Table 1. Response summaries for questions assessing views on newborn genetic screening. Response breakdowns are shown for family sub-groups (all, Type I, Type II/III combined, Type II and Type III) and patient sub-groups (all, Type II/III combined, Type II and Type III). Responses for each question were stratified as "agree" v "other" (other= disagree and neither disagree nor agree). This stratification was used to enable binominal logistic regression of each sub-group (**table 2**). Response distributions were compared using chi-squared analysis (P-value; significant differences are highlighted (p<0.05).

		Family Sub-Groups					Patient Sub-Groups				Statistical Comparison (chi-squared analysis)								
	All Responders (n=337)	Families (n=255)	Type I Families (n=120)	Type II/III Families (n=109)	Type II Families (n=87)	Type III Families (n=22)	Patients (n=82)	Type II and III Patients (n=58)	Type II Patients (n=27)	Type III Patients (n=31)	Families (all) vs Patients (all)	Type I vs II/III (Families)	Type I vs II (Families)	Type I vs III (Families)	Type II vs III (Families)	Type I vs II (Patients)	Type II/III (Families vs Patients)	Type II (Families vs Patients)	Type III (Families vs Patients)
Question											**P Value**	**P Value**	**P Value**	**P Value**	**P Value**	**P Value**	**P Value**	**P Value**	**P Value**
Identifying SMA at birth would lead to better support for children and families											0.01	0.18	0.15	0.75	0.77	0.11	0.009	0.003	0.71
Agree	282 (84%)	206 (81%)	101 (84%)	84 (77%)	66 (76%)	18 (82%)	76 (93%)	54 (93%)	27 (100%)	27 (87%)									
Other	55 (16%)	49 (19%)	19 (16%)	25 (23%)	21 (24%)	4 (18%)	6 (7%)	4 (7%)	0 (0%)	4 (13%)									
Identifying SMA at birth would extend life expectancy of SMA children											0.008	0.58	0.65	0.62	0.81	0.03	0.01	0.0009	0.79
Agree	127 (38%)	86 (34%)	40 (33%)	41 (38%)	32 (37%)	9 (41%)	41 (50%)	34 (59%)	20 (74%)	14 (45%)									
Other	210 (62%)	169 (66%)	80 (67%)	68 (62%)	55 (63%)	13 (59%)	41 (50%)	24 (41%)	7 (26%)	17 (55%)									
Identifying SMA at birth and not during pregancy removes parents ability to make informed decisions about bringing SMA children into the world											0.04	0.51	0.39	0.86	0.63	0.44	0.25	0.27	0.41
Agree	192 (57%)	153 (60%)	74 (62%)	62 (57%)	48 (55%)	14 (64%)	39 (48%)	27 (47%)	11 (41%)	16 (52%)									
Other	145 (43%)	102 (40%)	46 (38%)	47 (43%)	39 (45%)	8 (36%)	43 (52%)	31 (53%)	16 (59%)	15 (48%)									
Identifying SMA before symptoms emerge will prevent families and children enjoying life while they are symptom free											0.009	0.23	0.02	0.81	0.32	0.79	0.03	0.35	0.15
Agree	149 (44%)	123 (48%)	64 (53%)	49 (45%)	28 (32%)	11 (50%)	26 (32%)	16 (28%)	7 (26%)	9 (29%)									
Other	188 (56%)	132 (52%)	56 (47%)	60 (55%)	49 (68%)	11 (50%)	56 (68%)	42 (72%)	20 (74%)	22 (71%)									
Identifying SMA at birth will help research by enabling more children to be enrolled into clinical trials early on											0.57	0.54	0.51	0.95	0.57	0.55	0.57	0.96	0.76
Agree	251 (74%)	188 (74%)	88 (73%)	84 (77%)	68 (78%)	16 (73%)	63 (77%)	42 (72%)	21 (78%)	21 (68%)									
Other	86 (26%)	67 (26%)	32 (23%)	25 (23%)	19 (22%)	6 (27%)	19 (23%)	16 (23%)	6 (22%)	10 (32%)									
Identification of SMA at birth would interfere with the early bonding process											0.95	0.71	0.26	0.07	0.02	0.72	0.86	0.81	0.07
Agree	50 (15%)	38 (15%)	17 (14%)	18 (17%)	18 (21%)	0 (0%)	12 (15%)	9 (16%)	5 (19%)	4 (13%)									
Other	287 (85%)	217 (85%)	103 (86%)	91 (83%)	69 (79%)	22 (100%)	70 (85%)	49 (84%)	22 (81%)	27 (87%)									
Identification of SMA at birth would make the diagnosis easier for parents to accept											0.001	0.91	0.96	0.79	0.84	0.43	0.02	0.32	0.15
Agree	100 (30%)	64 (26%)	30 (25%)	28 (26%)	22 (25%)	6 (27%)	36 (44%)	25 (43%)	10 (37%)	15 (48%)									
Other	237 (70%)	191 (74%)	90 (75%)	81 (74%)	65 (75%)	16 (73%)	46 (56%)	33 (57%)	17 (63%)	16 (52%)									
Identifying SMA at birth would spare the difficulties associated with finding a diagnosis for a child later on											0.06	0.78	0.38	0.14	0.04	0.37	0.08	0.03	0.51
Agree	222 (66%)	161 (63%)	77 (64%)	68 (62%)	50 (57%)	18 (82%)	61 (74%)	44 (76%)	22 (81%)	22 (71%)									
Other	115 (34%)	94 (37%)	43 (36%)	41 (38%)	37 (43%)	4 (18%)	21 (26%)	14 (24%)	5 (19%)	9 (29%)									
Identifying SMA at birth is important, even if the Type can not be determined											0.01	0.27	0.19	0.83	0.34	0.75	0.01	0.11	0.34
Agree	225 (67%)	161 (63%)	79 (66%)	64 (59%)	49 (56%)	15 (68%)	64 (79%)	45 (78%)	20 (74%)	25 (81%)									
Other	112 (33%)	94 (37%)	41 (34%)	45 (41%)	38 (44%)	7 (32%)	18 (21%)	13 (22%)	7 (26%)	6 (19%)									
Identifying SMA at birth is important because it will enable parents to make informed decisions about future pregnancies											0.79	0.24	0.28	0.53	0.92	0.81	0.54	0.79	0.72
Agree	272 (81%)	205 (80%)	101 (84%)	85 (78%)	68 (78%)	17 (77%)	67 (82%)	48 (83%)	22 (81%)	26 (84%)									
Other	65 (19%)	50 (20%)	19 (16%)	24 (22%)	19 (22%)	5 (23%)	15 (18%)	10 (17%)	5 (19%)	5 (16%)									
It is unethical to screen newborns for conditions that have no effective treatment											0.51	0.47	0.44	0.65	0.86	0.52	0.75	0.91	0.72
Agree	27 (8%)	19 (7%)	8 (6%)	11 (10%)	9 (10%)	2 (9%)	8 (10%)	5 (8%)	3 (11%)	2 (6%)									
Other	310 (92%)	236 (93%)	112 (94%)	98 (90%)	78 (90%)	20 (91%)	74 (90%)	53 (92%)	24 (89%)	29 (94%)									
I would support an Newborn screening programme for SMA											0.61	0.57	0.54	0.86	0.81	0.97	0.15	0.34	0.53
Agree	236 (70%)	175 (68.6%)	84 (70%)	72 (66%)	57 (66%)	15 (68%)	61 (74.4%)	45 (78%)	21 (78%)	24 (77%)									
Other	101 (30%)	80 (31.4%)	36 (30%)	37 (34%)	30 (37%)	7 (32%)	21 (25.6%)	13 (22%)	6 (22%)	7 (23%)									

Table 2: Logistic regression analysis highlighting positive and negative drivers associated with responders views on newborn genetic screening. Odds ratios are presented for response breakdowns are shown for family sub-groups (all, Type I, Type II/III combined, Type II and Type III) and patient sub-groups (all, Type II/III combined, Type II and Type III). Responses for each question were stratified as “agree” v “other” (other= disagree and neither disagree nor agree). Odds ratios (OR) show the likelihood that responders who agreed with each individual question (variable) were also in favour of newborn genetic screening. Positive drivers are indicated by a odds ratio >1 and a p value <0.05); negative drivers are indicated by an odds ration <1 and a p value <0.05). Significant drivers are highlighted in orange. Several ORs are presented as NR (not returned); this is because of the low number of responders and included “other” responses.

	All		Fl(all)		Type II/III (n=120)		Type II and III F (n=109)		Type II/III F (n=87)		Type II/III F (n=22)		AwS (all)		Type II and III AwS (n=58)		Type II AwS (n=27)		Type III AwS (n=31)	
Question	**Odds Ratio (95% CI)**	**P-Value**	**Odds Ratio (95% CI)**	**P-Value**	**OR (95% CI)**	**P-Value**	**OR (95% CI)**	**P-Value**	**OR (95% CI)**	**P-Value**	**OR (95% CI)**	**P-Value**	**Odds Ratio (95% CI)**	**P-Value**	**OR (95% CI)**	**P-Value**	**OR (95% CI)**	**P-Value**	**OR (95% CI)**	**P-Value**
Identifying SMA at birth would lead to better support for children and families		<0.0001		<0.0001		<0.0001		<0.0001		<0.0001		0.06		0.03		0.03		0.99		0.02
Other	Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference	
Agree	18.16	(8.62-38.25)	21.94	(9.53-50.52)	36.68	(7.80-162.45)	15.76	(5.16-48.08)	17.32	(4.98-60.27)	10.50	(0.84-130.66)	6.94	(1.16-41.20)	13.20	(1.24-140.51)	NR*		17.25	(1.41-210.12)
Identifying SMA at birth would extend life expectancy of SMA children		<0.0001		<0.0001		0.005		0.005		0.02		0.11		0.44		0.31		0.64		0.32
Other	Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference	
Agree	3.10	(1.80-5.35)	4.15	(2.09-8.22)	4.42	(1.56-12.52)	3.83	(1.49-9.85)	3.35	(1.19-9.44)	6.85	(0.65-71.72)	1.47	(0.54-3.99)	1.92	(0.55-6.67)	1.60	(0.22-11.49)	2.50	(0.40-15.50)
Identifying SMA at birth and not during pregnancy removes parents ability to make informed decisions about bringing SMA children into the world		0.02		0.02		0.62		0.04		0.11		0.17		0.31		0.06		0.67		0.04
Other	Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference	
Agree	1.72	(1.08-2.76)	1.82	(1.06-3.11)	1.22	(0.55-2.71)	2.32	(1.03-5.21)	.08	(0.84-5.10)	3.66	(0.55-24.13)	1.67	(0.61-4.63)	3.81	(0.92-15.71)	1.50	(0.22-10.07)	10.00	(1.03-97.04)
Identifying SMA before symptoms emerge will prevent families and children enjoying life while they are symptom free		0.87		0.87		0.38		0.57		0.61		0.04		0.72		0.11		0.56		0.99
Other	Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference	
Agree	1.03	(0.64-1.66)	1.04	(0.61-1.77)	1.42	(0.64-3.10)	0.78	(0.36-1.76)	1.25	(0.51-3.08)	0.08	(0.008-0.89)	1.22	(0.41-3.16)	6.00	(0.71-50.59)	2.00	(0.19-20.89)	NR*	
Identifying SMA at birth will help research by enabling more children to be enrolled into clinical trials early on		<0.0001		<0.0001		<0.0001		<0.0001		<0.0001		0.008		0.06		0.09		0.08		0.49
Other	Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference	
Agree	6.30	(3.69-10.74)	8.07	(4.33-15.03)	6.10	(2.52-14.74)	11.61	(4.04-33.36)	9.10	(2.83-29.16)	35.00	(2.57-475.31)	2.79	(0.935-8.37)	3.00	(0.82-10.97)	6.00	(0.80-44.94)	1.82	(0.32-10.34)
Identification of SMA at birth would interfere with the early bonding process		<0.0001		<0.0001		0.002		0.03		0.03		0.99		0.045		0.09		0.31		0.18
Other	Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference	
Agree	0.26	(0.14-0.49)	0.27	(0.13-0.54)	0.17	(0.05-0.52)	0.33	(0.12-0.94)	0.32	(0.11-0.94)	NR*		0.27	(0.77-0.96)	0.28	(0.06-1.26)	0.33	(0.04-2.69)	0.22	(0.02-2.02)
Identification of SMA at birth would make the diagnosis easier for parents to accept		<0.0001		<0.0001		0.005		0.003		0.007		0.99		0.53		0.31		0.46		0.06
Other	Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference	
Agree	4.95	(2.51-9.76)	13.73	(4.15-45.35)	8.50	(1.90-37.96)	21.60	(2.79-166.69)	16.91	(2.14-133.37)	NR*		1.37	(0.50-3.80)	1.96	(0.52-7.33)	0.50	(0.07-3.14)	8.40	(0.87-81.08)
Identifying SMA at birth would spare the difficulties associated with finding a diagnosis for a child later on		<0.0001		<0.0001		<0.0001		<0.0001		<0.0001		0.99		0.13		0.17		0.99		0.01
Other	Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference	
Agree	6.71	(4.02-11.22)	9.23	(5.04-16.92)	4.71	(2.05-10.83)	14.12	(5.39-36.93)	13.53	(4.56-40.17)	NR*		2.27	(0.77-6.64)	2.50	(0.65-9.49)	NR*		12.50	(1.76-88.73)
Identifying SMA at birth is important, even if the Type I cannot be determined		<0.0001		<0.0001		<0.0001		<0.0001		<0.0001		0.003		<0.0001		<0.0001		0.02		0.99
Other	Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference	
Agree	22.09	(12.13-40.20)	28.25	(13.77-58.14)	15.00	(5.81-38.70)	62.84	(16.39-240.92)	65.80	(13.43-322.23)	84.00	(4.47-1576.51)	12.22	(3.65-40.86)	46.66	(8.17-266.56)	12.00	(1.48-97.17)	NR*	
Identifying SMA at birth is important because it will enable parents to make informed decisions about future pregnancies		<0.0001		<0.0001		<0.0001		<0.0001		<0.0001		0.02		0.002		0.001		0.04		0.007
Other	Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference	
Agree	11.66	(6.21-21.89)	14.18	(6.68-30.07)	10.55	(3.26-30.97)	14.14	(4.64-43.09)	13.25	(3.82-45.92)	18.66	(1.50-232.29)	6.87	(2.05-22.98)	16.33	(3.29-80.92)	9.50	(1.09-82.72)	30.66	(2.51-373.54)
It is unethical to screen newborns for conditions that have no effective treatment		<0.0001		0.001		0.01		0.03		<0.0001		0.02		0.002		0.01		0.99		0.36
Other	Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference		Reference	
Agree	0.12	(0.05-0.30)	0.18	(0.06-0.50)	0.12	(0.02-0.63)	0.25	(0.06-0.92)	13.25	(3.82-45.92)	18.66	(1.50-232.29)	0.03	(0.004-0.29)	0.05	(0.005-0.51)	NR*		0.26	(0.01-4.80)

Table 3: Levels of comparative support for the three potential SMA screening programmes (newborn screening, preconception genetic screening and prenatal screening). Support was compared in the following sub-groups: 1) Type I families; 2) Type II families; 3) Type III families; 4) adults with type II SMA; and 5) Adults with type III SMA. Agreement was assessed using a kappa analysis- Cohen's interpretation criteria are included; statistical significance of the Kappa (p-value) is shown (significance assigned using a <0.05 cut off).

Type I Families (n=120)				
	I would support a newborn screening programme			
Question	**Other**	**Agree**	**Kappa**	**P-Value**
I would support a pre-conception genetic screening programme			0.28	<0.0001
Other	10 (8%)	4 (3%)		
Agree	26 (22%)	80 (67%)		
I would support a prenatal screening programme			0.31	<0.0001
Other	11 (9%)	4 (3%)		
Agree	25 (21%)	80 (67%)		
Type II Families (n=87)				
	I would support a newborn screening programme			
Question	**Other**	**Agree**	**Kappa**	**P-Value**
I would support a pre-conception genetic screening programme			0.25	0.01
Other	13 (15%)	11 (13%)		
Agree	17 (20%)	46 (53%)		
I would support a prenatal screening programme			0.39	<0.0001
Other	16 (18%)	9 (10%)		
Agree	14 (16%)	48 (56%)		
Type III Families (n=22)				
	I would support a newborn screening programme			
Question	**Other**	**Agree**	**Kappa**	**P-Value**
I would support a pre-conception genetic screening programme			0.23	0.26
Other	3 (14%)	3 (14%)		
Agree	4 (18%)	12 (54%)		
I would support a prenatal screening programme			0.58	0.006
Other	5 (23%)	2 (9%)		
Agree	2 (9%)	13 (59%)		
Adults with Type II SMA (n=27)				
	I would support a newborn screening programme			
Question	**Other**	**Agree**	**Kappa**	**P-Value**
I would support a pre-conception genetic screening programme			0.48	0.01
Other	5 (19%)	5 (19%)		
Agree	1 (4%)	16 (58%)		
I would support a prenatal screening programme			0.32	0.05
Other	5 (19%)	8 (30%)		
Agree	1 (4%)	13 (47%)		
Adults with Type III SMA (n=31)				
	I would support a newborn screening programme			
Question	**Other**	**Agree**	**Kappa**	**P-Value**
I would support a pre-conception genetic screening programme			0.24	0.16
Other	2 (6%)	2 (6%)		
Agree	5 (16%)	22 (72%)		
I would support a prenatal screening programme			0.51	0.004
Other	4 (13%)	2 (6%)		
Agree	3 (9%)	22 (72%)		

Interpretation of Cohen's Kappa	
Kappa Range	**Interpretation**
0-0.2	No Agreement
0.21-0.39	Minimal Agreement
0.40-0.59	Weak Agreement
0.60-0.79	Moderate Agreement
0.80-0.90	Strong Agreement
>0.90	Almost Perfect Agreement