Supple 5.1 Previous population-based studies on trisomy 18 syndrome survival times





Supple 5.2 Flow chart of the selection process

Search strategy (MEDLINE):

1. Exp Trisomy 18 Syndromes/
2. ("Trisomy 18 Syndromes" or "Trisomy 18" or "Trisomy E Syndrome" or "Edwards Syndrome").tw.
3. 1 or 2

Records identified from initial searches (Apr. 2019)

MEDLINE (n = 2,498)

Initial exclusion of the reviews after title and abstract examination

(n = 2,457)

Full-text review articles examined for potential inclusion

(n = 2,500)

Full-text review articles excluded with reasons

(n = 20) Wrong outcome

(n = 7) Wrong population

(n = 12) Not population-based

(n = 39)

Total reviews included for synthesis

(n = 21)

Full-text review articles examined for potential inclusion

(n = 60)

Additional records

Hand search (n = 2)

Supple 5.3 List of excluded reviews with reasons (full-text screening)

|  |  |
| --- | --- |
| Studies | Reasons |
| Alberman, E. and Mutton, D. and Morris, J. K. (2012) Cytological and epidemiological findings in trisomies 13, 18, and 21: England and Wales 2004-2009.American Journal of Medical Genetics. Part A;158(5):1145-50. | Wrong outcome (no survival data) |
| Anneren, G. (1986) Incidence of trisomy 18 in Sweden in 1981-1982: clustering in a small region.Clinical Genetics;29(4):345-7. | Wrong outcome |
| Canfield, M. A. and Honein, M. A. and Yuskiv, N. and Xing, J. and Mai, C. T. and Collins, J. S. and Devine, O. and Petrini, J. and Ramadhani, T. A. and Hobbs, C. A. and Kirby, R. S.(2006)National estimates and race/ethnic-specific variation of selected birth defects in the United States, 1999-2001.Birth Defects Research;76(11):747-56. | Wrong outcome |
| Canfield, M. A. and Mai, C. T. and Wang, Y. and O'Halloran, A. and Marengo, L. K. and Olney, R. S. and Borger, C. L. and Rutkowski, R. and Fornoff, J. and Irwin, N. and Copel and , G. and Flood, T. J. and Meyer, R. E. and Rickard, R. and Alverson, C. J. and Sweatlock, J. and Kirby, R. S. and National Birth Defects Prevention, Network(2014)The association between race/ethnicity and major birth defects in the United States, 1999-2007.American Journal of Public Health;104(9):e14-23. | Wrong outcome |
| Carothers, A. D. (1994) A cytogenetic register of trisomies in Scotland: results of the first 2 years (1989, 1990).Clinical Genetics;46(6):405-9. | Wrong outcome |
| Chen, B. Y. and Hwang, B. F. and Guo, Y. L. (2009) Epidemiology of congenital anomalies in a population-based birth registry in Taiwan, 2002.Journal of the Formosan Medical Association;108(6):460-8. | Wrong outcome |
| Crider, K. S. and Olney, R. S. and Cragan, J. D. (2008) Trisomies 13 and 18: population prevalences, characteristics, and prenatal diagnosis, metropolitan Atlanta, 1994-2003.American Journal of Medical Genetics. Part A;146(7):820-6. | Wrong outcome |
| Emanuel, I. and Huang, S. W. and Chiang, W. T. and Yang, C. P. (1970) Trisomy 18 syndrome in Chinese infants. Clinical findings and incidence. Journal of Medical Genetics;7(2):138-41. | Wrong outcome |
| Farag, T. I. and Al-Awadi, S. A. and Al-Othman, S. A. and Sundareshan, T. S. and Krishna Murthy, D. S. and Usha, R. and Madg, S. A. and Uma, R. (1988) Down syndrome and trisomy 18 in the Bedouins. American Journal of Medical Genetics;29(4):943-4. | Wrong outcome |
| Forrester, M. B. and Merz, R. D. (2007) Potential impact of pregnancy outcome on the completeness of diagnosis of birth defects, Hawai'i, 1986-2001.Hawaii Medical Journal;66(2):32, 34-5. | Wrong outcome |
| Garfinkel, J. and Porter, I. H. (1971) Trisomy 18 in New York State.Lancet;2(7739):1421-2. | Wrong outcome |
| Goud, M. T. and Al-Harassi, S. M. and Al-Khalili, S. A. and Al-Salmani, K. K. and Al-Busaidy, S. M. and Rajab, A. (2005) Incidence of chromosome abnormalities in the Sultanate of Oman.Saudi Medical Journal;26(12):18810. | Wrong outcome |
| Haghnazarian, E. and Hu, J. and Song, A. Y. and Friedlich, P. S. and Lakshmanan, A. (2019) The association of Trisomy 13 and 18 and hospital discharge outcomes among neonates in California: A retrospective cohort study. Pediatrics & Neonatology;2():2. | Wrong outcome |
| Hartman, R. J. and Rasmussen, S. A. and Botto, L. D. and Riehle-Colarusso, T. and Martin, C. L. and Cragan, J. D. and Shin, M. and Correa, A. (2011) The contribution of chromosomal abnormalities to congenital heart defects: a population-based study. Pediatric Cardiology;32(8):1147-57.  | Wrong outcome |
| Kirby, R. S.(2017)The prevalence of selected major birth defects in the United States. Seminars in Perinatology;41(6):338-344. | Wrong outcome |
| Nair, D. B. and Tucker, D. and Hughes, R. and Greenacre, J. and Morgan, M. (2015)Unusual trend in the prevalence of trisomy 13 in mothers aged 35 and older: A population based study of national congenital anomaly data.Birth Defects Research;103(7):610-6. | Wrong outcome |
| Pakkasjarvi, N. and Koskimies, E. and Ritvanen, A. and Nietosvaara, Y. and Makitie, O. (2013) Characteristics and associated anomalies in radial ray deficiencies in Finland--a population-based study. American Journal of Medical Genetics. Part A;161(2):261-7. | Wrong outcome |
| Parker, S. E. and Mai, C. T. and Canfield, M. A. and Rickard, R. and Wang, Y. and Meyer, R. E. and Anderson, P. and Mason, C. A. and Collins, J. S. and Kirby, R. S. and Correa, A. and National Birth Defects Prevention, Network(2010)Updated National Birth Prevalence estimates for selected birth defects in the United States, 2004-2006.Birth Defects Research;88(12):1008-16. | Wrong outcome |
| Springett, A. and Wellesley, D. and Greenlees, R. and Loane, M. and Addor, M. C. and Arriola, L. and Bergman, J. and Cavero-Carbonell, C. and Csaky-Szunyogh, M. and Draper, E. S. and Garne, E. and Gatt, M. and Haeusler, M. and Khoshnood, B. and Klungsoyr, K. and Lynch, C. and Dias, C. M. and McDonnell, R. and Nelen, V. and O'Mahony, M. and Pierini, A. and Queisser-Luft, A. and Rankin, J. and Rissmann, A. and Rounding, C. and Stoianova, S. and Tuckerz, D. and Zymak-Zakutnia, N. and Morris, J. K.(2015)Congenital anomalies associated with trisomy 18 or trisomy 13: A registry-based study in 16 European countries, 2000-2011.American Journal of Medical Genetics. Part A;167(12):424657. | Wrong outcome |
| Tonks, A. M. and Gornall, A. S. and Larkins, S. A. and Gardosi, J. O. (2013)Trisomies 18 and 13: trends in prevalence and prenatal diagnosis - population based study. Prenatal Diagnosis;33(8):742-50. | Wrong outcome |
| Boghossian, N. S. and Hansen, N. I. and Bell, E. F. and Stoll, B. J. and Murray, J. C. and Carey, J. C. and Adams-Chapman, I. and Shankaran, S. and Walsh, M. C. and Laptook, A. R. and Faix, R. G. and Newman, N. S. and Hale, E. C. and Das, A. and Wilson, L. D. and Hensman, A. M. and Grisby, C. and Collins, M. V. and Vasil, D. M. and Finkle, J. and Maffett, D. and Ball, M. B. and Lacy, C. B. and Bara, R. and Higgins, R. D. and Eunice Kennedy Shriver National Institute of Child, Health and Human Development Neonatal Research, Network(2014)Mortality and morbidity of VLBW infants with trisomy 13 or trisomy 18.Pediatrics;133(2):226-35. | Wrong population(VLBW) |
| Boghossian, N. S. and Horbar, J. D. and Carpenter, J. H. and Murray, J. C. and Bell, E. F. and Vermont Oxford, Network (2012)Major chromosomal anomalies among very low birth weight infants in the Vermont Oxford Network. Journal of Pediatrics;160(5):774-780.e11. | Wrong population (VLBW) |
| Domingo, L. and Carey, J. C. and Eckhauser, A. and Wilkes, J. and Menon, S. C. (2019)Mortality and Resource Use Following Cardiac Interventions in Children with Trisomy 13 and Trisomy 18 and Congenital Heart Disease. Pediatric Cardiology;40(2):349-356. | Wrong population (underwent cardiac intervention) |
| Donovan, J. H. and Krigbaum, G. and Bruns, D. A.(2016)Medical interventions and survival by gender of children with trisomy 18.American Journal of Medical Genetics. Part C, Seminars in Medical Genetics;172(3):272-8. | Wrong population (children living 2 months or longer) |
| Ishitsuka, K. and Matsui, H. and Michihata, N. and Fushimi, K. and Nakamura, T. and Yasunaga, H. (2015)Medical procedures and outcomes of Japanese patients with trisomy 18 or trisomy 13: analysis of a nationwide administrative database of hospitalized patients. American Journal of Medical Genetics. Part A;167(8):1816-21. | Wrong population (hospitalized patients) |
| Laberge, J. M. and Flageole, H. and Pugash, D. and Khalife, S. and Blair, G. and Filiatrault, D. and Russo, P. and Lees, G. and Wilson, R. D. (2001)Outcome of the prenatally diagnosed congenital cystic adenomatoid lung malformation: a Canadian experience. Fetal Diagnosis & Therapy;16(3):178-86. | Wrong population (Congenital cystic adenomatoid malformation) |
| Peterson, J. K. and Kochilas, L. K. and Catton, K. G. and Moller, J. H. and Setty, S. P. (2017)Long-Term Outcomes of Children With Trisomy 13 and 18 After Congenital Heart Disease Interventions. Annals of Thoracic Surgery;103(6):1941-1949. | Wrong population (after congenital heart defect interventions) |
| Baty, B. J. and Blackburn, B. L. and Carey, J. C. (1994)Natural history of trisomy 18 and trisomy 13: I. Growth, physical assessment, medical histories, survival, and recurrence risk. American Journal of Medical Genetics;49(2):175-88. | Not population-based (parent questionnaires) |
| Burke, A. L. and Field, K. and Morrison, J. J. (2013) Natural history of fetal trisomy 18 after prenatal diagnosis. Archives of Disease in Childhood Fetal & Neonatal Edition;98(2):F152-4. | Not population-based |
| Bruns, D. A. (2010) Neonatal experiences of newborns with full trisomy 18.Advances in Neonatal Care;10(1):25-31. | Not population-based |
| Dereddy, N. R. and Pivnick, E. K. and Upadhyay, K. and Dhanireddy, R. and Talati, A. J. (2017) Neonatal Hospital Course and Outcomes of Live-born Infants with Trisomy 18 at Two Tertiary Care Centers in the United States. American Journal of Perinatology;34(3):270-275. | Not population-based |
| Hsiao, C. C. and Tsao, L. Y. and Chen, H. N. and Chiu, H. Y. and Chang, W. C. (2009) Changing clinical presentations and survival pattern in trisomy 18.Pediatrics & Neonatology;50(4):147-51. | Not population-based |
| Kato, E. and Kitase, Y. and Tachibana, T. and Hattori, T. and Saito, A. and Muramatsu, Y. and Takemoto, K. and Yamamoto, H. and Hayashi, S. and Yasuda, A. and Kato, Y. and Ieda, K. and Oshiro, M. and Sato, Y. and Hayakawa, M. (2019) Factors related to survival discharge in trisomy 18: A retrospective multicenter study. American Journal of Medical Genetics. Part A;3():3. | Not population-based |
| Morallo, L. M. and Rosenblum, H. and Esterly, K. L. and Johnson, W. D. and Storlazzi, J. J. and Narvaez, A. C. and Borgaonkar, D. S. (1982) Trisomy 18 (Edwards syndrome) in Delaware. Delaware Medical Journal;54(12):673-5. | Not population-based (case report) |
| Morallo, L. M. and Rosenblum, H. and Esterly, K. L. and Johnson, W. D. and Storlazzi, J. J. and Narvaez, A. C. and Borgaonkar, D. S. (1983)Trisomy 18 (Edwards syndrome) in Delaware. Delaware Medical Journal;55(1):27-31. | Not population-based (case report) |
| Naguib, K. K. and Al-Awadi, S. A. and Marafie, M. J. and Al-Hijji, S. Y. (1987)Trisomy 18 clustering in Kuwait. Clinical Genetics;32(6):379-82. | Not population-based (one center) |
| Naguib, K. K. and Al-Awadi, S. A. and Bastaki, L. and Moussa, M. A. and Abulhassan, S. A. and Tayel, S. and Murthy, K. (1999) Clustering of trisomy 18 in Kuwait: Genetic predisposition or environmental? Annals of Saudi Medicine;19(3):197-200. | Not population-based (one center) |
| Naguib, K. K. and Al-Awadi, S. A. and Moussa, M. A. and Bastaki, L. and Gouda, S. and Redha, M. A. and Mustafa, F. and Tayel, S. M. and Abulhassan, S. A. and Murthy, D. S. (1999) Trisomy 18 in Kuwait. International Journal of Epidemiology;28(4):711-6. | Not population-based (one center) |
| Weber WW (1967) Survival and the sex ratio in trisomy 17-18. Am J Hum Genet;19(3 Pt 2):369-77. | Not population-based (published sources and personal communications) |