Authors' objectives
To determine the efficacy and safety of vigabatrin in the treatment of infantile spasms in infants suffering from tuberous sclerosis complex.

Searching
A search going back to 1990 was conducted of MEDLINE and EMBASE for articles published in the English language. References of retrieved studies were reviewed.

Study selection
Study designs of evaluations included in the review
Studies that investigated the effects of vigabatrin on infantile spasms were included if they gave a breakdown of results for the effect in patients with tuberous sclerosis. Included studies were of the following designs: single blind; open prospective and retrospective.

Specific interventions included in the review
Vigabatrin in doses ranging from 20 to 400 mg/kg/day was studied.

Participants included in the review
Infants with infantile spasms with either a known underlying diagnosis of tuberous sclerosis complex or other causes or unknown causes were included. The age of infants (where stated) ranged from 4 to 26 months in age.

Outcomes assessed in the review
Outcomes assessed included: number with cessation of spasms considered by diagnosis (tuberous sclerosis or other); time taken to achieve cessation of spasms; and adverse reactions.

How were decisions on the relevance of primary studies made?
The authors do not state how the papers were selected for the review, or how many of the authors performed the selection.

Assessment of study quality
The authors do not state that they assessed validity.

Data extraction
The authors do not state how the data were extracted for the review, or how many of the authors performed the data extraction.

Methods of synthesis
How were the studies combined?
An overall average response rate was calculated for those with and without tuberous sclerosis.

How were differences between studies investigated?
The authors do not state how differences between the studies were investigated.

Results of the review
Eight studies were included (390 infants of whom 77 had a diagnosis of tuberous sclerosis complex).

Overall complete cessation of spasms occurred in 242 infants (62%).

Cessation of spasms occurred in 73 of 77 infants (95%) with tuberous sclerosis and in 169 of 313 infants (54%) without tuberous sclerosis.

Time taken to achieve cessation of spasms in those responding was reported as averaging 4 days (2 studies), within 7 days (1 study) and within 8 days (1 study) and within a month (1 study).

Adverse reactions (7 studies): a total of 87 adverse events were reported in 373 infants. The majority were transient and none continued after stopping vigabatrin. Adverse reactions resulted in 7 infants having vigabatrin withdrawn. Reasons for withdrawal were hypertonia, hypotonia, hyperactivity, irritability, and myoclonus. No deaths were attributed to vigabatrin. Other side effects included: drowsiness, behavioural problems, insomnia, diarrhoea, weight gain, and vomiting.

Authors' conclusions
Vigabatrin should be considered as first-line monotherapy for the treatment of infantile spasms in those with either a known diagnosis of tuberous sclerosis or who are at high risk of tuberous sclerosis such as those with a first degree relative with tuberous sclerosis complex. Paradoxically, in those without tuberous sclerosis complex, vigabatrin might be less efficacious than suggested by studies including patients with tuberous sclerosis complex.

CRD commentary
The aims and inclusion criteria were stated. Some relevant details of primary studies were clearly presented in tabular format. Side effects were included.

By limiting studies to those published in the English language identified from two databases some other relevant studies may have been omitted. No details were given of the methods used to select primary studies or extract data. No mention was made of diagnostic criteria used to define either tuberous sclerosis complex or infantile spasms in the primary studies. Neither validity nor heterogeneity were assessed. No mention was made of the duration of follow up.

In view of these limitations the authors conclusions cannot be supported.

Implications of the review for practice and research
Practice: The authors consider that vigabatrin should be considered the drug of choice for patients with infantile spasms and either a known diagnosis of tuberous sclerosis or who are at high risk of tuberous sclerosis complex before investigations are complete.

Research: The authors consider that more information is required on the incidence of both asymptomatic and symptomatic visual field defects in patients treated with vigabatrin and on the treatment of infantile spasms.

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Record Status
This is a critical abstract of a systematic review that meets the criteria for inclusion on DARE. Each critical abstract contains a brief summary of the review methods, results and conclusions followed by a detailed critical assessment on the reliability of the review and the conclusions drawn.