

Valuing the benefits and harms of antenatal and newborn screening programmes in health economic assessments (VALENTIA)

SUPPLEMENTARY MATERIAL 2

Results of thematic framework analysis

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~~Table 1 Thematic framework of benefits and harms adopted by health economic assessments evaluating antenatal and newborn screening programmes in the refresh search from 1st January 2021 to 22nd November 20212~~

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Table 1 Thematic framework of benefits and harms adopted by health economic assessments evaluating antenatal and newborn screening programmes in the refresh search from 1st January 2021 to 22nd November 2021

Theme no.	Theme	Subtheme level 1	Subtheme level 2	Subtheme level 3	Antenatal screening	Newborn screening
1	Diagnosis of screened for condition	born with condition	reduction in children born with condition through termination of pregnancy		1	-
1	Diagnosis of screened for condition	cases diagnosed at screening			2-5	-
1	Diagnosis of screened for condition	cases diagnosed at screening that would have become symptomatic			-	6
1	Diagnosis of screened for condition	cases missed at screening			2	-
1	Diagnosis of screened for condition	confirmatory test and additional tests to reach diagnosis of screened for condition	unnecessary due to false positive		5,7	6,8
1	Diagnosis of screened for condition	confirmatory test and additional tests to reach diagnosis of screened for condition	unnecessary due to false positive		2,9	-

Theme no.	Theme	Subtheme level 1	Subtheme level 2	Subtheme level 3	Antenatal screening	Newborn screening
	condition	tests to reach diagnosis of screened for condition	positive [invasive]			
1	Diagnosis of screened for condition	screened for condition related complications	morbidity		10	-
2	Life years and health status adjustments	infant life years post birth	DALYs		10	-
2	Life years and health status adjustments	infant life years post birth	morbidity		4,9-11	-
2	Life years and health status adjustments	infant life years post birth	mortality		3,4,11,12	-
2	Life years and health status adjustments	infant life years post birth	QALYs		12	6,8,13
2	Life years and health status adjustments	maternal life years	mortality		7	-
2	Life years and health status adjustments	maternal life years	QALYs		1,5,7,12	-
3	Treatment	comparison of earlier treatment	reduction in treatment	costs	12	-

Theme no.	Theme	Subtheme level 1	Subtheme level 2	Subtheme level 3	Antenatal screening	Newborn screening
		after screen detection and later after symptomatic detection	required			
3	Treatment	comparison of earlier treatment after screen detection	adverse complications of screened for condition averted		7,11,12	-
3	Treatment	hospital stay			4	-
3	Treatment	treatment related harm	adverse reaction to treatment		11	-
3	Treatment	unnecessary due to false positive			3,4,7,9-12	-
6	Pregnancy loss	termination of pregnancy			2,9	-
6	Pregnancy loss	treatment/test related			1,9	-
6	Pregnancy loss	treatment/test related	unaffected		2	-
7	Spillover effects				-	8

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Table 2 Summary of thematic framework of benefits and harms adopted by health economic assessments evaluating antenatal and newborn screening programmes by conditions

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Screening type	Core theme	Developmental	Endocrinology	Genetic	Gestational cardiorenal	Haematology	Infection	Intrauterine fetal demise	Maternal mental health	Metabolic	Neurodevelopment	Neurological	Nutrition	Structural	Urology
Antenatal	Total no. of studies	0 (0%)	24 (100%)	77 (100%)	5 (100%)	18 (100%)	76 (100%)	1 (100%)	1 (100%)	0 (0%)	1 (100%)	0 (0%)	4 (100%)	36 (100%)	1 (100%)
	No themes*	0 (0%)	9 (38%)	24 (31%)	1 (20%)	8 (44%)	22 (29%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	1 (25%)	16 (44%)	0 (0%)
	Theme 1	0 (0%)	8 (33%)	49 (64%)	3 (60%)	7 (39%)	32 (42%)	0 (0%)	1 (100%)	0 (0%)	0 (0%)	0 (0%)	2 (50%)	14 (39%)	1 (100%)
	Theme 2	0 (0%)	13 (54%)	14 (18%)	2 (40%)	4 (22%)	38 (50%)	1 (100%)	1 (100%)	0 (0%)	1 (100%)	0 (0%)	2 (50%)	14 (39%)	0 (0%)
	Theme 3	0 (0%)	11 (46%)	8 (10%)	4 (80%)	7 (39%)	47 (62%)	1 (100%)	1 (100%)	0 (0%)	0 (0%)	0 (0%)	2 (50%)	7 (19%)	1 (100%)
	Theme 4	0 (0%)	4 (17%)	15 (19%)	1 (20%)	3 (17%)	36 (47%)	0 (0%)	0 (0%)	0 (0%)	1 (100%)	0 (0%)	0 (0%)	8 (22%)	1 (100%)
	Theme 5	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	2 (3%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)
	Theme 6	0 (0%)	0 (0%)	43 (56%)	0 (0%)	2 (11%)	2 (3%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	1 (25%)	6 (17%)	0 (0%)
	Theme 7	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)
Newborn	Total no. of	6	4	12	0	12	3	1	6	32	3	1	0	25	0

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<u>Screening type</u>	<u>Core theme</u>	<u>Developmental</u>	<u>Endocrinology</u>	<u>Genetic</u>	<u>Gestational cardiorenal</u>	<u>Haematology</u>	<u>Infection</u>	<u>Intrauterine fetal demise</u>	<u>Maternal mental health</u>	<u>Metabolic</u>	<u>Neurodevelopment</u>	<u>Neurological</u>	<u>Nutrition</u>	<u>Structural</u>	<u>Urology</u>
	<u>studies</u>	(100%)	(100%)	(100%)	(0%)	(100%)	(100%)	(100%)	(0%)	(100%)	(100%)	(100%)	(0%)	(100%)	(0%)
	<u>No themes*</u>	3 (50%)	1 (25%)	1 (8%)	0 (0%)	3 (25%)	0 (0%)	0 (0%)	3 (0%)	9 (28%)	1 (33%)	0 (0%)	0 (0%)	3 (12%)	0 (0%)
	<u>Theme 1</u>	2 (33%)	2 (50%)	8 (67%)	0 (0%)	8 (67%)	1 (33%)	1 (100%)	2 (0%)	22 (69%)	1 (33%)	0 (0%)	0 (0%)	19 (76%)	0 (0%)
	<u>Theme 2</u>	3 (50%)	1 (25%)	8 (67%)	0 (0%)	9 (75%)	0 (0%)	1 (100%)	3 (0%)	19 (59%)	2 (67%)	1 (100%)	0 (0%)	8 (32%)	0 (0%)
	<u>Theme 3</u>	1 (17%)	1 (25%)	3 (25%)	0 (0%)	7 (58%)	1 (33%)	1 (100%)	1 (0%)	14 (44%)	1 (33%)	1 (100%)	0 (0%)	11 (44%)	0 (0%)
	<u>Theme 4</u>	1 (17%)	2 (50%)	5 (42%)	0 (0%)	4 (33%)	1 (33%)	1 (100%)	1 (0%)	17 (53%)	1 (33%)	0 (0%)	0 (0%)	7 (28%)	0 (0%)
	<u>Theme 5</u>	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	3 (9%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)
	<u>Theme 7</u>	0 (0%)	0 (0%)	1 (8%)	0 (0%)	0 (0%)	2 (67%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)

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Theme 1: Diagnosis of screened for condition, Theme 2: Life years and health status adjustments, Theme 3: Treatment, Theme 4: Long-term cost associated with screened for condition, Theme 5: Overdiagnosis, Theme 6: Pregnancy loss, Theme 7: Spillover effects.

*Limited information about benefits and harms could be extracted to inform our bespoke form as the majority in this category were conference abstracts.

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FIGURES

Note: "No themes" in the following figures refers to articles/reports with limited information about benefits and harms that could be extracted to inform our bespoke form.

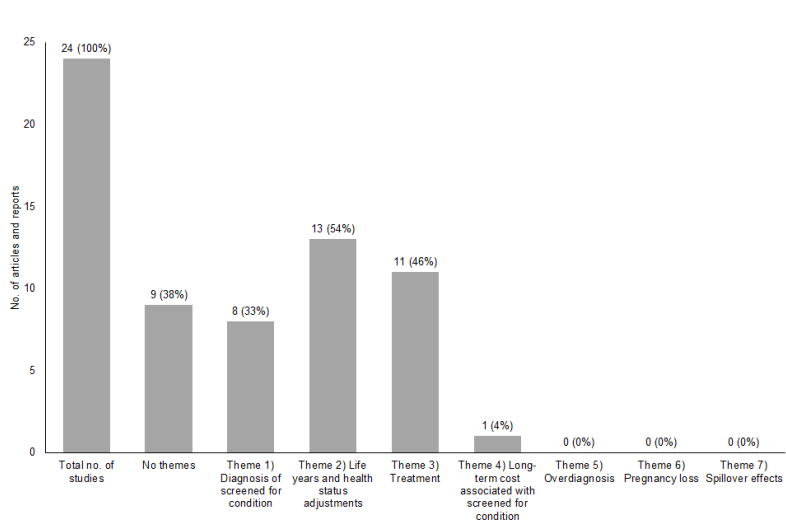


Figure 1 Benefits and harms adopted by health economic assessments evaluating antenatal screening programmes for endocrinological conditions using thematic framework

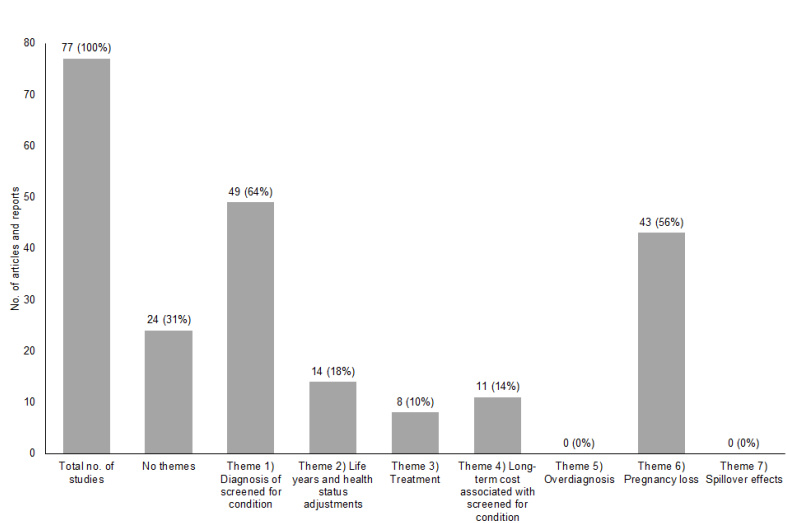


Figure 2 Benefits and harms adopted by health economic assessments evaluating antenatal screening programmes for genetic conditions using thematic framework

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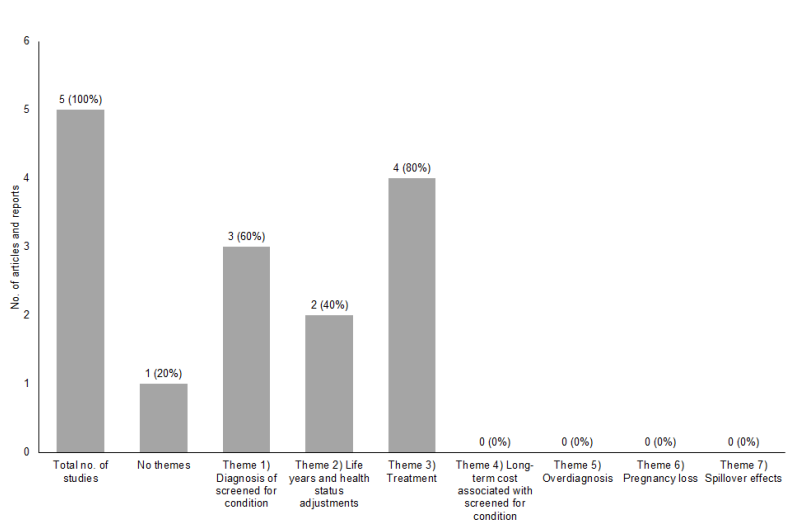


Figure 3 Benefits and harms adopted by health-economic assessments evaluating antenatal screening programmes for gestational cardiorenal conditions using thematic framework

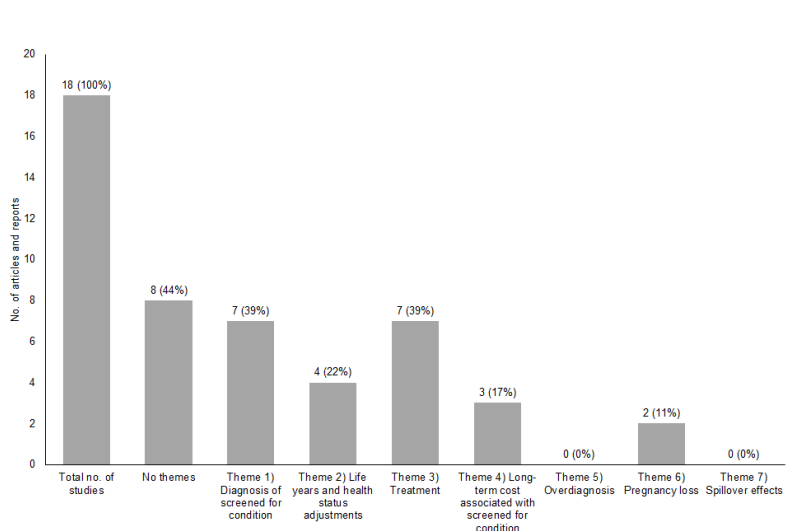
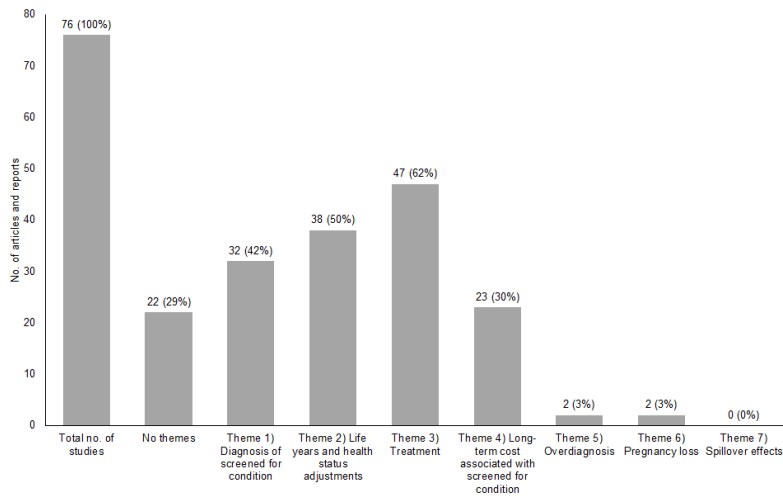


Figure 4 Benefits and harms adopted by health-economic assessments evaluating antenatal screening programmes for haematological conditions using thematic framework

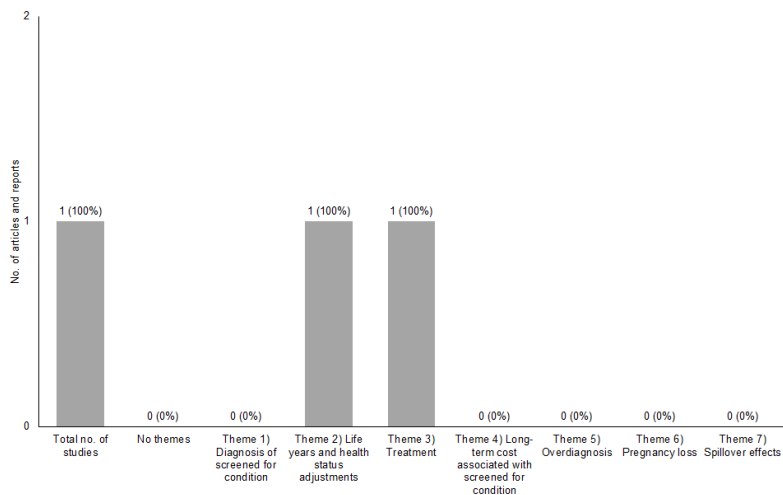
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Figure 5 Benefits and harms adopted by health-economic assessments evaluating antenatal screening programmes for infectious diseases using thematic framework



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Figure 6 Benefits and harms adopted by health-economic assessments evaluating antenatal screening programmes for intrauterine fetal demise using thematic framework

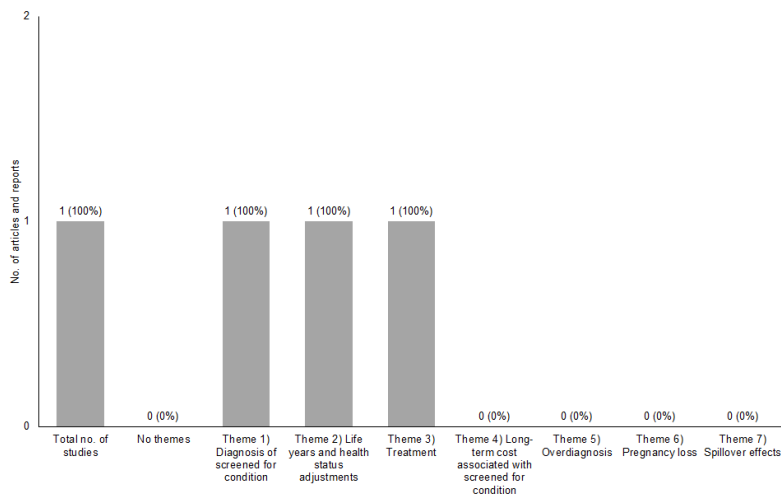


Figure 7 Benefits and harms adopted by health-economic assessments evaluating antenatal screening programmes for maternal mental health conditions using thematic framework

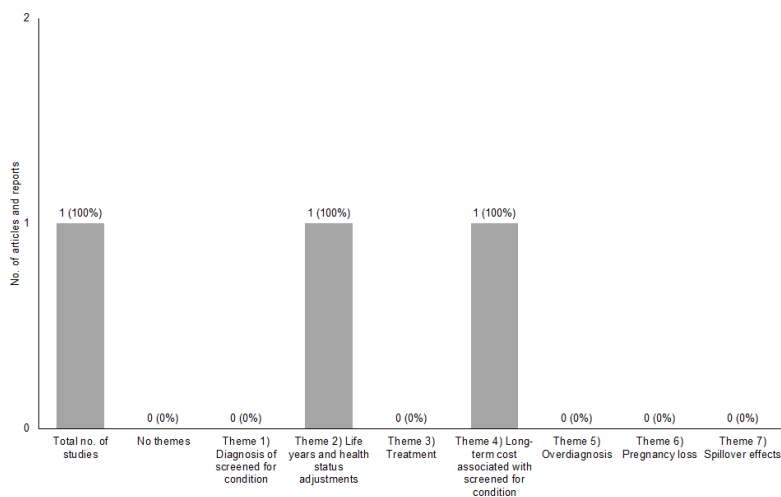
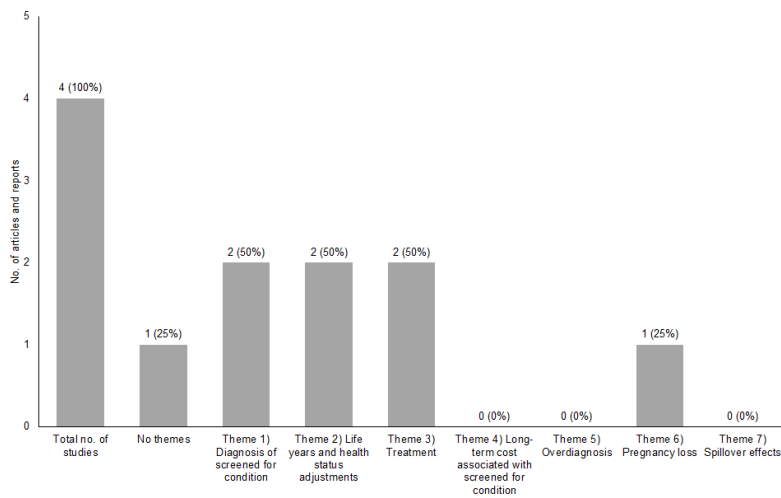


Figure 8 Benefits and harms adopted by health-economic assessments evaluating antenatal screening programmes for neurodevelopmental conditions using thematic framework

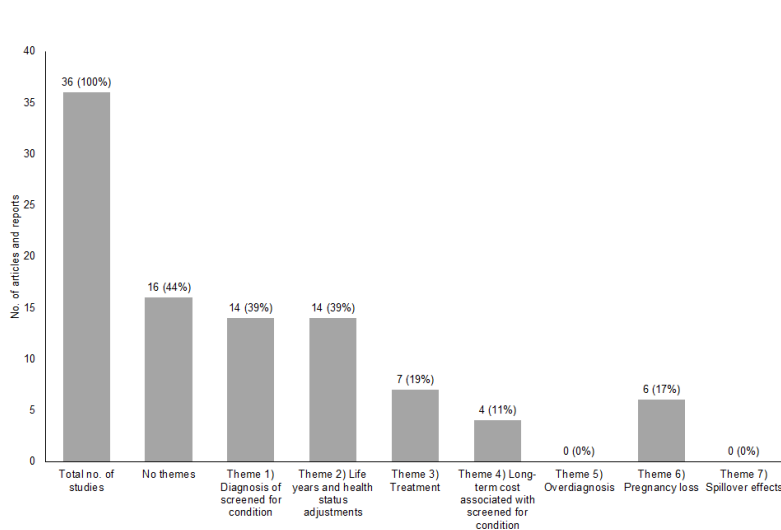
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Figure 9 Benefits and harms adopted by health-economic assessments evaluating antenatal screening programmes for nutritional conditions using thematic framework



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Figure 10 Benefits and harms adopted by health-economic assessments evaluating antenatal screening programmes for structural conditions using thematic framework

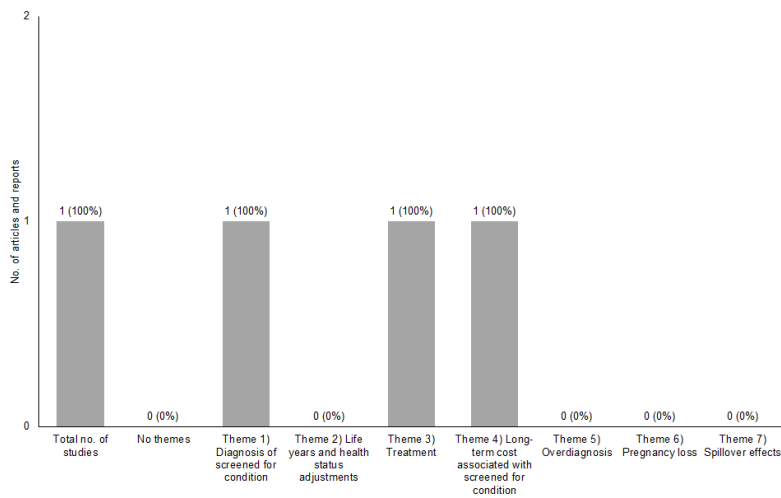


Figure 11 Benefits and harms adopted by health economic assessments evaluating antenatal screening programmes for urological conditions using thematic framework

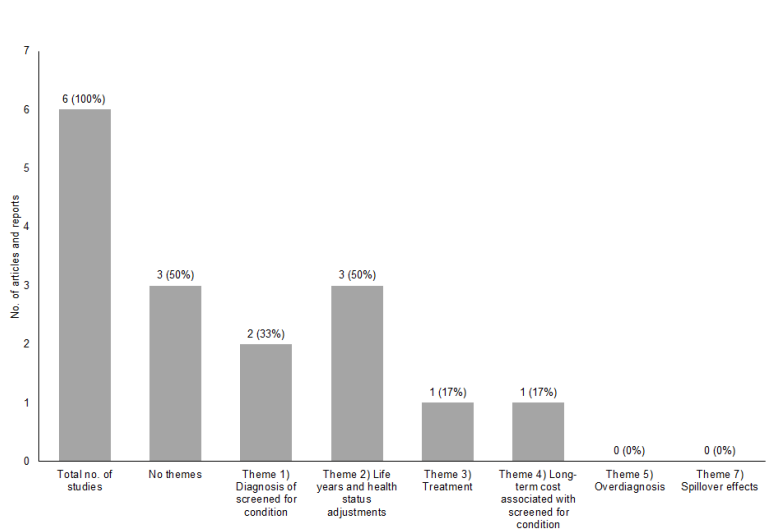
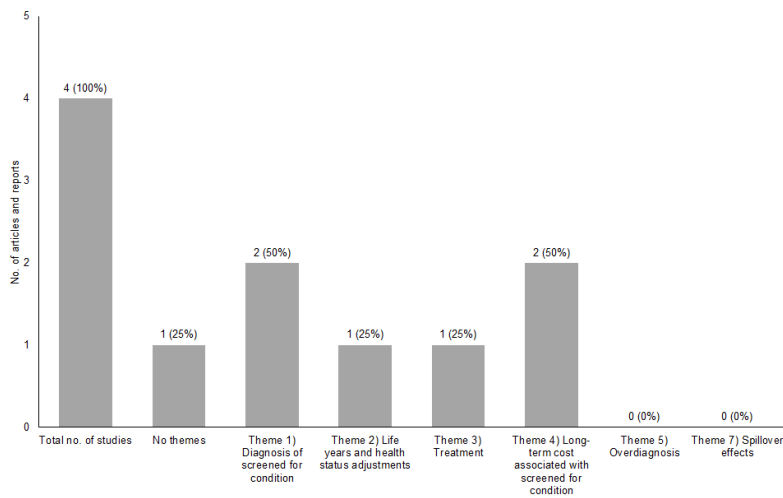


Figure 12 Benefits and harms adopted by health economic assessments evaluating newborn screening programmes for developmental conditions using thematic framework

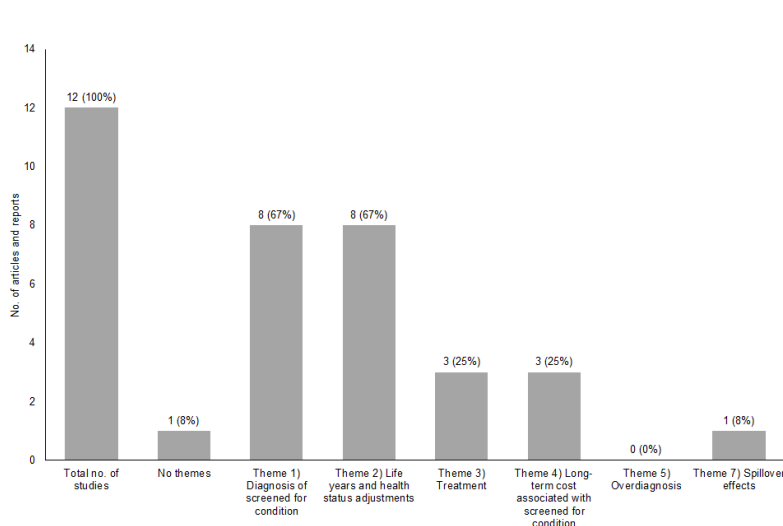
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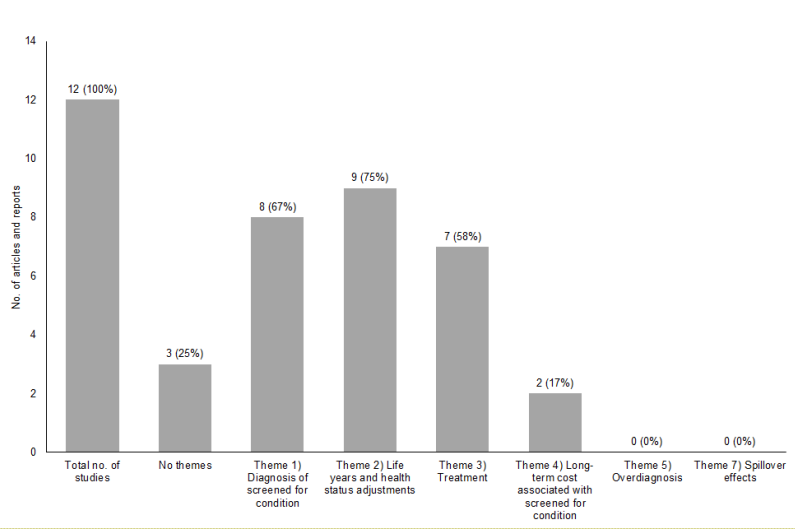
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Figure 13 Benefits and harms adopted by health economic assessments evaluating newborn screening programmes for endocrinological conditions using thematic framework



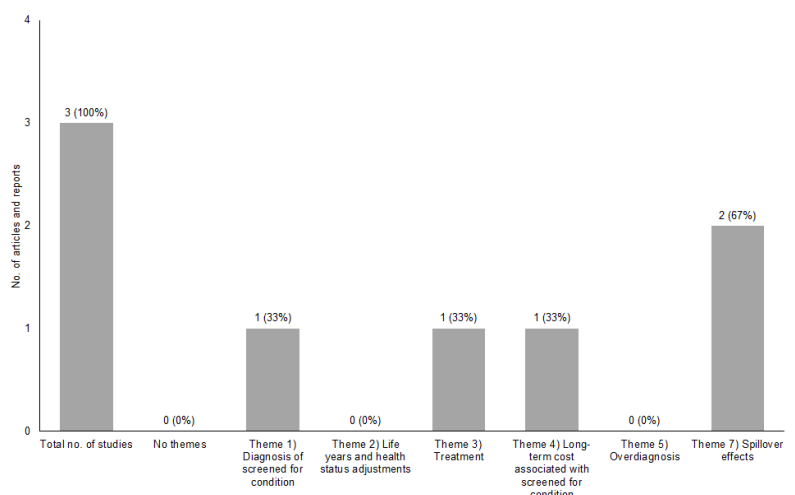
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Figure 14 Benefits and harms adopted by health economic assessments evaluating newborn screening programmes for genetic conditions using thematic framework



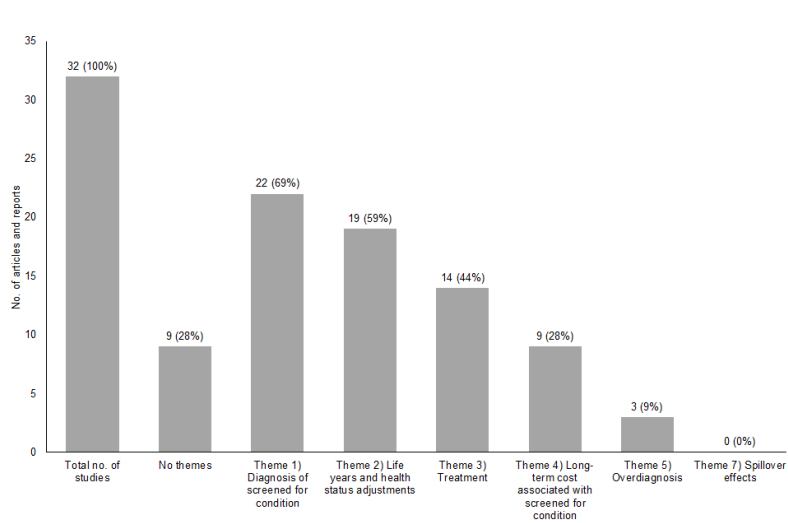
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Figure 15 Benefits and harms adopted by health economic assessments evaluating newborn screening programmes for haematological conditions using thematic framework



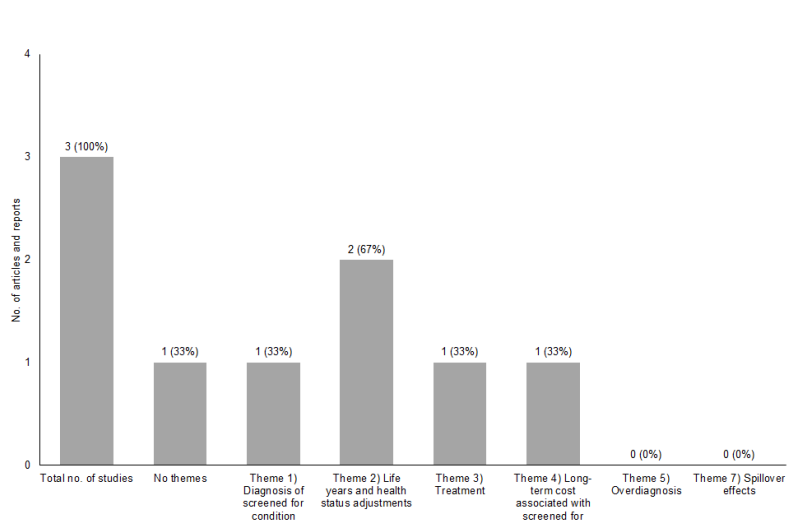
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Figure 16 Benefits and harms adopted by health economic assessments evaluating newborn screening programmes for infectious diseases using thematic framework



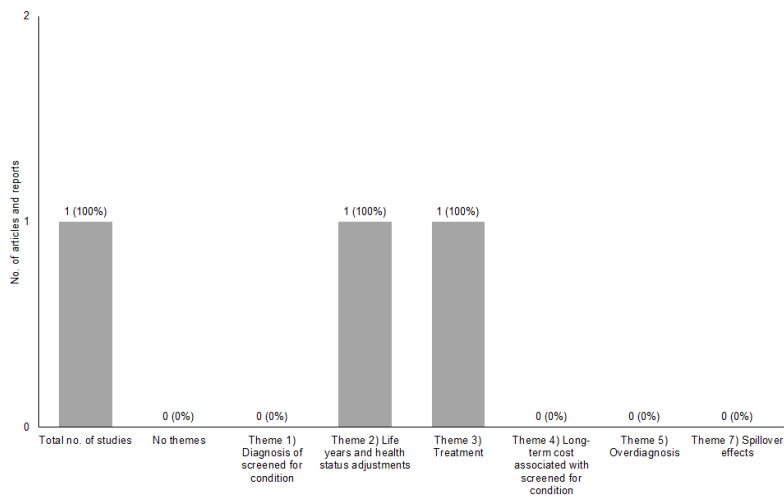
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Figure 17 Benefits and harms adopted by health economic assessments evaluating newborn screening programmes for metabolic conditions using thematic framework



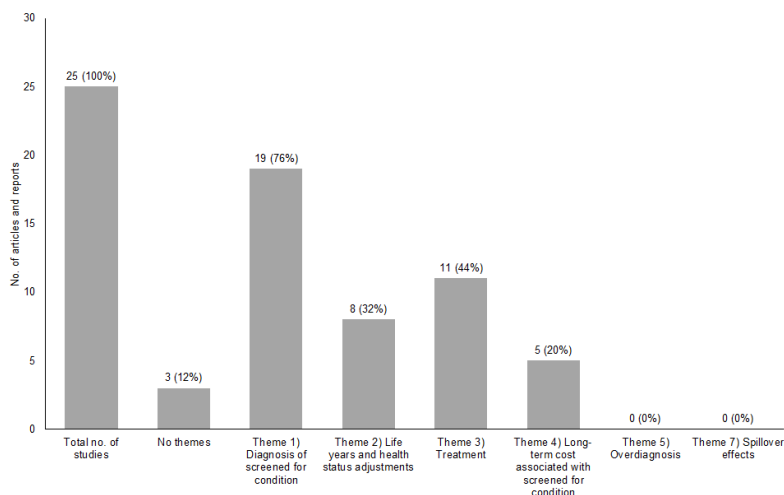
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Figure 18 Benefits and harms adopted by health economic assessments evaluating newborn screening programmes for neurodevelopmental conditions using thematic framework



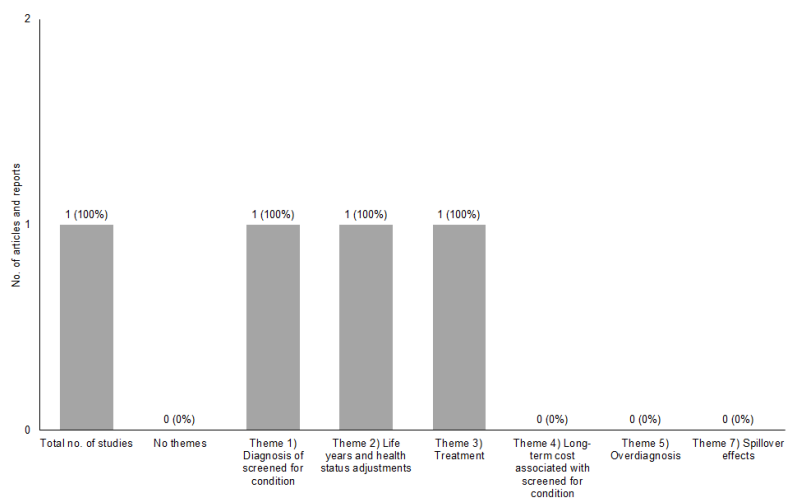
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Figure 19 Benefits and harms adopted by health economic assessments evaluating newborn screening programmes for neurological conditions using thematic framework



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Figure 20 Benefits and harms adopted by health economic assessments evaluating newborn screening programmes for structural conditions using thematic framework



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Figure 21 Benefits and harms adopted by health economic assessments evaluating newborn screening programmes for sudden infant death syndrome using thematic framework