hold some promise. Two hundred and eight children received an average of 30 hr of intervention per week from licensed professionals. After 3 years, 28% had a lower degree of autism, improved speech and higher developmental age (Darrou et al., 2010). No association between the intensity of the intervention (a proxy for intervention costs) and outcomes was found, and the authors suggest that the focus for future research should be on the *type* of intervention rather more than the number of intervention hours. A new systematic review is underway of all interventions for preschool children with ASD (Tachibana, Green, Hwang, & Emsley, 2012).

Attention deficit hyperactivity disorder (ADHD)

Introduction. There is considerable discussion on the diagnosis and treatment of ADHD. It has a worldwide prevalence of 5.3%, although this is often considered to be highest in the United States (Polanczyk, Silva de Lima, Lessa Horta, Biederman, & Rohde, 2007). Bloom, Cohen, and Freeman (2010), for example, report a prevalence rate of 9% from the 2009 National Health Interview Survey (12% for boys and 5% for girls), a 2% rise on the 2006 figures (11% for boys and 4% for girls; Bloom & Cohen, 2007). There has been an exponential growth in published papers on ADHD over the 25 years to 2005, nearly half of which derive from the United States and nearly half of which concern medication (López-Muños et al., 2008). The current review found 15 journal articles that estimated the costs associated with ADHD, six of which adopted a wider cost perspective (see Table 2).

Results. Pelham, Foster, and Robb (2007) reviewed the US literature published between 2001 and 2005 and found 13 studies from which to build a cost of illness study for ADHD, eight more than had been identified 4 years earlier (Leibson & Long, 2003). Most of the 13 studies focussed on health care costs with just two estimating the costs of ADHD to the education system, and two looking at costs borne by the juvenile justice system. Health care costs (12 studies) were between \$790 and \$5,518 per child annually (mean \$2,636) including average medication costs of \$459 (five studies). More than half of the mean total annual cost of \$14,576 per child comprised juvenile justice and crime costs. Annual societal costs for childhood ADHD in the United States were estimated to be \$42.5 billion (2005 prices).

In the 6 years following this review, Doshi et al. (2012) identified an additional four papers for their literature-based (1990–2011) cost of illness model for childhood ADHD. Health care costs were most commonly reported (nine papers) and again showed a wide difference in the reported estimates (\$621–

\$2,720 per annum). The authors based the costs of additional educational support on three papers (Jones & Foster, 2009; Marks et al., 2009; Robb et al., 2011) and costs to the justice system on just one paper (Jones & Foster, 2009).

Other studies have identified the costs of childhood ADHD for specific parts of the US system. Higher inpatient costs were found for children and adolescents with a secondary diagnosis of ADHD than for those without (Meyers, Classi, Wietecha, & Candrilli, 2010), high costs accrue to the school system (Robb et al., 2011) and in the longer term, to the justice system (Fletcher & Wolfe, 2009). When compared with their peers without ADHD, high service use rates among preschool children were found for speech and language therapy, occupational therapy, physiotherapy and special education, with the associated costs being four times higher (Marks et al., 2009).

Analysis of the Cardiff Longitudinal ADHD Study mirrors these raised nonhealth care costs for the United Kingdom (Telford et al., 2013). Service-use data were collected from parents for 143 young people aged 12-18 years who had been treated for ADHD 5 years earlier. The total mean cost over the previous 12 months was £5,490 (median=£2,330, 2010 prices) with education services accounting for 76%, even though one in 10 of the young people had already left school. NHS health care and medication each absorbed 12% of the total costs. Using the data from the BCAMH large survey (see above), Snell et al. (2013) found observed mean costs for those with hyperkinetic disorders to be slightly lower at £3,108 per annum. Neither of these studies was able to include costs borne by justice system or the family.

Two other European studies add information on family impacts and health care costs. A Dutch study found that the mean direct medical costs for children with ADHD (n = 70) were far higher than for the 35 children with other behaviour problems or the 60 children with no behaviour problems: €2,040, €288 and €177 per annum for each group respectively (Hakkaart-van Roijen et al., 2007). Annual medical costs for mothers attending services were also significantly higher where they had a child with ADHD: €728 compared with €202 (behaviour problems group), €154 (no behaviour problems). Mothers from the ADHD group missed an average of 17 days work per year compared with 3 and 6 days for the other groups. In Flanders, Belgium, parents reported their children's use of health care, social care and other nonmedical resources including out-of-pocket expenses (De Ridder & Graeve, 2006). Compared with their siblings, out-of-pocket expenditure was more than six times higher for the child with ADHD (€588 v. €92 per annum; 2002 prices), and public sector costs were twice as high (€779 v. €371 per annum).

Where medication costs were separately identified, they comprised around 20% of health care costs.

Global spending on ADHD medication grew ninefold to US \$2.4 billion between 1993 and 2003. This spending growth was concentrated in developing countries with the United States absorbing over 80% of the global market (Scheffler, Hinshaw, Modrek, & Levine, 2007), although one study found that the United States may pay higher prices for this medication than other countries (Lang, Scheffler, & Hu, 2010). A systematic review of papers published between 1990 and 2011 found ADHD medication to be more cost-effective than no treatment, placebo, behavioural therapy or community care, but insufficient evidence on the relative cost-effectiveness for different groups of children and adolescents, or for the different pharmaceutical preparations (Wu et al., 2012). Findings from the cost-effectiveness analysis of the Multimodal Treatment Study of Children with ADHD (Jensen et al., 2005) were included in that 2012 review. Other analysis of these data suggests that as children's needs rise, behaviour therapy with or without medication may cost more today, but may generate higher cost-savings in the future (Foster, Jensen et al., 2007).

Conclusion. Table 2 shows that the direct health care costs estimated across four countries vary widely, from US\$ *PPP* \$660 to \$3,140 (both estimates are from US studies). Among the European countries, the UK studies estimate both the lowest and the highest health and mental health care costs. These two studies have a different sampling frame: a longer term follow-up of a clinical/research sample (Telford et al., 2013) and a national survey (Snell et al., 2013), but include a similar scope of costs.

Education costs are estimated for four studies shown in Table 2, and for three are around \$5,000 per annum (US\$ PPP). Crime-related costs, derived from existing literature, were estimated in just two of these studies. The lower figure (Doshi et al., 2012) uses data from two earlier studies, one of which includes only the costs to the justice system. The higher figure includes victim costs (Pelham et al., 2007; p719). As with studies estimating the cost of ASD, most analyses focus on health care costs (see text) with just one recent US article from the Fast Track study additionally incorporating costs to the education (32%–57% of total costs over the age groups) and justice (5%–25%) systems (Jones & Foster, 2009).

Studies looking at the relative cost-effectiveness of treatment options are rare. Six studies including a nonmedication treatment option were included in Wu et al. (2012), only two of which were published since 2005 (Foster, Jensen et al., 2007; Jensen et al., 2005). Both these analyses use the Multimodal Treatment Study, which evaluated three treatment modes. The main cost-effectiveness results show that medication treatment dominated community care plus behavioural therapy and behavioural therapy alone (Jensen et al., 2005). This literature search revealed no other articles reporting cost-effectiveness analyses of nonmedication ADHD treatments.

Conduct disorder (CD)

Introduction. Conduct disorder (CD) is another relatively common childhood psychiatric disorder. With a prevalence rate of about 5%, it is likely to persist into adulthood in about half of all childhood cases. CD is characterised by repetitive patterns of antisocial, aggressive or defiant conduct. Behaviours such as lying, bullying, threatening or intimidating others are common, as is truanting from school. Preschool diagnosis is common; a short screener completed by teachers can identify at-risk kindergarten children (Jones, Dodge, Foster, Nix, & Conduct Problems Prevention Research Group, 2002). The literature search identified a small cluster of cost papers based around the US Fast Track study, two separate UK studies, and two reviews of the economic evidence for parenting programmes and CD. Table 3 shows the support costs derived from four studies where costs to nonhealth care sectors have been estimated.

Results. The Fast Track Study has provided some of the most up-to-date US data on the costs of conduct disorder. Recruitment for the study began in 1990 and continued for 5 years, and the study makes use of interview and administrative data to calculate a wide range of support costs (2000 prices; Foster, Damon, & Jones, 2005). Costs over school years 7-13 were far higher for children with CD (n = 59), than for those with oppositional defiant disorder (n = 78), those within one criterion of being diagnosed (n = 40) or those with no behaviour problems (n = 488). School costs accounted for the largest proportion, followed by juvenile justice costs (20%). Cumulative costs by school year 13 for those with CD amounted to over \$70,000 more than for those with no behaviour problems. By linking the Fast Track intervention costs with its impact on conduct disorder, subsequent analysis found that such expensive interventions are likely to be cost-effective if they can be targeted on those who will be costly to society when left untreated (p1289; Foster, Jones, & the Conduct Problems Prevention Research Group, 2006).

Two studies provide some insights into the high costs of conduct disorders in the United Kingdom. Table 3 shows that both Romeo et al. (2006) and Clark, O'Malley, Woodham, Barrett, and Byford (2005) include a wider range of cost categories than do Snell et al. (2013). Persistent antisocial behaviour is a core feature of conduct disorder, and total support costs for 80 children, all assessed as being in the worst 1% for disruptive behaviour, were between £49 and £19,940 per annum, around a mean of £5,960

(2002–2003 prices; Romeo et al., 2006). Service costs (£1,277) fell mainly to the health care system and education, but the nonservice family-borne costs (time taken off work, additional time spent preparing meals, shopping, cleaning, etc.) far outweighed the service-related costs at 78% of the total. Greater severity of behaviour problems raised costs but explained only 7% of the cost variation.

In the second UK study, local services were asked to identify their 'most concerning youth' (Clark et al., 2005). Just five of the 56 young people had no psychiatric diagnosis and 53% were diagnosed with conduct disorder. In contrast to the first study, more than half (60%) had lived in specialist facilities away from home over the year for which costs were calculated. Mean annual public sector costs were therefore much higher at just over £52,000 per year (£1,017 per week, 2000-2001 prices) with half accruing to social care services, and a third to education. Costs to health care and the justice system were surprisingly low (5% of total costs each), in part due to high use of specialist nondomestic accommodation with relatively high levels of staffing (52% of total costs). Costs were significantly higher for younger children, those living away from home, showing inappropriate sexual behaviour and if the child was from a white ethnic background. Measures of mental health needs (severity) did not influence costs.

The high costs of supporting children and adolescents with conduct disorder and the likely downstream implications (see above, Knapp et al., 2011) suggest that early treatment is key to reducing costs. An economic case has been made for a number of childhood interventions in the United Kingdom: parenting programmes, school-based social and emotional learning programmes, school-based interventions to reduce bullying, and early detection and intervention for psychosis (Knapp, 2011). Evidence-based parenting programmes, for example, may be cost-saving to the public sector within 5-8 years, and save £16,450 per family over 25 years (see also, Bonin, Stevens, Beecham, Byford, & Parsonage, 2011). Empirical analyses of 20 years of US trials (n = 459) supported 'stacking' parent, teacher and child components of the Incredible Years programme as a means of providing cost-effective (intervention costs only) treatment for behaviour problems in children age 3 to 8 years old (Foster, Olchowski, & Webster-Stratton, 2007).

Implementation is key to both shorter term cost-effectiveness and achieving the predicted savings for early interventions. However, there is a paucity of evidence on whether parenting programmes can be rolled-out and run effectively in routine practice, how to encourage parents to complete the course, how to identify and target groups of parents likely to benefit, how to engage hardto-reach populations and whether the programme will engender benefits into adulthood (Bonin et al., 2011; Stevens, 2012).

Conclusion. In the United Kingdom, support costs for children with CD appear to be lower than for children with ADHD (see Tables 2 and 3; Snell et al., 2013). For the United States, the Fast Track study suggests that the support costs for children with CD might be higher than for those with ADHD, and that public sector costs for children with both ADHD and CD are twice as high as for ADHD alone (Jones & Foster, 2009). However, when looking at the data in Table 3, it is important to take each sample's age range into acount. As Foster et al. (2005) note, costs linked to crime increase as children age, and thus were less relevent in the Romeo et al. study (2006; children ages 3-8 years) and they were excluded from the analyses undertaken by Snell et al. (2013) even though the age range was 5-15 years. The analysis by Clark et al. (2005) sits slightly apart from the other three papers as only half the young people were diagnosed with conduct disorder. Moreover, much higher use of costly highly supervised nonhospital residential establishments than in any of the other studies raised total costs considerably. Notably, there is far less attention paid to costs borne by families with a child with CD than for families including a child with ASD or ADHD.

Intervening early, as symptoms of conduct disorder become apparent, is recommended and has been shown to save costs downstream. The economic evidence to support parenting programmes for CD is positive but limited (Charles, Bywater, & Edwards, 2011), and as with ADHD, we know little about the longer term outcomes.

Anxiety and depression

Introduction. There are fewer articles reporting cost-related aspects of depression and anxiety than for the three disorders considered so far. Lynch and Clarke (2006) found only five articles containing data up to April 2005, four from the United States and one from the United Kingdom. They concluded that there was insufficient evidence to estimate the economic burden of depression in children and adolescents. Today, the picture is only a little more encouraging. The search identified only one article providing a cost of illness estimate for young people with anxiety and two estimating the costs of supporting those with depression (see Table 4). Three studies describing the impacts of depression into young adulthood were also identified, as well as four cost-effectiveness evaluations published since 2007, two of which came from the same study.

Results. A Dutch cost of illness study focused on 118 anxious young people aged 8–18 who were referred to community mental health services. Mean annual costs per child were \notin 2,748 per annum for direct health care, direct nonmedical care, and indirect and out-of-pocket expenses (2003 prices;

Bodden, Dirksen, & Bögels, 2008). Inpatient care, day treatment, and parental productivity losses each contributed around a quarter to the total costs. The annual national cost was calculated to be €20,294,000. These data formed the baseline of a cost-effectiveness study, the results of which favoured individual cognitive behaviour therapy (CBT) over family CBT 1 year after treatment, mainly due to slightly (but not statistically significant) improved outcome scores (Bodden, Dirksen, Bögels, Nauta et al., 2008). In the analysis of the British Birth Cohort Surveys, Knapp et al. (2011, see above) found childhood anxiety problems to be associated with lower income at age 30 but not with being

employed or occupational status. The costs of supporting adolescents with depression in the United States were also estimated using baseline data from a trial (TADS, see below). Service use data were collected from individual participants and their families over a 3-month period (Domino, Burns, Mario et al., 2009). With the wide population coverage of andministrative and insurance-based databases, such an approach is rare in the United States, but does mean that a broad public sector prespective can be taken – here, health and mental health care, social care, education and the justice system – as well as family costs. Annual costs per family, estimated by multiplying the reported 3-month cost by four, were \$1,124 (2003 prices).

Major depressive disorders (MDD) in childhood have both health and economic impacts in adulthood, as findings from four recent studies show. A longitudinal study in the United States showed that while controlling for current depression (at age 20), those diagnosed with depression at age 15 had higher health care use, poorer self-perceived general health and increased levels of work impairment (Keenan-Miller, Hammen, & Brennan, 2007). Smith and Smith (2010) also highlighted the link between childhood psychological conditions and physical health in adulthood. Their analysis of the US Panel Study of Income Dynamics showed that after controlling for physical ailments in childhood and family members, and neighbourhood characteristics, both educational achievement and family income were lower for those who had a childhood psychological condition.

Similar findings come from analysis of data on a cohort of young adults born in New Zealand in 1977 and participating in the Christchurch Health and Development Study (Fergusson, Boden, & Horwood, 2007). Thirty-five per cent of the cohort met the criteria for at least one episode of major depression between 16 and 21 years old. By age 21–25, and after adjusting for family and personal characteristics, increased frequency of depressive episodes was significantly associated with poorer mental health outcomes (depression, anxiety and suicidal behaviour), poorer educational achievement, and a greater likelihood of being dependant on welfare payments and being unemployed.

Progress in economic studies across different disorders 11

Information about cost-effective interventions in childhood, which may mitigate poor child and adult outcomes, is rare with just two recent studies, each comparing cognitive behavioural therapy (CBT) and medication.

The UK-based trial randomly assigned 208 adolescents (11–17 years) to clinical care plus additional selective serotonin reuptake inhibitors (SSRI, mainly fluoxetine) or clinical care plus SSRIs plus 19 sessions of CBT (Byford, Barratt, Roberts, Wilkinson et al., 2007). Costs across the public sector were measured, plus travel costs and productivity losses for the intervention sessions (2003–2004 prices). At 28 weeks, there were no significant differences in clinical outcomes, QALYs (from the EQ-5D) or costs. The cost-effectiveness analysis suggested that the probability of SSRI+CBT being more cost-effective than SSRIs alone is very small.

However, the US-based trial for adolescent depression (TADS) came to a different conclusion. This RCT compared fluoxetine, CBT, and a combined intervention of CBT and fluoxetine in 327 adolescents age 12–18. Depression-free days were estimated and costs included the intervention and health care, caregiver reports of other health care, school-based counselling, criminal justice and social care services, and caregiver time and travel for the intervention (2003 prices). At 12 weeks, the results favoured treatment by fluoxetine alone (Domino et al., 2008), but at 36 weeks, the analysis showed SSRI+CBT to be the more cost-effective option (Domino, Burns, Silva et al., 2008; Domino, Foster et al., 2009).

Conclusion. Comparing the 2012 US\$ costs for supporting children and adolescents with depression (two studies; see Table 4) with costs shown in the earlier tables, this disorder appears to be absorbing fewer health and mental health care resources, and is less costly to the education system. Direct health care costs for anxiety (one study) are closer to costs for ADHD and CD. However, these disorders are less well served by cost-related research than ASD, CD or ADHD, although both have implications for poorer adult health and quality of life. The question of how best to use resources to treat these disorders in childhood also remains unanswered, even when considering medication (see also, Schlander, 2010; p2455). This absence of research evidence hampered provision of value for money advice to health policy makers in Australia and the authors had to draw on adult studies for some parameters in their analysis of childhood depression (p. e724, Mihalopoulos, Vos, Pirkis, & Carter, 2012).

Other disorders

Introduction. The four disorder groups considered so far absorb the majority of the current literature on

cost-related research. In this section, cost papers relating to eating disorders (five journal articles), psychosis (two articles) and bipolar disorder (one article) are summarised.

Results. A recent UK estimate for the health care associated with eating disorders is costs £80 m-£100 m, accompanied by GDP reductions of between £0.23bn and £2.9bn (Pro Bono Economics & beat, 2012). Baseline data from a randomised controlled trial showed that the public sector costs associated with supporting 85 young people with bulimia nervosa and eating disorder not-otherwise specified (EDNOS) were around £700 over 3 months (2003-2004 prices; Schmidt et al., 2007, 2008). Subsequent family therapy was found to cost almost twice as much as self-guided care, but 12 months later, there was no difference in either outcome (bingeing) or support costs between the two intervention groups, although public sector costs had reduced for both groups (Schmidt et al., 2007) and those with EDNOS responded less well to treatment (Schmidt et al., 2008).

An internet-delivered intervention for young people with bulimic symptoms showed positive outcomes, some reduction in service contacts and reduced family expenditure (Pretorius et al., 2009). Again incorporating newer technologies, telemedicine CBT for young people with bulimia nervosa living in the United States generated similar outcomes for young people as face-to-face CBT but at a reduced cost (Crow et al., 2009). Using a narrow cost perspective (initial evaluation, laboratory tests and psychotherapy visits), the cost per recovered (abstinent) person was \$9,325 for usual care and \$7,300 for telemedicine (2005 prices). A recent review suggests that tele-psychiatry has the potential to be a useful treatment option but that questions remain about confidentiality, legality, reimbursement and cost-effectiveness (Paing et al., 2009).

The final study of eating disorder services compared inpatient and outpatient treatment and general CAMHS and specialist eating disorder services in the United Kingdom. There were no outcome differences between the treatment options for these adolescents with anorexia nervosa (n = 167), but higher costs for those using generalist CAMHS services, mainly because of increased hospital admissions (Byford, Barratt, Roberts, Clark et al., 2007). Specialist outpatient care proved to be the 'dominant' option when comparing incremental cost-effectiveness ratios, supporting the current clinical guidelines for England.

Turning to interventions for psychosis, two modelling studies suggest that a change in the way services were delivered to children and adolescent with psychosis would save money. Using a narrow cost perspective (inpatient care and the early intervention service), a UK-based decision-analytic model identified that cost-savings of £4,814 per young person over 6 months were an early intervention service to replace standard care (2010 prices; McCrone et al., 2013). Wong et al. (2011) used a matched group design to show that outcome were similar for a new early assessment service in Hong Kong (n = 65 young people), but offered 35% savings per one point improvement on a clinical scale (PANNS) over the previous service. Finally, one US study on bipolar disorder among children and adolescents (n = 4,973) found the annual hospital and pharmacy costs to equal that for adults; 71% of the total was for mental health care with psychotropic medications absorbing about a third of that cost (Dusetzina et al., 2012).

Conclusion. It is difficult to formulate any overall conclusions from such a small disparate set of studies. However, most of these are recent publications suggesting that, although lagging behind research for other disorders, this may represent the start of an improved evidence base.

Discussion

The first JCPP review of economics and child and adolescent mental health was published more than 15 years ago (Knapp, 1997) and since then, there have been three reviews of economic evaluations in child and adolescent mental health published in peer-review journals. Looking at the literature from 1966 to 2003, Romeo et al. (2005) identified 21 studies that measured both costs and outcomes, although seven did not report individual-level outcomes; nine of these studies were published after 1996. Seven of the 14 studies published between 1992 and 2004 and discussed in a review of mental health promotion and mental illness prevention focused on children and adolescents (Zechmeister et al., 2008). A third systematic review of economic evaluations of interventions published between 2001 and 2009 found 12 studies using primary data and seven that developed simulation models (Kilian et al., 2010). The general conclusion from these reviews was that although the number of economic evaluations was rising, studies suffered from small sample sizes, short time horizons and narrow cost perspectives. For simulation models, there was a lack of clarity in methods and too many assumptions made in the absence of supporting evidence. The most recent of these reviews (Kilian et al., 2010) suggests that there is still a continuing dependence on a narrow payer-only perspective, but that the quality of papers is improving.

In their review of the benefits and costs of prevention (papers published up to 2007), O'Connell et al. (2009) arrive at a similar conclusion. They also suggest that to develop the limited evidence base, guidelines might improve the quality of economic

evaluations, and help ensure that an economic evaluation always accompanies an effectiveness study. Four other focussed reviews of economic evaluations have been published since 2009: two considered the economic evidence for parenting programmes to intervene in childhood behaviour problems (Charles et al., 2011; Stevens, 2012); and two reviewed studies of medication (Schlander, 2010; Wu et al., 2012). All four of these reviews found full cost-effectiveness analyses to be rare and methodological problems to be common.

This review has collated the data from studies published in peer-reviewed journals since 2005 with the aim of identifying support costs, adult consequences, and cost-effective child and adolescent interventions. The wider aims of this paper led to the overlap in dates with the earlier reviews. In this discussion, the focus is on studies published from 2009, including two identified in the most recent review (Kilian et al., 2010).

From 2009 to 2012, this review identified seven studies of costs and outcomes (analyses concerned only with medication are excluded), including three modelling studies, plus five analyses looking at the longer term consequences of childhood mental health problems. Seventeen papers reported the costs of treating and supporting children with psychiatric disorders, predominantly focussing on health care costs. Studies using data from the United States (n = 39) and the United Kingdom formed the majority and there were just three articles concerning low- or middle-income countries (Kawakami et al., 2012; Lee et al., 2009; Lund et al., 2009). Limiting the search to English language papers may have contributed to this bias; however, Kilian et al. (2010) revealed a similar geographical spread but had no language restrictions, as long as there was an English title on the database. This review identified just one additional paper from Germany that had an English title and abstract (Schlander, Trott, & Schwarz, 2010).

Notably, the search identified only one full cost-effectiveness study published since 2009, evaluating medication and CBT for children and adolescents with depression. The previous service use and cost paper and shorter term cost-effectiveness evaluation from this study added to a very limited evidence base on support and interventions for adolescent depression in the United States. The UK-based cost-effectiveness evaluation of medication and CBT fulfils a similar role (Byford, Barratt, Roberts, Wilkinson et al., 2007). Two cost-effectiveness studies examining similar treatments for similar conditions are to be welcomed, but this also serves to highlight gaps in the current evidence base and the difficulties of linking individual studies. In the first place, medication and CBT are only two of a range of treatments available; for example, no cost-effectiveness analyses were found in a review of psychotherapy interventions for depressed children and adolescents (Watanabe, Hunot, Omori, Churchill, & Furukawa, 2007). That is not to say that these treatments are not more (or less) cost-effective than other options, just that as yet there is no evidence either way. In the second place, these two studies of medication and CBT are not fully comparable because although the methods are robust and clearly stated, the cost perspective is different, the studies use different outcome measures, there are potential differences in the intervention, etc. And third, there are differences between the countries' health care organisation and financing systems; these make it very difficult to compare cost-effectiveness findings between countries. To come to a definitive view on relative cost-effectiveness, data from many more trials are needed for meta-analysis or analysis of combined individual-level data sets (see, for example, Bower et al., 2003; Foster, Jensen et al., 2007). This review also uncovered five trial protocols that included an economic evaluation, although none comparing medication and CBT. Other trials, of course, may well be underway, but not have published their protocol in the areas searched.

Ten articles published since 2009 report the costs of supporting or treating groups of children and adolescents with various disorders, one of which drew on existing literature. Cost of illness studies need good quality diagnostic or epidemiologic information and details of resources (services) used, as well as (unit) costs for those services. Each of the studies reported here drew on robust diagnostic instruments to ensure that the appropriate populations were included. However, the World Health Organisation found there was no meaningful epidemiologic information for about two thirds of the 192 countries worldwide, yet up to 20% of children and adolescents may suffer from a disabling mental illness (Belfer, 2008).

Among the nine data-based cost of illness studies, there are, broadly speaking, two types. On the one hand, there are costs data derived from larger surveys, routine data collections or claims' databases (four papers); on the other are costs calculated from studies where primary, usually prospective, data are collected for a specific sample (five papers). The division is not always as clear-cut as this of course, but both broad methods have their positive attributes, and both have their limitations. For example, large survey and databases are more likely to provide a representative sample of children, but they also tend to be more limited in the scope of resource use that is measured and thus have a narrower cost perspective. Commonly, these studies focus on health care service use and costs, although databases for education and justice systems have also proved useful.

Primary data tends to come from smaller, often prospective, studies. They often include more detailed information on a wider range of service and

supports (although this can be for a closely specified sample) allowing a broad perspective to be presented in the cost calculations. Findings from these studies show that for children and adolescents with psychiatric disorders, it is very rare for the health sector to bear a large proportion of total support costs. Where all public sector costs are considered, it is common for the largest proportion to fall to the justice system in studies of adolescents and to the education system for younger children. However, where a societal perspective is taken and family-borne costs are included, by far the largest cost impact is accounted for by the value of work absence. Assessing the extent of support received from informal care (time parents spend caring in particular) is particularly difficult in evaluations of children's services - when does parental care end and additional support due to the disorder begin? There are additional questions about measuring the impact of a child's disorder on the family. For example, should the costs of additional services used by parents and siblings be included? When estimating the costs of parental lost productivity, should the study also look at how the child's problems changed parents' employment trajectories?

There are also differences in the way data on service use are collected: claims' databases or management information systems collate data from services, other studies (surveys, smaller prospective studies) often rely on parents' recall, and occasionally interview the young person. Comparisons between the two methods have shown mixed results with greater concordance for larger items (such as hospital care) and for shorter time spans. For all the above reasons, it is very difficult to compare the costs of supporting or treating children across studies. Moreover, the studies discussed in this review have such an array of participant needs, perspectives and approaches to (unit) costs that simple comparisons can confuse rather than clarify. One common finding was that almost all studies found cost data to be highly skewed across the sample. Most children and adolescents have little contact with services (low costs), and in any sample, there are usually a few participants who use services intensively and generate high costs. Mean costs provide useful information for planners, but cost variation analyses can help providers manage their service appropriately and can highlight equity issues (see, for example, Howell & Teich, 2008). Where such analyses have been undertaken, it is rare for the proportion of cost variation statistically 'explained' to reach more than 30%, but there are some cost associations that appear reasonably frequently: age, severity of condition, comorbidity, whether a community or clinic sample and some family characteristics related to deprivation.

Three of the papers published since 2009 report adult economic outcomes for childhood disorders. Studies that estimate longer term morbidity and economic outcomes commonly use longitudinal studies or population surveys from which certain groups are selected. For child and adolescent mental health problems, subsequent adult impacts can generally be seen in their poorer health, quality of life and family life, as well as negative impacts on employment status and income.

Findings such as these can indicate where intervention in childhood and adolescence is needed to try to avert poor outcomes, whereas modelling studies (four since 2009) more commonly aim to show that investing money today in particular services will save money to the public purse or society in the future. Such models are often developed because there is not enough evidence from primary research to draw firm conclusions about longer term cost-effectiveness. Generally speaking, in the papers described here, methods were clearly stated and data sources identified, although the cost perspective was very narrow in two of the studies. However, the challenges in generating these savings in the real world are not necessarily to do with putting these services in place, because the models are usually based on innovative services already in existence. The challenges are more about the ability to predict which individuals are 'at risk' (need the services) and will benefit most, targeting the service on these groups, and in ensuring that those who attend receive an effective 'dose' of the service.

This paper has attempted to do something rather different from earlier reviews of economic evaluations, which have assessed studies that measure both costs and outcomes of child and adolescent mental health interventions. As well as identifying these cost and outcome analyses, this paper aimed to pull together the information on the costs of support and treatment of child and adolescent psychiatric disorders and their longer term (adulthood) costs. Measures commonly employed to assess the quality of studies, such as the BMJ guidelines for economic evaluations, are less helpful because the intention is to range across different types of economic studies. Generally speaking, this has brought forth a slightly more rosy view of the quantity and quality of the economic studies. Taken overall, these studies have reported their methods and cost perspective in a good depth of detail. Some studies continue to use a narrow perspective, but others are broadening the view to include nonhealth care providers. They are also beginning to highlight the high cost to parents - not just due to absence from work but also the costs that families bear for providing additional care and support to a child with mental health problems. Analytic approaches are generally well described, although lack of space means that the details cannot be replicated here. Similarly, many papers explore cost or other variations within a population sample; these complex analyses generate far finer level of results and findings than would fit into this article.

Conclusion

The demands for economic evaluation remain the same today as when described 15 years ago (Knapp, 1997). Prevalence of child and adolescent disorders continues to grow whether in real terms - perhaps because of wider cultural, social or economic changes - or due to improved detection (Collishaw, Maughan, Goodman, & Pickles, 2004; McClellan, Bresnahan, Echeverria, Knox, & Susser, 2009; McCrone, Dhanasiri, Patel, Knapp, & Lawton-Smith, 2008; Sourander et al., 2005). Wider economic pressures and government cutbacks are a major challenge for commissioners and service providers in the United States and almost every European country. The policy and practice backdrop is one of continually developing health care systems and markets in which there is an increasing need to show value for money. This means that there is still a need for better economic evidence to inform policy and practice.

There is no doubt that the evidence base is improving and while the articles published post-2005, and particularly the post-2009 articles, have provided better methodological information, this should not lead to complacency. The publication rate for cost-related articles in child and adolescent psychiatry is about 10 per year since 2009. Although only a handful of these would meet the criteria for the earlier reviews, this is good progress. However, when looking through the lens of specific disorder groups, the resulting evidence base is perhaps less encouraging. For example, it has only been possible to identify more than a handful of published journal articles for four disorders: autism spectrum disorder, attention deficit hyperactivity disorder, conduct disorder and depression. Moreover, most of the studies, including those that identify impacts in adulthood, refer to the US or British context. There is, therefore, little additional information to help address the questions identified in 1997: we still do not know enough about what support and treatment should be provided; when, where or to whom it should be provided; or how support and treatment should be provided. Despite the increase in cost-related studies and the progress made over the last decade, a considerable amount of research is still needed before there is a sound evidence base to inform decisions on how best to use scarce resources to support and treat children with mental health problems.

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Key Points

- There are far fewer economic studies for child and adolescent psychiatric disorders than for adult disorders. Earlier reviews have found the quality of studies that combine costs and outcomes to be poor.
- Costs falling to nonhealth agencies such as those providing social care, education, justice and family costs can be high, but are rarely assessed. This wide perspective matches the breadth of consequences that childhood psychiatric disorders can have.
- High nonhealth costs in childhood and poor economic outcomes in adulthood make a strong case for increased investment in treatment for childhood psychiatric disorders.
- Many more good quality economic studies, including cost-effectiveness analyses of new and existing interventions, are required to provide a sufficient body of evidence to help commissioners and providers make decisions about resource allocation.

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