pediatric hematological malignancies has made it difficult to judge whether these advances in medical science have contributed to the welfare of children across the country.

In order to resolve these issues, the Japanese Society of Pediatric Hematology (JSPH), which unified with the Japan Society of Pediatric Oncology to the Japanese Society of Pediatric Hematology/Oncology since January, 2012, began a registry of newly diagnosed hematological diseases including non-malignant diseases partly in conjunction with the Japanese Society of Hematology in 2006, and planned complementary research into the prognoses (outcomes regarding dead or alive) as part of a research project intended to grasp the total number of pediatric patients with hematological diseases. This is the first report to describe survival times for the nationwide patients with pediatric hematological malignancies as well as the incidence of them from the JSPH disease registry [15, 16].

Materials and methods

The disease registry survey was conducted on the treatment facilities, where JSPH members are working and also pre-registered to JSPH Disease Registry Project. Using electronic or paper-based survey forms, the participating facilities voluntarily and continuously registered the cases of patients below the age of 20, who were diagnosed as hematological malignancies or benign hematological disorders after 2006. As for the electronic registration, E-DMS online by the e-Trial Co., Ltd. was used until December 2011 when it was replaced by Patient Data Organizing System (Ptosh) developed by the National Hospital Organization Nagoya Medical Center Clinical Research Center in collaboration with a non-profit organization, Organization for Supporting Clinical Research (NPO-OSCR). For fax registrations, the disease registry data were sent to the Data Management Department of NPO-OSCR. The database prepared for the registry was coordinated with the registry for epidemiological researches/clinical trials organized by Japanese Pediatric Leukemia/Lymphoma Study Group (JPLSG) in an attempt to "unify the registration" so as to provide more convenience to the participating facilities and to prevent non-registration.

In order to maintain the uniformity of the diagnoses concerning diseases to be registered, JSPH Disease Registry Committee prepared a guideline for diagnosis, to which the participants were requested to conform [17]. Acute lymphoblastic leukemia (ALL), acute myeloid leukemia (AML), rare leukemia, myelodysplastic syndromes and/or myeloproliferative neoplasms (MDS and/or MPN), or transient abnormal myelopoiesis associated with Down syndrome (Down-TAM), non-Hodgkin (NHL) and Hodgkin lymphomas (HL), histiocytosis including Langerhans cell histiocytosis (LCH) and hemophagocytic lymphohistiocytosis (HLH), other lymphoproliferative disorders (LPD) and other hematological malignancies were defined as the hematological malignancies to be registered. Underlying diseases, pathological/immunological/cytogenetic characteristics, pathogenetic forms (primary/secondary), and other natures of diseases were also recorded.

When a patient is affected by multiple diseases, each disease was registered as one entry. Patients' genders, places of residence at the initial diagnosis, dates of birth, dates of diagnosis, etc., were registered as the basic patient information. The outcomes of the respective diseases (alive or death), along with the diagnosed disease information, were recorded up to the end of May and were registered for every calendar year.

The registered data were compiled according to diagnoses, diagnosed years, genders, age categories (0, 1–4, 5–9, 10–14, and 15–19) and residential areas at diagnosis (Hokkaido, Tohoku, Kanto-Koshinetsu, Tokai-Hokuriku, Kinki, Chugoku-Shikoku, and Kyushu-Okinawa) to indicate the numbers of cases, respectively. A crude incidence rate is the number of new cases by diagnoses in a gender/age-specified populations under age 20 as of each diagnosed year, expressed as the number of hematological malignancies per 100,000 population at risk. Overall survival (OS) was defined as the length of time from the diagnosis of hematological malignancies to death from any cause. Patients were censored at the time of loss of follow-up or June 15, 2011. OS was estimated using the Kaplan–Meier method, and 2-year survival rate (2y-OS) was measured with a 95 % confidence interval (95 % CI) using Greenwood's formula. All statistical analyses were carried out using the SAS software Release 9.1 (SAS Institute Inc., Cary, NC, USA).

This registry project is operated upon obtaining the approval of JSPH Clinical Research Review Committee, followed by consents from the head of each participating institute.

Results

Numbers of registered institutions and cases

The number of institutions registered had increased by 16 from 223 institutions of the 2007 survey to 239 by the time of the 2011 survey (including the 4 institutions which had withdrawn during this period), with participation from 47 prefectures throughout Japan. Registration of cases with hematological malignancies was conducted by 187 (78.2 %) among the 239 institutions. Since retrospective registration was allowed for cases diagnosed since 2006, increases in the number of registered cases were found up

to 3 years in plateau after the diagnoses. A total of 5,287 cases were registered as hematological malignancies from 2006 to 2010, and the numbers by year were 2006: 967 cases, 2007: 1,053 cases, 2008: 1,116 cases, 2009: 1,081 cases and 2010: 1,070 cases.

Incidences

The results of broadly classifying hematological malignancies into the disease groups of ALL, AML, MDS and/or MPN, NHL, HL, histiocytosis, LPD, other hematological malignancies and Down-TAM, and tabulating by the year of diagnosis are shown in Table 1. A total of 5,287 cases were registered in 5 years, which was an annual incidence of hematologic malignancies of 4.5 cases per 100,000 people. The greatest number of cases was ALL with 2,464 cases (46.6 %), followed by AML with 891 cases (16.9 %), NHL with 628 cases (11.9 %) and histiocytosis with 624 cases (11.8 %), and this trend remained nearly constant without any dependence on the year of diagnosis. On the other hand, the number of cases reported as Down-TAM in 2010 had increased by about 1.7 times from the average number of cases reported in previous years. In addition, there were a large number of cases reported as other LPD in 2007, about two times more than in other years. The number of registered cases including rare leukemia (36 cases), other LPD (51 cases) and other hematological malignancies (6 cases) were small.

Table 2 shows the number of registrations by gender, age category, and residential areas at diagnosis for each disease group classification.

In the tabulation of ALL by the immunophenotypic classification, the most part was accounted for by B-precursor ALL with 2,110 cases (85.6 %), followed by T-ALL with 1,269 cases (10.9 %) and mature B-ALL with 60 cases (2.4 %). The peak incidence of ALL occurs between 1 and 4 years of age. In addition, the incidence of ALL is slightly higher among male children than female children, and this difference is consistent regardless of the classification by immunophenotype. Genetic abnormalities in 2,464 cases with ALL included 281 cases (11.4 %) with hyperdiploid karyotype over 50 chromosomes, 247 cases (10.0 %) with ETV6-RUNX1, 135 cases (5.5 %) with MLL rearrangement, 113 cases (4.6 %) with E2A-PBX1, 106 cases (4.3 %) with BCR-ABL1 gene rearrangement, 29 cases (1.2 %) with t(v;8q24) and 583 cases (23.7 %) with other abnormalities.

In the tabulation of AML by FAB classification, overall the greatest number was M2 with 218 cases (24.5 %), followed by M7 with 212 cases (21.5 %), M5 with 124 cases (13.9 %) and M4 with 112 cases (12.6 %). The distribution by age category showed the greatest numbers of M2 for ages 5–9 and 10–14 years (the peak of incidence during age 10–14 years), but for ages 0 and 1–4 years the incidence of M7 was extremely high (the peak of incidence during age 1–4 years), making up almost half of the incidences. Half of patients diagnosed with M7 AML were associated with Down syndrome ($n = 114$), corresponding to 94.2 % of 121 AML patients with Down syndrome. No clear difference in the number of cases of disease was found between the genders.

Table 1 Numbers of cases and incidence rates of hematological malignancies in Japanese children and adolescents, diagnosed between 2006 and 2010

Disease	Total (%)	Crude incidence rate[a]	Year of diagnosis				
			2006	2007	2008	2009	2010
Acute lymphoblastic leukemia	2,464 (46.6)	2.1	444	506	532	504	478
Acute myeloid leukemia	891 (16.9)	0.8	167	165	184	193	182
Rare leukemia	36 (0.7)	0.0	9	7	4	10	6
Myelodysplastic syndrome and/or myeloproliferative neoplasms	296 (5.6)	0.3	61	60	46	54	75
Non-Hodgkin lymphoma	628 (11.9)	0.5	118	129	138	137	106
Hodgkin lymphoma	107 (2.0)	0.1	19	21	24	14	29
Histiocytosis	624 (11.8)	0.5	114	108	138	128	136
Transient abnormal myelopoiesis associated with Down syndrome	184 (3.5)	0.2	26	37	37	31	53
Other hematological malignancies	6 (0.1)	0.0	0	0	5	1	0
Other lymphoproliferative disorders	51 (1.0)	0.0	9	20	8	9	5
Hematological malignancies, Total	5,287 (100.0)	4.5	967	1,053	1,116	1,081	1,070

[a] Crude incidence rate is the number of new cases by diagnoses in a gender/age-specified populations under age 20 as of each diagnosed year, expressed as the number of hematological malignancies per 100,000 population at risk

Table 2 Numbers of incidences of hematological malignancies according to gender, age category, and residential areas at diagnosis in Japanese children and adolescents, diagnosed between 2006 and 2010

Disease	Subtype	*n* (%)	*n* (%)	Gender, *n* (%)		Age, *n* (%)					Residential areas at diagnosis, *n* (%)						
				Male	Female	0	1–4	5–9	10–14	15–19	Hokkaido	Tohoku	Kanto-Koshinetsu	Tokai-Hokuriku	Kinki	Chugoku-Shikoku	Kyushu-Okinawa
Acute lymphoblastic leukemia		2,464 (46.6)		1,411 (57.3)	1,053 (42.7)	108 (4.4)	1044 (42.4)	711 (28.9)	499 (20.3)	102 (4.1)	105 (4.3)	192 (7.8)	971 (39.4)	347 (14.1)	415 (16.8)	190 (7.7)	244 (9.9)
	B-precursor		2,110 (85.6)	1,151	959	102	979	580	372	77	83	168	829	306	350	164	210
	Mature B		60 (2.4)	38	22	5	9	24	20	2	4	2	22	6	11	7	8
	T cell		269 (10.9)	206	63	1	46	101	99	22	15	19	112	34	46	18	25
	Unknown		25 (1.0)	16	9	0	10	6	8	1	3	3	8	1	8	1	1
Acute myeloid leukemia		891 (16.9)		451 (50.6)	440 (49.4)	109 (12.2)	306 (34.3)	168 (18.9)	240 (26.9)	68 (7.6)	32 (3.6)	69 (7.7)	348 (39.1)	121 (13.6)	134 (15.0)	78 (8.8)	109 (12.2)
	M0		33 (3.7)	15	18	4	7	10	7	5	1	4	15	4	4	0	5
	M1		73 (8.2)	35	38	2	17	19	28	7	4	4	30	12	10	6	7
	M2		218 (24.5)	109	109	3	37	73	88	17	11	17	78	28	35	20	29
	M3, M3v		70 (7.9)	37	33	2	12	17	29	10	1	2	22	11	17	7	10
	M4, M4Eo		112 (12.6)	57	55	12	29	20	38	13	3	12	37	19	21	8	12
	M5a, M5b		124 (13.9)	63	61	28	33	16	39	8	6	9	47	15	20	13	14
	M6a, M6b		14 (1.6)	8	6	0	7	3	3	1	0	1	6	1	2	2	2
	M7		212 (23.8)	104	108	55	150	2	3	2	6	17	99	28	19	16	27
	Unknown		35 (3.9)	23	12	3	14	8	5	5	0	3	14	3	6	6	3
Rare leukemia		36 (0.7)		24 (66.7)	12 (33.3)	5 (13.9)	8 (22.2)	6 (16.7)	14 (38.9)	3 (8.3)	2 (5.6)	2 (5.6)	14 (38.9)	4 (11.1)	4 (11.1)	5 (13.9)	5 (13.9)
Myelodysplastic syndrome (MDS) and/or Myeloproliferative neoplasms (MPN)		296 (5.6)		171 (57.8)	125 (42.2)	42 (14.2)	73 (24.7)	66 (22.3)	92 (31.1)	23 (7.8)	13 (4.4)	10 (3.4)	127 (42.9)	40 (13.5)	43 (14.5)	34 (11.5)	29 (9.8)
	MPN		111 (37.5)	62	49	2	13	33	57	6	4	3	46	17	19	13	9
	MDS/MPN		49 (16.6)	32	17	27	18	1	1	2	2	2	23	7	5	3	7
	MDS		136 (45.9)	77	59	13	42	32	34	15	7	5	58	16	19	18	13
Non-Hodgkin lymphoma		628 (11.9)		446 (71.0)	182 (29.0)	7 (1.1)	96 (15.3)	237 (37.7)	240 (38.2)	48 (7.6)	26 (4.1)	45 (7.2)	218 (34.7)	107 (17.0)	107 (17.0)	56 (8.9)	69 (11.0)
	Lymphoblastic-T-precursor		136 (21.7)	100	36	0	14	48	63	11	8	14	51	21	24	8	10
	Lymphoblastic-B-precursor		71 (11.3)	42	29	5	20	31	12	3	5	4	25	14	7	9	7
	Burkitt		154 (24.5)	128	26	0	30	75	43	6	3	9	52	28	30	14	18
	Diffuse large B cell		121 (19.3)	80	41	0	12	42	50	17	4	9	35	17	22	17	17
	Anaplastic large cell		100 (15.9)	71	29	0	13	31	49	7	4	5	35	21	17	7	11

Table 2 continued

Disease	Subtype	*n* (%)	*n* (%)	Gender, *n* (%)		Age, *n* (%)					Residential areas at diagnosis, *n* (%)						
				Male	Female	0	1–4	5–9	10–14	15–19	Hokkaido	Tohoku	Kanto-Koshinetsu	Tokai-Hokuriku	Kinki	Chugoku-Shikoku	Kyushu-Okinawa
	Other		46 (7.3)	25	21	2	7	10	23	4	2	4	20	6	7	1	6
Hodgkin lymphoma		107 (2.0)		61 (57.0)	46 (43.0)	0 (0.0)	6 (5.6)	28 (26.2)	55 (51.4)	18 (16.8)	4 (3.7)	9 (8.4)	37 (34.6)	19 (17.8)	15 (14.0)	7 (6.5)	16 (15.0)
Histiocytosis		624 (11.8)		329 (52.7)	295 (47.3)	113 (18.1)	259 (41.5)	139 (22.3)	95 (15.2)	18 (2.9)	27 (4.3)	36 (5.8)	213 (34.1)	96 (15.4)	125 (20.0)	55 (8.8)	72 (11.5)
	Langerhans cell histiocytosis		345 (55.3)	199	146	66	143	81	50	5	20	20	110	50	66	39	40
	Hemophagocytic lymphohistiocytosis		265 (42.5)	123	142	44	108	56	44	13	6	15	97	44	55	16	32
	Other		14 (2.2)	7	7	3	8	2	1	0	1	1	6	2	4	0	0
Transient abnormal myelopoiesis associated with Down syndrome		184 (3.5)		100 (54.4)	84 (45.7)	182 (98.9)	2 (1.1)	0 (0.0)	0 (0.0)	0 (0.0)	4 (2.2)	3 (1.6)	86 (46.7)	23 (12.5)	28 (15.2)	13 (7.1)	27 (14.7)
Other	lymphoproliferative disorders		51 (1.0)		24 (47.1)	27 (52.9)	1 (2.0)	9 (17.7)	19 (37.3)	18 (35.3)	4 (7.8)	0 (0.0)	4 (7.8)	13 (25.5)	8 (15.7)	9 (17.7)	11 (21.6)
6 (11.8)																	
Other hematologic malignancies		6 (0.1)		2 (33.3)	4 (66.7)	5 (83.3)	0 (0.0)	1 (16.7)	0 (0.0)	0 (0.0)	2 (33.3)	0 (0.0)	2 (33.3)	1 (16.7)	0 (0.0)	0 (0.0)	1 (16.7)
Hematological malignancies	Total	5,287 (100.0)		3,019 (57.1)	2,268 (42.9)	572 (10.8)	1,803 (34.1)	1,375 (26.0)	1,253 (23.7)	284 (5.4)	215 (4.1)	370 (7.0)	2,029 (38.4)	766 (14.5)	880 (16.6)	449 (8.5)	578 (10.9)

Among NHL patients, those with Burkitt lymphoma (BL, 24.5 %), precursor T-lymphoblastic lymphoma (21.7 %), or diffuse large B cell lymphoma (DLBCL, 19.3 %), respectively, accounted for more than 20 %. When combined with those with anaplastic large cell lymphoma (ALCL, 15.9 %) and those with precursor B-lymphoblastic lymphoma (11.3 %), patients with these types of lymphoma accounted for 92.7 % of all NHL patients. Three subtypes, i.e., nodular sclerosis (37 cases), nodular lymphocyte predominance (29 cases), and mixed cellularity (31 cases), accounted for 90.7 % of all HL patients. While NHL patients were predominantly male, accounting for 2.5 times the number of female patients, no significant gender difference was observed for HL, with only 1.3 times male predominance. Peak incidences of both NHL and HL occurred at 5 years of age or older. While the incidence of BL peaked between ages 5 and 9, the incidence of precursor T-lymphoblastic lymphoma, DLBCL, and ALCL increased with age and peaked between ages 10 and 14.

The majority of histiocytosis included LCH, at 345 cases (55.3 %), followed by HLH at 265 cases (42.5 %). Incidences of both HLH and LCH mainly occurred in children aged four and under.

Among patients with MDS and/or MPN, those with MDS made up the majority (136 cases, 45.9 %), followed by MPN (111 cases, 37.5 %), including chronic myeloid leukemia (CML), and MDS/MPN (49 cases, 16.6 %), including juvenile myelomonocytic leukemia (JMML) and chronic myelomonocytic leukemia (CMML). The breakdown of registered cases of MDS by the JSPH guideline for diagnosis showed that refractory anemia (33 cases) account for 22 %, refractory cytopenia with multi-lineage dysplasia (25 cases) for 18 %, refractory anemia with excess blasts (RAEB)-1 (22 cases) for 16 %, and RAEB2 (19 cases) for 14 %, respectively.

Down-TAM accounted for 3.5 % of all cases, with the incidence rate being slightly higher in males (1.2 times higher than females).

No significant regional difference was observed when looking at age-specific incidences (data not shown).

Survival

Table 3 and Figs. 1, 2, 3, 4, 5 and 6 show disease-specific prognostic information for 5,287 cases of patients who were diagnosed with pediatric hematological malignancies between 2006 and 2010. The median observation period (range) was 1.7 (0.0–5.3) years. The point estimate (95 %CI) of 2y-OS for all pediatric patients with hematological malignancies was 91.6 (90.7–92.5) %. No difference was observed in survival rates in terms of gender (2y-OS 91.5 % for male, 91.9 % for female; log-rank test p value = 0.76) or residential areas at diagnosis (2y-OS 88.0 % for Hokkaido, 93.3 % for Tohoku, 91.5 % for Kanto-Koshinetsu, 91.7 % for Tokai-Hokuriku, 94.2 % for Kinki, 92.2 % for Chugoku-Shikoku, and 88.1 % for Kyushu-Okinawa; p value = 0.11). Survival rates in different age categories showed that children aged 5–9 years had the best prognosis (94.4 %). This was followed by children aged 1–4 years (93.8 %) and those aged 10–14 years (90.8 %), indicating a 2y-OS of more than 90 %. On the other hand, the 2y-OS for patients aged 15–19 years was about 10 % points lower at 80.5 %. The 2y-OS for infants less than 1 year old did not reach 90 % either at 85.6 % (log-rank test p value < 0.0001).

The comparison of 2y-OS among different diseases indicates that patients with HL (95.2 %) had the best prognosis.

Table 3 Survival for Japanese children and adolescents diagnosed with hematological malignancies

Disease	n	1 year		2 year		3 year		4 year		5 year	
		1-yr OS	95 % CI	2-yr OS	95 % CI	3-yr OS	95 % CI	4-yr OS	95 % CI	5-yr OS	95 % CI
Acute lymphoblastic leukemia	2,464	97.3	96.5–97.9	94.2	93.0–95.2	91.1	89.4–92.5	89.1	87.0–90.9	88.7	86.4–90.6
B-precursor	2,110	98.0	97.3–98.6	96.2	95.1–97.0	93.6	92.0–94.9	91.8	89.7–93.5	91.3	88.9–93.2
Mature B	60	95.6	83.4–98.9	84.7	68.8–92.9						
T cell	269	92.4	87.9–95.2	81.3	74.3–86.5	71.0	61.8–78.3	66.9	56.3–75.4		
Unknown	25	90.5	67.0–97.5	75.4	33.3–93.0						
Acute myeloid leukemia	891	91.2	88.9–93.0	83.3	80.1–86.1	77.4	73.3–80.9	76.3	71.9–80.1	75.2	70.3–79.4
M0	33	81.2	60.5–91.8	71.8	49.3–85.7	59.9	30.0–80.3				
M1	73	87.7	75.8–94.0	80.5	66.3–89.2	76.8	61.0–86.9				
M2	218	93.5	88.8–96.3	85.9	79.2–90.6	78.9	70.2–85.3	76.4	66.4–83.8	72.2	58.8–81.9
M3, M3v	70	95.6	87.0–98.6	95.6	87.0–98.6						
M4, M4Eo	112	91.7	84.0–95.8	79.0	68.1–86.6	72.4	59.5–81.7				

Table 3 continued

Disease	*n*	1 year		2 year		3 year		4 year		5 year	
		1-yr OS	95 % CI	2-yr OS	95 % CI	3-yr OS	95 % CI	4-yr OS	95 % CI	5-yr OS	95 % CI
M5a, M5b	124	83.7	74.9–89.6	76.7	66.4–84.2	70.5	58.3–79.7	67.4	54.1–77.6		
M6a, M6b	14	90.9	50.8–98.7	90.9	50.8–98.7						
M7	212	94.0	89.1–96.8	84.8	77.0–90.1	80.2	70.8–86.9				
Unknown	35	90.7	73.9–96.9	82.9	63.2–92.6	56.8	21.9–81.0				
Rare leukemia	36	90.6	73.5–96.9	75.2	51.5–88.5	68.9	43.9–84.5				
Myelodysplastic syndrome (MDS) and/or myeloproliferative neoplasms (MPN)	296	96.2	93.1–98.0	92.8	88.4–95.6	87.9	81.8–92.1	85.3	76.6–91.0		
MPN	111	100.0	–	98.6	90.2–99.8	96.7	87.4–99.2				
MDS/MPN	49	90.6	76.5–96.4	86.6	69.9–94.4	80.0	57.2–91.4				
MDS	136	95.0	89.2–97.7	90.0	81.9–94.6	82.6	71.3–89.7	76.2	57.8–87.4		
Non-Hodgkin lymphoma	628	94.6	92.3–96.2	92.1	89.3–94.2	90.5	87.0–93.1				
Lymphoblastic-T-precursor	136	96.4	90.6–98.6	90.4	82.2–95.0	86.9	77.0–92.7	83.1	69.8–90.9		
Lymphoblastic-B-precursor	71	98.4	89.3–99.8	96.2	85.3–99.1						
Burkitt	154	93.3	87.4–96.4	91.1	84.4–95.0						
Diffuse large B cell	121	97.1	91.3–99.1	97.1	91.3–99.1						
Anaplastic large cell	100	91.0	82.7–95.4	91.0	82.7–95.4						
Other	46	90.7	76.9–96.4	83.8	66.7–92.6						
Hodgkin lymphoma	107	98.7	91.2–99.8	95.2	85.5–98.4						
Histiocytosis	624	94.9	92.8–96.5	93.9	91.4–95.6						
Langerhans cell histiocytosis	345	99.3	97.3–99.8	98.7	95.8–99.6						
Hemophagocytic lymphohistiocytosis	265	88.9	84.3–92.3	87.7	82.8–91.4						
Other	14	100.0	–	90.0	47.3–98.5						
Transient abnormal myelopoiesis associated with Down syndrome	184	89.8	84.3–93.4	89.8	84.3–93.4						
Other lymphoproliferative disorders	51	89.5	76.6–95.5	86.3	71.6–93.7						
Other hematologic malignancies	6	83.3	27.3–97.5	83.3	27.3–97.5						
Hematological malignancies, Total	5,281	95.2	94.6–95.8	91.6	90.7–92.5	88.8	87.6–89.8	87.5	86.2–88.8	87.0	85.4–88.3
Gender											
Female	2,268	95.3	94.2–96.1	91.9	90.5–93.1	89.3	87.4–90.8	87.6	85.5–89.5	86.8	84.3–88.9
Male	3,019	95.2	94.3–96.0	91.5	90.2–92.6	88.4	86.8–89.8	87.5	85.7–89.0	87.2	85.3–88.8
Age											
0	572	88.0	84.8–90.5	85.6	82.0–88.5	83.7	79.7–87.0				
1–4	1,803	96.7	95.7–97.5	93.8	92.4–95.0	91.4	89.5–93.0	90.9	88.8–92.6		
5–9	1,375	97.1	96.0–97.9	94.4	92.7–95.7	91.0	88.7–92.9	89.5	86.7–91.7		
10–14	1,253	95.4	93.9–96.5	90.8	88.6–92.5	88.1	85.5–90.3	85.9	82.7–88.6	84.0	79.6–87.5
15–19	284	90.6	86.2–93.7	80.5	74.2–85.5	73.7	65.8–80.1			69.1	56.6–78.6
Residential areas											
Hokkaido	215	92.8	87.9–95.8	88.0	81.7–92.3	85.9	78.8–90.8			82.6	72.1–89.4
Tohoku	370	96.6	93.9–98.1	93.3	89.4–95.8	89.1	83.7–92.7	86.9	80.5–91.3		
Kanto-Koshinetsu	2,029	95.0	93.9–96.0	91.5	89.9–92.8	87.8	85.6–89.6	86.5	84.0–88.6		
Tokai-Hokuriku	766	95.5	93.6–96.8	91.7	89.1–93.7	90.8	88.0–93.0	90.3	87.3–92.7	89.0	84.7–92.2
Kinki	880	97.1	95.6–98.0	94.2	92.1–95.7	90.7	87.7–93.0	89.0	85.3–91.8	88.0	83.7–91.2
Chugoku-Shikoku	449	94.0	91.1–96.0	92.2	88.8–94.6	88.1	83.4–91.6				
Kyushu-Okinawa	578	93.6	91.1–95.4	88.1	84.6–90.9	87.3	83.7–90.9	87.3	83.7–90.2	85.1	80.5–88.7

yr year, *OS* overall survival

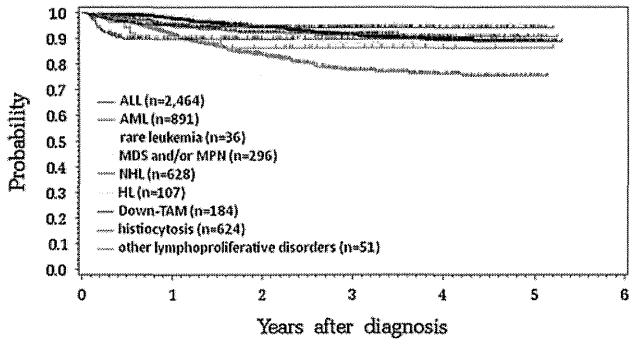

Fig. 1 Overall survival for patients diagnosed with hematological malignancies ($n = 5{,}287$). *ALL* acute lymphoblastic leukemia, *AML* acute myeloid leukemia, *MDS and/or MPN* myelodysplastic syndrome and/or myeloproliferative neoplasms, *NHL* non-Hodgkin lymphoma, *HL* Hodgkin lymphoma, *Down-TAM* transient abnormal myelopoiesis associated with Down syndrome

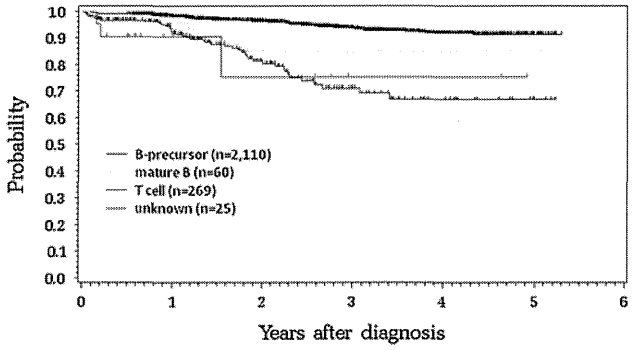

Fig. 2 Overall survival for patients diagnosed with acute lymphoblastic leukemia

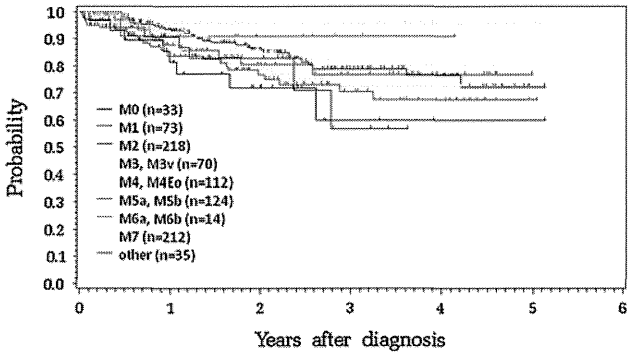

Fig. 3 Overall survival for patients diagnosed with acute myeloid leukemia

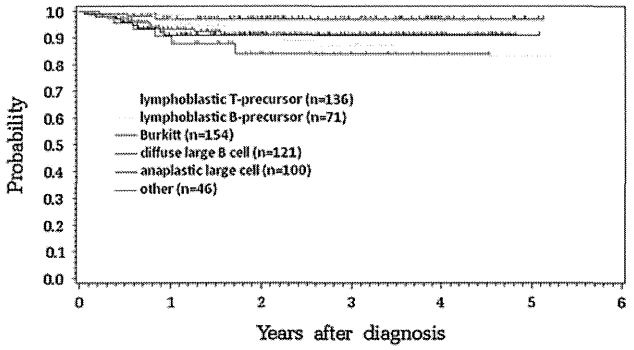

Fig. 4 Overall survival for patients diagnosed with non-Hodgkin lymphoma

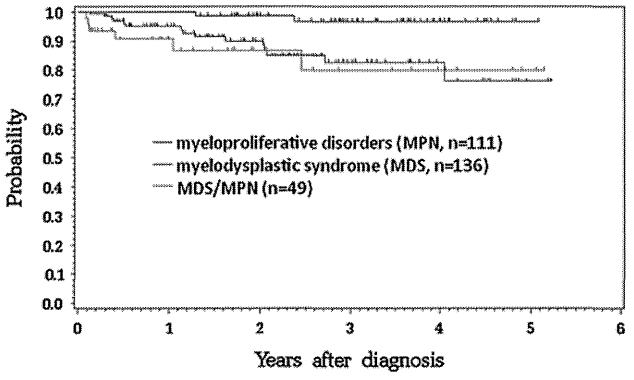

Fig. 5 Overall survival for patients diagnosed with myelodysplastic syndrome and/or myeloproliferative. *MDS* myelodysplastic syndrome, *MPN* myeloproliferative neoplasms

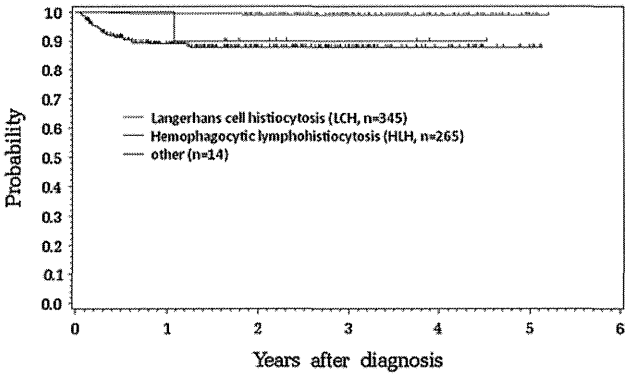

Fig. 6 Overall survival for patients diagnosed with histiocytosis

This was followed by patients with ALL (94.2 %), histiocytosis (93.9 %), MDS/MPN (92.4 %), and NHL (92.1 %). All patients had a survival rate of 90 % or more within 2 years after their disease was diagnosed. The 2y-OS for patients with Down-TAM (89.8 %) and those with other LPD (86.3 %) was estimated more than 85 %, while that for AML (83.3 %) and rare leukemia (75.2 %) was inferior to it.

Examination based on the immunophenotypic classification of ALL shows that patients with B-precursor ALL had the highest 2y-OS at 96.2 %, surpassing the 2y-OS for patients with mature B cell ALL (84.7 %) or T cell ALL (81.3 %).

Examination based on the FAB classification of AML indicates that M3 had the best prognosis (2y-OS 95.6 %),

followed by M6 (90.6 %). The 2y-OS accounted for more than 80 % with the exception of M0 (71.8 %), M5 (76.7 %), and M4 (79.0 %) whose survival rates did not reach 80 %.

Any type of NHL, without being based on the immunological classification, indicated a survival rate of more than 90 % within 2 years after diagnosis; especially, DLBCL (97.1 %) and precursor B-lymphoblastic lymphoma (96.2 %) had excellent prognoses.

Among histiocytosis, LCH indicated an extremely excellent 2y-OS of 98.7 % while that of HLH was 87.7 %. As for MDS and/or MPN, the 2y-OS of MPN was 98.6 % and the best, followed by MDS (90.0 %) and MDS/MPN (86.6 %).

Discussion

This study, which includes the largest childhood cohorts of hematological malignancies ever reported in Japan, documented progressive improvements in survival for children enrolled onto the disease registry project of the JSPH between 2007 and 2011. Considering the number of participating institutions, we estimated that our patient sample collected through the system represented about 80 % of all the cases of hematological malignancies in Japan. As cases newly diagnosed during the 5 years from January 1, 2006 to December 31, 2010, it has reported 5,287 cases of hematological malignancies from 187 institutions (diagnosis and treatment departments) across 47 prefectures nationwide; this result is equivalent to the prevalence of 4.5 cases per 100,000 people per year. The number of registered cases for age 15–19 years is much lower than those at age 14 or younger, which may be reflecting the fact that patients over the age of 16 usually visit internists rather than pediatrician. In order to figure out the exact trends in disease incidence for this age category in Japan, it is necessary to establish a registration system that can be accessed both by internists and pediatricians.

Regarding the incidence by disease group, ALL accounted for approximately half of hematological malignancies and more than 80 % when combined with AML, NHL and histiocytosis, which accounted for 10–15 %, respectively. The incidence by disease was nearly constant regardless of the diagnosis year. The reason why reports of Down-TAM almost doubled in 2010 is inferred that such increase accompanied wider recognition and utilization of the TAM central diagnosis system realized through a nationwide clinical observational study, JPLSG TAM-10, started in the same year (UMIN # 000005418).

According to the immunophenotypic classification counting of ALL, although the percentage of B-precursor ALL in Japan is higher than that in the US (85.6 vs. 63 %), T-ALL and mature B-ALL showed almost the same ratios [18]. It was also consistent with the findings in the US that the age of peak incidence was under 4 years old and that the incidence of ALL is slightly higher among male children than female children [19]. It is reported that introduction of risk-stratified treatment and improvement in supportive care have helped to achieve better treatment results of ALL with its 5y-OS higher than 85 % [14]. Our data showed that the 2y-OS of ALL was 94.2 % while its 5y-OS also exceeded 80 %. This indicates an improvement compared to the results of the European disease registry data during the second half of the twentieth century (1978–1997) [20], suggesting that prognoses as good as those in the results of recent foreign clinical studies have been achieved nationwide [14].

A dramatic improvement in the success rate of the treatment of AML has been seen [21], from about 20 % during the 1970s to 55 % in the decade following 2000. Although there are differences in thinking among different groups studying the treatment, such as those with regard to chemotherapy as well as hematopoietic stem cell transplantation depending on the disease risk, the overall survival in clinical trial has also been improved to have reached 42 ~ 62 % [22–24]. In the AML99 clinical trials (2000–2002) [25–28] conducted in Japan, good results of a 5y-OS of over 76 % were obtained. In the present study also, it was found that by and large a good 2y-OS of higher than 80 % has been obtained even when M3 (2y-OS: 95.6 %), with the best prognosis, is excluded.

The incidence of HL in our data was very low compared to that from other countries. There are reports, both domestic and from overseas, that the incidences of both NHL and HL are relatively high in adolescents, and that the ratio of male to female children is high [29–33]. In our data, NHL is uncommon in infant, and the incidence of NHL increases throughout life. Although NHL is more common than HL in children younger than 15 years, the relative incidence of HL increases in children older than 10 years, making the incidence of HL in children aged between 15 and 19, almost twice that of NHL. In addition, higher incidence of NHL in male children was observed, while there was a slight male predominance in the incidence of HL, with an incidence ratio of 1.3 in male and female children. For NHL, favorable outcomes of treatment were obtained in more than 90 % of the cases within 2 years after diagnosis regardless of the immunological classification, which were similar to reports of clinical trials from inside and outside of the country [34–37].

Similarly, there was a high incidence of LCH, which accounts for the majority of cases of histiocytosis, in the age group of 1–4 years as in the overseas reports [38], and the prognosis was also good [39]. About half of the patients (42.5 %) with histiocytosis were diagnosed as having

HLH. In accordance with the previous literature in Japan, our data showed that the incidence of HLH cases per year was about 50 (mean 53, range 43–64) [40]. And clinical outcomes of HLH were considerably improved compared to the results of HLH-94 study [41].

With regard to MDS and/or MPN, hematopoietic stem cell transplantation, rather than conventional chemotherapy, has come to be a good indication [42] in cases in which there is an HLA-matched sibling donor. Children with low-risk MDS, including refractory anemia and refractory anemia with ring sideroblasts, were not candidates for hematopoietic stem cell transplantation [43]. Although the 5y-OS of children under the age of 16 was, respectively, 50–67 % for MDS and 51–75 % for MPN, depending on the type of transplant, in the national survey results (2011 report) from 1991 to 2010 by the Japan Society for Hematopoietic Cell Transplantation (JSHCT), our data showed that there was an improvement to 86.6 % for MDS/MPN including JMML, for which the prognoses are the worst [44], although the follow-up period was not sufficient.

In treatments for hematological malignancies during childhood and adolescents, long-term toxicity, including treatment-related deaths and secondary neoplasms, still remain as important issues. Therefore, we will continuously evaluate the trends in the national levels of diagnosis and treatment through the JSPH disease registry project and will show data concerning trends in disease incidence and deaths accompanied by prognostic information. Continuous activity to monitor the level of medical care is considered quite important in aiming at the development of more effective treatments which maintain long-term safety.

Acknowledgments The survey on hematological malignancies incidence in Japan was conducted with contributions from the 187 institutions, described in Appendix 1. The authors thank deeply the members, especially Kaori Nagai, Kazumi Takeuchi, Maki Nishimura, and Midori Otomo of the Data Management Department of the NPO-OSCR, for their support in the management of the electronic or paper-based survey system and in the cleaning and tabulation of the registered data.

Conflict of interest The authors have no financial relationship to declare.

Appendix

See appendix Table 4.

Table 4 Institutions with registered cases of hematological malignancies

S. no.	District	Institutions
1	Hokkaido	Oji General Hospital
2	Hokkaido	Sapporo Medical University Hospital
3	Hokkaido	Hokkaido Medical Center for Child Health and Rehabilitation
4	Hokkaido	Sapporo Hokuyu Hospital
5	Hokkaido	Hokkaido University Hospital
6	Hokkaido	KKR Sapporo Medical Center
7	Hokkaido	Asahikawa Medical University Hospital
8	Hokkaido	Hospital Hakodate Hokkaido
9	Hokkaido	Kushiro city General Hospital
10	Hokkaido	National Hospital Organization Hokkaido Cancer Center
11	Tohoku	Hirosaki University school of Medicine and Hospital
12	Tohoku	Nakadori General Hospital
13	Tohoku	Akita University Hospital
14	Tohoku	Iwate Medical University Hospital
15	Tohoku	Iwate Prefectural Chubu Hospital
16	Tohoku	Iwaki Kyoritsu Hospital
17	Tohoku	Fukushima Medical University Hospital
18	Tohoku	Tohoku University Hospital
19	Tohoku	Miyagi Children's Hospital
20	Tohoku	Yamagata University Hospital
21	Tohoku	Sendai City Hospital
22	Kanto and Koshinetsu	Ibaraki Children's Hospital
23	Kanto and Koshinetsu	Tsukuba University Hospital

Table 4 continued

S. no.	District	Institutions
24	Kanto and Koshinetsu	Yokohama City University Hospital
25	Kanto and Koshinetsu	Saiseikai Yokohama City Nanbu Hospital
26	Kanto and Koshinetsu	Kitasato University Hospital
27	Kanto and Koshinetsu	Tokai University Hospital
28	Kanto and Koshinetsu	Showa University Fujigaoka Hospital
29	Kanto and Koshinetsu	Kanagawa Children's Medical Center
30	Kanto and Koshinetsu	St. Marianna University School of Medicine Hospital
31	Kanto and Koshinetsu	Gunma Children's Medical Center
32	Kanto and Koshinetsu	Gunma University Hospital
33	Kanto and Koshinetsu	Saitama Medical Center
34	Kanto and Koshinetsu	Saitama Children's Medical Center
35	Kanto and Koshinetsu	National Defense Medical College Hospital
36	Kanto and Koshinetsu	Teikyo University Chiba Medical Center
37	Kanto and Koshinetsu	Kameda Medical Center
38	Kanto and Koshinetsu	Nippon Medical School Chiba Hokusoh Hospital
39	Kanto and Koshinetsu	Kokuho Asahi General Hospital
40	Kanto and Koshinetsu	Japanese Red Cross Narita Hospital
41	Kanto and Koshinetsu	Chiba University Hospital
42	Kanto and Koshinetsu	Chiba Children's Hospital
43	Kanto and Koshinetsu	Matsudo City Hospital
44	Kanto and Koshinetsu	National Center for Global Health and Medicine
45	Kanto and Koshinetsu	Nihon University Itabashi Hospital
46	Kanto and Koshinetsu	Japanese Red Cross Musashino Hospital
47	Kanto and Koshinetsu	Teikyo University Hospital
48	Kanto and Koshinetsu	Tokyo Medical And Dental University Hospital Faculty of Medicine
49	Kanto and Koshinetsu	The Jikei University Daisan Hospital
50	Kanto and Koshinetsu	Tokyo Metropolitan Children's Medical Center
51	Kanto and Koshinetsu	The Jikei University Hospital
52	Kanto and Koshinetsu	Nippon medical School Hospital
53	Kanto and Koshinetsu	Tokyo Women's Medical University Medical Center East
54	Kanto and Koshinetsu	The University of Tokyo Hospital
55	Kanto and Koshinetsu	Keio University Hospital
56	Kanto and Koshinetsu	Tokyo Metropolitan Cancer and Infectious diseases Center Komagome Hospital
57	Kanto and Koshinetsu	Toho University Omori Medical Center
58	Kanto and Koshinetsu	Showa University Hospital
59	Kanto and Koshinetsu	Juntendo University Hospital
60	Kanto and Koshinetsu	National Center for Child Health and Development
61	Kanto and Koshinetsu	St. Luke's International Hospital
62	Kanto and Koshinetsu	Kyorin University Hospital
63	Kanto and Koshinetsu	Tokyo Dental College Ichikawa General Hospital
64	Kanto and Koshinetsu	Dokkyo Medical University Hospital
65	Kanto and Koshinetsu	Jichi Medical University Hospital
66	Kanto and Koshinetsu	Shinshu University Hospital
67	Kanto and Koshinetsu	Nagano Children's Hospital
68	Kanto and Koshinetsu	Niigata University Medical and Dental Hospital
69	Kanto and Koshinetsu	Niigata Cancer Center Hospital
70	Kanto and Koshinetsu	University of Yamanashi Hospital
71	Kanto and Koshinetsu	Japanese Red Cross Maebashi Hospital

Table 4 continued

S. no.	District	Institutions
72	Kanto and Koshinetsu	Saitama Medical University International Medical Center
73	Kanto and Koshinetsu	Yokosuka Kyosai Hospital
74	Kanto and Koshinetsu	Kofu Municipal Hospital
75	Kanto and Koshinetsu	Teikyo University School of medicine University Hospital, Mizonokuchi
76	Tokai and Hokuriku	Fujita Health University
77	Tokai and Hokuriku	Aichi Medical University Hospital
78	Tokai and Hokuriku	Komaki City Hospital
79	Tokai and Hokuriku	National Hospital Organization Nagoya Medical Center
80	Tokai and Hokuriku	Nagoya Daini Red Cross Hospital
81	Tokai and Hokuriku	Anjo Kosei Hospital
82	Tokai and Hokuriku	Japanese Red Cross Nagoya Daiichi Hospital
83	Tokai and Hokuriku	Nagoya University Hospital
84	Tokai and Hokuriku	Kasugai Municipal Hospital
85	Tokai and Hokuriku	Nagoya City University Hospital
86	Tokai and Hokuriku	Toyohashi Municipal Hospital
87	Tokai and Hokuriku	Ichinomiya Municipal Hospital
88	Tokai and Hokuriku	Okazaki City Hospital
89	Tokai and Hokuriku	Kanazawa University Hospital
90	Tokai and Hokuriku	Ishikawa Prefectural Central Hospital
91	Tokai and Hokuriku	Kanazawa Medical University Hospital
92	Tokai and Hokuriku	Gifu Municipal Hospital
93	Tokai and Hokuriku	Toki Municipal General Hospital
94	Tokai and Hokuriku	Gifu University Hospital
95	Tokai and Hokuriku	Hamamatsu Medical Center
96	Tokai and Hokuriku	Hamamatsu University School of Medicine, University Hospital
97	Tokai and Hokuriku	Shizuoka Children's Hospital
98	Tokai and Hokuriku	Iwata City Hospital
99	Tokai and Hokuriku	Seirei Hamamatsu General Hospital
100	Tokai and Hokuriku	Toyama University Hospital
101	Tokai and Hokuriku	Fukui Red Cross Hospital
102	Tokai and Hokuriku	University of Fukui Hospital
103	Tokai and Hokuriku	Mie University Hospital
104	Tokai and Hokuriku	National Mie Hospital
105	Tokai and Hokuriku	Nagoya City East Medical Center
106	Kinki	National Hospital Organization Osaka National Hospital
107	Kinki	Osaka City University Hospital
108	Kinki	Kinki University Hospital
109	Kinki	Yao Municipal Hospital
110	Kinki	Matsushita Memorial Hospital
111	Kinki	Osaka Medical Center and Research Institute for Maternal and Child Health
112	Kinki	Toyonaka Municipal Hospital
113	Kinki	Osaka University Hospital
114	Kinki	Sakai Hospital Kinki University Faculty of Medicine
115	Kinki	Osaka Medical College Hospital
116	Kinki	Kansai Medical University Hirakata Hospital
117	Kinki	Kitano Hospital, The Tazuke Kofukai Medical Research Institute
118	Kinki	Osaka City General Hospital
119	Kinki	Osaka Red Cross Hospital

Table 4 continued

S. no.	District	Institutions
120	Kinki	Osaka General Medical Center
121	Kinki	Nakano Children's Hospital
122	Kinki	Kishiwada City Hospital
123	Kinki	Japanese Red Cross Kyoto Daiichi Hospital
124	Kinki	Kyoto-Katsura Hospital
125	Kinki	Kyoto University Hospital
126	Kinki	Kyoto City Hospital
127	Kinki	National Hospital Organization Maizuru Medical Center
128	Kinki	University Hospital, Kyoto Prefectural University of Medicine
129	Kinki	Takashima General Hospital
130	Kinki	Shiga University of Medical Science Hospital
131	Kinki	Shiga Medical Center for Children
132	Kinki	Otsu Red Cross Hospital
133	Kinki	Tenri Hospital
134	Kinki	Nara Medical University Hospital
135	Kinki	Kobe University Hospital
136	Kinki	Kobe City Medical Center General Hospital
137	Kinki	Japanese Red Cross Society Himeji Hospital
138	Kinki	Akashi Municipal Hospital
139	Kinki	Hyogo Prefectural Kobe Children's Hospital
140	Kinki	Hyogo College of Medicine Hospital
141	Kinki	Nishi-Kobe Medical Center
142	Kinki	Japanese Red Cross Society Wakayama Medical Center
143	Kinki	Wakayama Medical University Hospital
144	Chugoku and Shikoku	Ehime Prefectural Central Hospital
145	Chugoku and Shikoku	Ehime University Hospital
146	Chugoku and Shikoku	National Hospital Organization Okayama Medical Center
147	Chugoku and Shikoku	Okayama University Hospital
148	Chugoku and Shikoku	Okayama Saiseikai General Hospital
149	Chugoku and Shikoku	Kawasaki Medical School Hospital
150	Chugoku and Shikoku	Kurashiki Central Hospital
151	Chugoku and Shikoku	National Hospital Organization Kagawa Children's Hospital
152	Chugoku and Shikoku	Kagawa University Hospital
153	Chugoku and Shikoku	National Hospital Organization Kochi Medical Center
154	Chugoku and Shikoku	Japanese Red Cross Kochi Hospital
155	Chugoku and Shikoku	Kochi Medical School Hospital
156	Chugoku and Shikoku	Shimane University Hospital
157	Chugoku and Shikoku	Shimane Prefectural Central Hospital
158	Chugoku and Shikoku	Tokushima University Hospital
159	Chugoku and Shikoku	Tottori University Hospital
160	Chugoku and Shikoku	Tottori Prefectural Chuou Hospital
161	Chugoku and Shikoku	Hiroshima University Hospital
162	Chugoku and Shikoku	Hiroshima Red Cross Hospital and Atomic-bomb Survivors Hospital
163	Chugoku and Shikoku	Yamaguchi University Hospital
164	Chugoku and Shikoku	Tokushima Red Cross Hospital
165	Chugoku and Shikoku	Matsue Red Cross Hospital
166	Kyushu and Okinawa	National Hospital Organization Beppu Medical Center
167	Kyushu and Okinawa	Oita Prefectural Hospital

Table 4 continued

S. no.	District	Institutions
168	Kyushu and Okinawa	Oita University Hospital
169	Kyushu and Okinawa	Hospital, University of the Ryukyus
170	Kyushu and Okinawa	Okinawa Prefectural Nanbu Medical Center and Children's Medical Center
171	Kyushu and Okinawa	Kagoshima City Hospital
172	Kyushu and Okinawa	Kagoshima University Medical And Dental Hospital
173	Kyushu and Okinawa	National Hospital Organization Kumamoto Medical Center
174	Kyushu and Okinawa	Kumamoto university hospital
175	Kyushu and Okinawa	Japanese Red Cross Kumamoto Hospital
176	Kyushu and Okinawa	Saga University Hospital
177	Kyushu and Okinawa	Nagasaki University Hospital
178	Kyushu and Okinawa	Kitakyushu Municipal Medical Center
179	Kyushu and Okinawa	Center for Pediatric Emergency Medicine Kitakyushu Municipal Yahata Hospital
180	Kyushu and Okinawa	Kurume University Hospital
181	Kyushu and Okinawa	University Hospital of Occupational and Environmental Health
182	Kyushu and Okinawa	Kyushu University Hospital
183	Kyushu and Okinawa	National Hospital Organization Kyushu Cancer Center
184	Kyushu and Okinawa	Fukuoka University Hospital
185	Kyushu and Okinawa	University of Miyazaki Hospital
186	Kyushu and Okinawa	Sasebo Municipal General Hospital
187	Kyushu and Okinawa	General Hospital Hamanomachi

References

1. The Research Program for the Treatment of Chronic Pediatric Diseases of Specified Categories: Department of Health Policy. Available from URL: http://www.nch.go.jp/policy/shoumann.htm. Accessed October, 2012.
2. Research group of the Treatment of Chronic Pediatric Diseases of Specified Categories. Available from URL: http://www.aiiku.or.jp/aiiku/mch/syoman/syo.html. Accessed October, 2012.
3. Matsuda T, Marugame T, Kamo K, Katanoda K, Ajiki W, Sobue T. Cancer incidence and incidence rates in Japan in 2006: based on data from 15 population-based cancer registries in the monitoring of cancer incidence in Japan (MCIJ) project. Jpn J Clin Oncol. 2012;42(2):139–47.
4. Ikeda H, Matsuyama S, Tanimura M. Association between hepatoblastoma and very low birth weight: a trend or a chance? J Pediatr. 1997;130(4):557–60.
5. The Committee of the Japan Children's Cancer Registry. The Japan Children's Cancer Registry: outline, results, and perspectives. In: Watanabe S, Tominaga S, Kakizoe T, editors. Cancer Treatment and survival. Tokyo: CRC Press; 1995.
6. Ajiki W, Hanai A, Tsukuma H, Hiyama T, Fujimoto I. Incidence of childhood cancer in Osaka, Japan, 1971–1988: reclassification of registered cases by Birch's scheme using information on clinical diagnosis, histology and primary site. Jpn J Cancer Res. 1994;85(2):139–46.
7. Conter V, Arico M, Basso G, Biondi A, Barisone E, Messina C, et al. Long-term results of the Italian Association of Pediatric Hematology and Oncology (AIEOP) Studies 82, 87, 88, 91 and 95 for childhood acute lymphoblastic leukemia. Leukemia. 2010;24(2):255–64.
8. Hunger SP, Lu X, Devidas M, Camitta BM, Gaynon PS, Winick NJ, et al. Improved survival for children and adolescents with acute lymphoblastic leukemia between 1990 and 2005: a report from the children's oncology group. J Clin Oncol. 2012;30(14):1663–9.
9. Mitchell C, Richards S, Harrison CJ, Eden T. Long-term follow-up of the United Kingdom medical research council protocols for childhood acute lymphoblastic leukaemia, 1980–2001. Leukemia. 2010;24(2):406–18.
10. Moricke A, Zimmermann M, Reiter A, Henze G, Schrauder A, Gadner H, et al. Long-term results of five consecutive trials in childhood acute lymphoblastic leukemia performed by the ALL-BFM study group from 1981 to 2000. Leukemia. 2010;24(2):265–84.
11. Schmiegelow K, Forestier E, Hellebostad M, Heyman M, Kristinsson J, Soderhall S, et al. Long-term results of NOPHO ALL-92 and ALL-2000 studies of childhood acute lymphoblastic leukemia. Leukemia. 2010;24(2):345–54.
12. Veerman AJ, Kamps WA, van den Berg H, van den Berg E, Bokkerink JP, Bruin MC, et al. Dexamethasone-based therapy for childhood acute lymphoblastic leukaemia: results of the prospective Dutch Childhood Oncology Group (DCOG) protocol ALL-9 (1997–2004). Lancet Oncol. 2009;10(10):957–66.
13. Vrooman LM, Silverman LB. Childhood acute lymphoblastic leukemia: update on prognostic factors. Curr Opin Pediatr. 2009;21(1):1–8.
14. Pui CH, Mullighan CG, Evans WE, Relling MV. Pediatric acute lymphoblastic leukemia: where are we going and how do we get there? Blood. 2012;120(6):1165–74.
15. Disease Registry Committee for the Japanese Society of Pediatric Hematology. Annual Report of Nationwide Survey 2008. Jpn J Pediatr Hematol. 2009;23:213–9.
16. Disease Registry Committee for the Japanese Society of Pediatric Hematology. Annual Report of Nationwide Survey 2009. Jpn J Pediatr Hematol. 2010;24:182–9.

17. Disease Registry Committee for the Japanese Society of Pediatric Hematology, Diagnostic guidelines for hematological diseases. Jpn J Pediatr Hematol. 2009;21 Suppl 5/6:1–76.
18. Rubnitz JE, Pui CH. Recent advances in the treatment and understanding of childhood acute lymphoblastic leukaemia. Cancer Treat Rev. 2003;29(1):31–44.
19. Jemal A, Siegel R, Ward E, Hao Y, Xu J, Thun MJ. Cancer statistics, 2009. CA Cancer J Clin. 2009;59(4):225–49.
20. Coebergh JW, Reedijk AM, de Vries E, Martos C, Jakab Z, Steliarova-Foucher E, et al. Leukaemia incidence and survival in children and adolescents in Europe during 1978–1997. Report from the Automated Childhood Cancer Information System project. Eur J Cancer. 2006;42(13):2019–36.
21. Rubnitz JE, Inaba H. Childhood acute myeloid leukaemia. Br J Haematol. 2012;159(3):259–76.
22. Amadori S, Testi AM, Arico M, Comelli A, Giuliano M, Madon E, et al. Prospective comparative study of bone marrow transplantation and postremission chemotherapy for childhood acute myelogenous leukemia. The Associazione Italiana Ematologia ed Oncologia Pediatrica Cooperative Group. J Clin Oncol. 1993;11(6):1046–54.
23. Webb DK, Harrison G, Stevens RF, Gibson BG, Hann IM, Wheatley K. Relationships between age at diagnosis, clinical features, and outcome of therapy in children treated in the Medical Research Council AML 10 and 12 trials for acute myeloid leukemia. Blood. 2001;98(6):1714–20.
24. Woods WG, Neudorf S, Gold S, Sanders J, Buckley JD, Barnard DR, et al. A comparison of allogeneic bone marrow transplantation, autologous bone marrow transplantation, and aggressive chemotherapy in children with acute myeloid leukemia in remission. Blood. 2001;97(1):56–62.
25. Tsukimoto I, Tawa A, Horibe K, Tabuchi K, Kigasawa H, Tsuchida M, et al. Risk-stratified therapy and the intensive use of cytarabine improves the outcome in childhood acute myeloid leukemia: the AML99 trial from the Japanese Childhood AML Cooperative Study Group. J Clin Oncol. 2009;27(24):4007–13.
26. Imaizumi M, Tawa A, Hanada R, Tsuchida M, Tabuchi K, Kigasawa H, et al. Prospective study of a therapeutic regimen with all-trans retinoic acid and anthracyclines in combination of cytarabine in children with acute promyelocytic leukaemia: the Japanese childhood acute myeloid leukaemia cooperative study. Br J Haematol. 2011;152(1):89–98.
27. Kudo K, Kojima S, Tabuchi K, Yabe H, Tawa A, Imaizumi M, et al. Prospective study of a pirarubicin, intermediate-dose cytarabine, and etoposide regimen in children with Down syndrome and acute myeloid leukemia: the Japanese Childhood AML Cooperative Study Group. J Clin Oncol. 2007;25(34):5442–7.
28. Imamura T, Iwamoto S, Kanai R, Shimada A, Terui K, Osugi Y, et al. Outcome in 146 patients with paediatric acute myeloid leukaemia treated according to the AML99 protocol in the period 2003–06 from the Japan Association of Childhood Leukaemia Study. Br J Haematol. 2012;159(2):204–10.
29. Koga Y, Kumagai M, Takimoto T, Mimaya J, Nakazawa A, Horibe K, et al. Retrospective analysis of 157 patients with pediatric Hodgkin lymphoma in Japan: investigation by four pediatric cancer study groups. Rinsho Ketsueki. 2012;53(4):443–9.
30. Manipadam MT, Nair S, Viswabandya A, Mathew L, Srivastava A, Chandy M. Non-Hodgkin lymphoma in childhood and adolescence: frequency and distribution of immunomorphological types from a tertiary care center in South India. World J Pediatr. 2011;7(4):318–25.
31. Wright D, McKeever P, Carter R. Childhood non-Hodgkin lymphomas in the United Kingdom: findings from the UK Children's Cancer Study Group. J Clin Pathol. 1997;50(2):128–34.
32. Spitz MR, Sider JG, Johnson CC, Butler JJ, Pollack ES, Newell GR. Ethnic patterns of Hodgkin's disease incidence among children and adolescents in the United States, 1973–82. J Natl Cancer Inst. 1986;76(2):235–9.
33. Percy CL, Smith MA, Linet M, Gloeckler Ries LA, Friedman DL. Lymphoma and reticuloendothelial neoplasms. In: Ries LAG, Smith MA, Gurney JG, et.al., eds. Cancer incidence and survival among children and adolescents. United States SEER Program 1975–1995. Bethesda, MD: National Cancer Institute, SEER Program, 1999;35–49. Available at: http://www.acco.org/LinkClick.aspx?fileticket=CGsKkvHaH1Y%3D&tabid=670. Accessed November, 2012.
34. Fujita N, Kobayashi R, Takimoto T, Nakagawa A, Ueda K, Horibe K. Results of the Japan Association of Childhood Leukemia Study (JACLS) NHL-98 protocol for the treatment of B-cell non-Hodgkin lymphoma and mature B-cell acute lymphoblastic leukemia in childhood. Leuk Lymphoma. 2011;52(2):223–9.
35. Kikuchi A, Mori T, Fujimoto J, Kumagai M, Sunami S, Okimoto Y, et al. Outcome of childhood B-cell non-Hodgkin lymphoma and B-cell acute lymphoblastic leukemia treated with the Tokyo Children's Cancer Study Group NHL B9604 protocol. Leuk Lymphoma. 2008;49(4):757–62.
36. Tsurusawa M, Katano N, Hirota T, Ito M, Yanase T, Asami K, et al. Studies of childhood non-Hodgkin's lymphoma-treatment results with the CCLSG NHL 960 protocol. Children's Cancer and Leukemia Study Group (CCLSG). Rinsho Ketsueki. 1998;39(11):1092–8.
37. Reiter A, Schrappe M, Tiemann M, Ludwig WD, Yakisan E, Zimmermann M, et al. Improved treatment results in childhood B-cell neoplasms with tailored intensification of therapy: a report of the Berlin-Frankfurt-Munster Group Trial NHL-BFM 90. Blood. 1999;94(10):3294–306.
38. Bhatia S, Nesbit ME Jr, Egeler RM, Buckley JD, Mertens A, Robison LL. Epidemiologic study of Langerhans cell histiocytosis in children. J Pediatr. 1997;130(5):774–84.
39. Morimoto A, Ikushima S, Kinugawa N, Ishii E, Kohdera U, Sako M, et al. Improved outcome in the treatment of pediatric multifocal Langerhans cell histiocytosis: results from the Japan Langerhans Cell Histiocytosis Study Group-96 protocol study. Cancer. 2006;107(3):613–9.
40. Ishii E, Ohga S, Imashuku S, Yasukawa M, Tsuda H, Miura I, et al. Nationwide survey of hemophagocytic lymphohistiocytosis in Japan. Int J Hematol. 2007;86(1):58–65.
41. Trottestam H, Horne A, Arico M, Egeler RM, Filipovich AH, Gadner H, et al. Chemoimmunotherapy for hemophagocytic lymphohistiocytosis: long-term results of the HLH-94 treatment protocol. Blood. 2011;118(17):4577–84.
42. Kikuchi A, Hasegawa D, Ohtsuka Y, Hamamoto K, Kojima S, Okamura J, et al. Outcome of children with refractory anaemia with excess of blast (RAEB) and RAEB in transformation (RAEB-T) in the Japanese MDS99 study. Br J Haematol. 2012;158(5):657–61.
43. Hasegawa D, Manabe A, Yagasaki H, Ohtsuka Y, Inoue M, Kikuchi A, et al. Treatment of children with refractory anemia: the Japanese Childhood MDS Study Group trial (MDS99). Pediatr Blood Cancer. 2009;53(6):1011–5.
44. Yoshida N, Hirabayashi S, Watanabe S, Zaike Y, Tsuchida M, Yoshimi A, et al. Prognosis of 75 patients with juvenile myelomonocytic leukemia: prospective study by MDS committee in the Japanese Society of Pediatric Hematology. Rinsho Ketsueki. 2011;52(12):1853–8.

小児がん診療に関する放射線治療の実態調査

副島　俊典[1]*，正木　英一[2]，原　純一[3]，田口　智章[4]，上田　孝文[5]，堀部　敬三[6]，
柳澤　隆昭[7]，吉峰　俊樹[8]，柴田亜希子[9]，小田　慈[10]，瀧本　哲也[11]，多田羅竜平[12]

[1]兵庫県立がんセンター放射線治療科
[2]総合南東北病院小児がん放射線治療研究所
[3]大阪市立総合医療センター小児血液腫瘍科
[4]九州大学小児外科
[5]国立病院機構大阪医療センター整形外科
[6]国立病院機構名古屋医療センター臨床研究センター
[7]埼玉医科大学国際医療センター脳脊髄腫瘍科
[8]大阪大学脳神経外科
[9]国立がん研究センターがん対策情報センター
[10]岡山大学病院小児血液腫瘍科
[11]国立成育医療研究センター臨床研究推進室
[12]大阪市立総合医療センター緩和医療科

National Survey of Radiation Therapy about Management of Pediatric Cancer

Toshinori Soejima[1]*, Hidekazu Masaki[2], Junichi Hara[3], Tomoaki Taguchi[4], Takafumi Ueda[5], Keizo Horibe[6], Takaaki Yanagisawa[7], Toshiki Yoshimine[8], Akiko Shibata[9], Megumi Oda[10], Tetsuya Takimoto[11], Ryuhei Tatara[12]

[1]*Department of Radiation Oncology, Hyogo Cancer Center*
[2]*Research Institute for Pediatric Radiation Oncology, Southern Tohoku General Hospital*
[3]*Department of Pediatric Hematology and Oncology, Osaka City General Hospital*
[4]*Department of Pediatric Surgery, Kyushu University Graduate School of Medicine*
[5]*Department of Orthopaedic Surgery, National Hospital Organization Osaka Medical Center*
[6]*National Hospital Organization Nagoya Medical Center, Clinical Research Center*
[7]*Division of Pediatric Neuro-Oncology, Department of Neuro-Oncology/Neurosurgery, Comprehensive Cancer Center, International Medical Center, Saitama Medical University*
[8]*Department of Neurosurgery, Osaka University Graduate School of Medicine*
[9]*National Cancer Center Division of Cancer Information Services and Surveillance*
[10]*Department of Pediatric Hematology and Oncology, Okayama University Hospital*
[11]*Division of Clinical Research, National Medical Center for Children and Mothers, National Center for Child Health and Development*
[12]*Department of Palliative Medicine, Osaka General Hospital*

Abstract

We conducted a survey on the treatment system of radiation therapy based on the number of cases treated with radiation therapy, and opinions of treatment for pediatric cancer in 113 radiation departments of hospitals certified by JASTRO and children's hospitals. The response to our questionnaire was 66% (75 facilities): 71 departments answered radiation therapy for pediatric cancer was possible. However, there were few facilities where special radiation therapy devices, such as IMRT, were used as sedative necessary for pediatric cancer treatment. In addition, many radiation oncologists thought that centralized treatment for pediatric cancer is necessary.

Key words: pediatric cancer, radiation therapy, national survey

要　旨

放射線腫瘍学会認定施設および小児病院合計113施設に放射線治療の診療体制，放射線治療診療実績，今後の小児がん放射線治療に対する方向性についてアンケート調査を行った．回答は75施設（66%）で得られた．その結果，小児がんの放射線治療は多くの施設（71施設）が可能であると回答した．しかし，強度変調放射線治療などの特殊な放射線治療装置が鎮静の必要な小児にも使用される施設は少数であった．また，多くの放射線治療医が小児がんの放射線治療は集約すべきであると考えていた．

キーワード：小児がん，放射線治療，実態調査

2013年4月5日受付，2013年12月5日受理
* 別刷請求先：〒673-8558 明石市北王子町13-70
兵庫県立がんセンター　副島俊典
E mail: soe@hp.pref.hyogo.jp

I　はじめに

がん対策推進基本計画が見直され[1]，小児がん対策の充実も求められるようになった．希少疾患である小児がんは

表1　アンケート（放射線科用）

1. 施設と診療体制について	
①がん診療連携拠点病院の指定	国指定都道府県拠点　国指定地域拠点　都道府県指定拠点 いずれでもない
②小児の放射線治療は可能ですか	はい　いいえ
③ガンマナイフ	鎮静の必要な小児にも可能　成人のみ　実施していない
④サイバーナイフ	鎮静の必要な小児にも可能　成人のみ　実施していない
⑤強度変調照射	鎮静の必要な小児にも可能　成人のみ　実施していない
⑥放射線治療専門医	専従　診断と兼務　非常勤
⑦専従医がおられる場合，何名でしょうか	名
⑧医学物理士	いる　いない
⑨放射線治療専門放射線技師	いる　いない
⑩鎮静が必要な場合，だれが鎮静処置を行っていますか	放射線科医　小児科医　麻酔科医
⑪集学的診療体制で診療が行われていますか	はい　いいえ
⑫小児がんを対象としたキャンサーボードが行われていますか	はい　いいえ
2. 診療実績について	
年間の小児の放射線治療初診症例の概数をお答えください.	
造血幹細胞移植前処置としての全身照射	0 1–5 6–10 11–15 16–20 21
脳腫瘍	0 1–5 6–10 11–15 16–20 21
骨軟部腫瘍	0 1–5 6–10 11–15 16–20 21
その他の固形がん	0 1–5 6–10 11–15 16–20 21
上記の合計は何例程度ですか	0 1–5 6–10 11–15 16–20 21
3. 今後の小児がん放射線治療に対する方向について	
①今後積極的に症例を増やしていきたい	はい　いいえ
②現状維持でよい	はい　いいえ
今後，取り扱いをやめたい/取り扱う予定はない	はい　いいえ
小児の放射線治療はできればやりたくない	はい　いいえ
上記の理由	自由記載
小児の放射線治療は集約すべきとお考えでしょうか	はい　いいえ
その理由	自由記載

症例が分散されると治療の質も問題となる．そこで全国で15病院が小児がん拠点病院に決定され，小児がんの集約化も検討されている．

小児がんの集学的治療のなかで，放射線治療は重要である[2]．しかし，放射線治療の現場は急激な症例数の増加と高精度化による治療時間の増加のため非常に多忙であり，この状況下で小児がんに関わることの困難さも予想される．さらに放射線治療技師や医学物理士の欠如などの問題もあり，小児がんの放射線治療の質の担保も早急に解決すべき問題である．小児がん診療は専門的知識が必要であるにもかかわらず，小児がん治療経験の少ない放射線治療医が多いことも問題となる．

われわれは小児がん拠点病院において放射線治療の質の確保のためにはどのような指定要件が必要か検討するために，放射線治療の主要な施設にアンケート調査を実施した．既に小児がん拠点病院の指定要件は決定され，小児がん拠点病院も発表されているが，小児がんの放射線治療は必ずしも十分とはいえない状況であり，小児がんの放射線治療の実情を検討することは重要と考え，アンケート調査の結果を報告する．

II　対象および方法

放射線腫瘍学会認定施設およびリニアックを有する小児病院合計113施設に診療体制，診療実績，今後の小児がん放射線治療に対する方向性についてのアンケート調査を行った（表1）．アンケート調査は2012年4月から郵送で行った．

回答が得られた施設は75施設（66%）であった．そのなかでがん診療連携拠点病院の指定を受けているのは国指定都道府県がん診療連携拠点病院（以下国指定都道府県）が24施設，国指定地域がん診療連携拠点病院（以下国指定地域）が26施設，都道府県指定がん拠点病院（以下都道府県指定）が16施設であった．いずれでもない施設（指定なし）は9施設であり，うち小児病院が6施設あった．

III　結　果

1. 放射線治療の診療体制について（表2）

1）小児がん放射線治療の可否

小児がんの放射線治療が可能かどうかの質問では71施

表2　放射線治療の診療体制について

小児の放射線治療は可能か				
可能	71	95%		
不可能	3	4%		
無記載	1	1%		
特殊照射装置		ガンマナイフ	サイバーナイフ	IMRT
鎮静の必要な小児にも可能		3 (4%)	3 (4%)	24 (32%)
成人のみ		1 (1%)	2 (3%)	27 (36%)
実施していない		68 (91%)	68 (91%)	22 (29%)
無記載		3 (4%)	2 (3%)	2 (3%)
放射線治療に関わるスタッフ				
放射線治療専門医				
専従	70	93%		
診断と兼務	0	0%		
非常勤	5	7%		
医学物理士				
いる	54	72%		
いない	20	27%		
無記載	1	1%		
放射線治療技師				
いる	69	92%		
いない	5	7%		
無記載	1	1%		
鎮静施行医（重複あり）				
放射線科医	0	0%		
小児科医	66	88%		
麻酔科医	10	13%		
無記載	6	8%		
集学的診療体制				
行われている	66	88%		
行われていない	9	12%		
小児がんを対象としたキャンサーボード				
実施している	32	43%		
実施していない	39	52%		
無記載	4	5%		

IMRT; intensity modulated radiation therapy 強度変調放射線治療

設（95%が可能と答えた．不可能と答えたのは3施設（4%）のみで，国指定都道府県で1施設，都道府県指定で1施設，いずれでもない施設が1施設の3施設であった．不可能と答えた国指定都道府県の1施設は15歳以上を対象疾患としており，同じ都道府県内に小児病院がある施設であった．

2）特殊放射線治療

ガンマナイフ，サイバーナイフ，強度変調放射線治療（Intensity Modulated Radiation Therapy，以下IMRT）が小児がんに使用できるかどうかについて調査した．鎮静の必要な小児にもガンマナイフが可能な施設は3施設（4%）（国指定地域2，都道府県指定1），成人のみ使用可能な施設が1施設（1%）（都道府県指定1），実施していない施設が68施設（91%）であった．

サイバーナイフに関しては，鎮静の必要な小児にも可能な施設が3施設（4%）（国指定都道府県2，国指定地域1），成人のみ使用可能な施設が2施設（国指定都道府県1，国指定地域1），実施していない施設が68施設（91%）であった．

IMRTに関しては，鎮静の必要な小児にも可能な施設が24施設（32%）（国指定都道府県11，国指定地域10，都道府県指定2，指定なし1），成人のみ可能な施設が27施設（36%）（国指定都道府県8，国指定地域11，都道府県指定6，指定なし2），実施していない施設が22施設（25%）であった．小児病院でIMRTを施行している施設はなかった．

3）放射線治療の関わるスタッフ

放射線治療専門医が専従しているのが70施設（90%）で，放射線診断医が放射線治療を兼務している施設はなかった．非常勤の施設が5施設（7%）で，全て小児病院であった．

表3　放射線治療診療実績（年間小児がん放射線治療初診症例数）

	全身照射	脳腫瘍	骨軟部	その他	合計
0例	23	10	22	20	7
1–5例	33	46	47	43	18
6–10例	12	11	5	8	22
11–15例	5	5	0	1	10
16–20例	0	3	0	0	5
21例以上	0	0	0	2	10

医学物理士が従事する施設が54施設（72%）で，小児病院では1施設のみであった．放射線治療技師は大部分の施設に従事していた．

4）放射線治療施行時の鎮静施行医

鎮静が必要な場合，鎮静処置を行うのは多くは小児科医（66施設88%）で，麻酔科医が行う施設は10施設（13%），放射線腫瘍医が行う施設はなかった．

5）集学的診療体制

66施設（88%）が集学的診療体制で診療が行われていると回答し，9施設（12%）は行われていないと回答していた．小児がんを対象としたキャンサーボードが行われている施設が32施設（43%）であった．

2. 小児がんの放射線治療診療実績について（表3）

年間の小児がんの放射線治療初診症例数では，0例7施設（10%），1例から5例18施設（25%），6例から10例22施設（31%），11例から15例10施設（14%），16例から20例5施設（7%）で，21症例以上の症例数がある施設が10施設（14%）であった．疾患別には全身照射については0症例23施設11症例以上の施設が5施設であった．脳腫瘍，骨軟部腫瘍，その他の疾患に関して症例0の施設がそれぞれ10施設，22施設，20施設で，逆に11症例以上の施設がそれぞれ8施設，0施設，3施設であった．

3. 今後の小児がん放射線治療に対する方向について（表4）

今後積極的に症例を増やしていきたいと回答した施設は26施設（35%），いいえと答えた施設が45施設（60%）であった．また，現状維持でよいと回答した施設が52施設（69%），いいえと答えた施設が17施設（23%）であった．今後，取り扱いをやめたいもしくは取り扱う予定はないと回答した施設が6施設（8%）であったがこの回答に対するいいえと回答した施設は60施設（80%）であった．小児がんの放射線治療はできればやりたくないと回答した施設は，11施設（15%）であったが，いいえと答えた施設は57施設（76%）であった．小児がんの放射線治療について肯定的な回答をした施設の理由としては小児がん治療の中核病院であるからという回答が多く，否定的な意見の理由としては，専門的な知識が必要，時間的な制約などの回答が多かった．小児がんの放射線治療は集約化すべきと考えている施設は67施設（89%）で，いいえと答えた施設は2施設（3%）のみであった．多くの施設が集約化すべきと考えており，その理由として，症例数が少ない，専門性の問題を解答する施設が多かった．

表4　今後の小児がん放射線治療に対する方向について

今後積極的に症例を増やしていきたい		
はい	26	35%
いいえ	45	60%
無記載	4	5%
現状維持でよい		
はい	52	69%
いいえ	17	23%
無記載	6	8%
今後，取り扱いをやめたい／取り扱う予定はない		
はい	6	8%
いいえ	60	80%
無記載	9	12%
小児がんの放射線治療はできればやりたくない		
はい	11	15%
いいえ	57	76%
無記載	7	9%
小児がんの放射線治療は集約化すべきとお考えでしょうか		
はい	67	89%
いいえ	2	3%
無記載	6	8%

IV　考　察

今回のアンケート調査では小児の放射線治療が可能と回答する施設が多かったが，年間に経験する症例数が20症例以上経験する施設はわずかに14%と少数であった．また，小児がんの放射線治療の症例数を積極的に増やしていこうとする施設は35%と半数以下であり，それ程多くない実態が判明した．また，特殊な放射線治療機器も鎮静可能な小児には適応していないとの回答も多くみられた．特にIMRTにおいては小児がんにおける有用性が指摘されているにもかかわらず[3]，小児がんに適応せずに成人のみに適応すると答えている施設が27施設あった．臓器合併症を減ずるためにはIMRTは小児がんの放射線治療には有効であるが，小児を扱う煩雑さのためか適応としていないことは小児がんの治療に当たって大きな問題と感じられる．

また，小児病院での放射線治療の設備，スタッフの少なさも浮き彫りになった．放射線治療専門医が常勤でなく，非常勤の施設が5施設であり，医学物理士がいる小児病院では1施設しかなく，IMRTを行っている施設はなかった．小児がん治療の中心的役割を果たしていくべき小児病院が放射線治療に関して人材および設備的には十分でないこ

とがわかった．これは年間の放射線治療患者数が200名以下の場合は経営的に赤字となるといわれており，年間の患者数が50名以下の小児病院では設備投資や人材確保をするべき予算がないことが原因として考えられる．また，常勤の放射線腫瘍医が不在であることが，設備や医学物理士などのスタッフの充実を行えない理由であることも想像される．症例数が少なくても治療方針の決定や放射線治療患者の急性合併症や晩期合併症に対する対応，および院内外への医療スタッフへの教育など多くの業務が放射線腫瘍医に求められ，小児病院においても常勤化が必要と考えられる．

小児がんの放射線治療を全くしていない施設も多くみられた．治療の質向上には医師だけでなく，放射線技師や放射線治療に従事する看護師にも診療の経験が必要と思われる．症例数にかかわらず，質が保たれていれば問題はないが，症例数が少ない場合，その都度ガイドラインなどを見る必要があり，必要な子どものためのリソース（環境や人材）を用意できなかったり，多職種腫瘍カンファレンスが開催されていないなどの問題がありえる．多くの放射線腫瘍医は小児がんの集約化には賛成と答えていた．専門性の問題，希少疾患であること，時間など手間がかかることなどが理由であった．放射線腫瘍医への調査結果から放射線治療の観点からのみ考えると今後小児がんの治療施設の集約化の方向が望ましいと思われた．集約された小児がん拠点病院の放射線治療では晩期合併症を減らすためにIMRTが可能であることが望まれる．また，放射線治療の質の担保のために医学物理士の常勤が必要不可欠と思われる．しかし集約化は簡単に進むわけではないので，現状でどのようにこれらの問題に対処していくのか検討が必要と考える．

V 結　語

小児がんに対する放射線治療についてアンケート調査を行った．その結果IMRTなどの特殊な放射線治療装置が鎮静の必要な小児にも使用される施設が少なかった．また，多くの放射線治療施設が小児がんの放射線治療を集約すべきであると考えていた．

この研究は厚生労働省科学研究費補助金，がん対策推進基本計画とがん診療連携拠点病院の小児がん診療体制への適応に関する研究（研究代表者　原純一）の援助を受け，同研究の一部としてアンケート調査を行った．

文　献

1) 厚生労働省：がん対策推進基本計画　平成24年6月 http://www.mhlw.go.jp/bunya/kenkou/dl/gan_keikaku02.pdf
2) 野崎美和子：小児固形腫瘍における放射線治療の役割．小児がん，47: 419–423, 2010.
3) Sterzing F, Stoiber EM, Nill S, et al: Intensity modulated radiotherapy (IMRT) in the treatment of children and adolescents — a single institution's experience and a review of the literature. Radiat Oncol, 23: 37, 2009.