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Resuscitative endovascular balloon occlusion of the aorta versus aortic cross clamping among patients with critical trauma: a nationwide cohort study in Japan

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Abstract

Background: Measures of aortic occlusion (AO) for resuscitation in patients with severe torso trauma remain controversial. Our aim was to characterize the current use of resuscitative endovascular balloon occlusion of the aorta (REBOA) and resuscitative open aortic cross-clamping (ACC), and to evaluate whether REBOA should be an alternative method to resuscitative open ACC.

Methods: This study was a retrospective cohort study between 2004 and 2013 from a nationwide trauma registry in Japan. Participants were selected who underwent either REBOA or ACC. Their characteristics, interventions, and outcomes were analyzed to compare REBOA and ACC directly. The primary outcome was in-hospital mortality and the secondary outcome was mortality in the emergency department. Logistic regression analysis was performed to compare the outcomes between REBOA and ACC with adjustment for severity; 1:1 propensity score matching was also performed.

Results: Of the 159,157 trauma patients, 903 were eligible based on the selection criteria. Overall, 405/607 patients (67%) who had REBOA died compared to 210/233 patients (90%) who had ACC. Patients with REBOA had higher revised trauma score (RTS) (mean \pm SD, 5.2 ± 2.0 vs. 4.2 ± 2.2 ; $P < 0.001$) but higher Injury Severity Score (ISS) (median (interquartile); 34 (25) vs. 34 (20); $P < 0.001$), and higher probability of survival (0.43 ± 0.36 vs. 0.27 ± 0.30 ; $P < 0.001$) compared to those with ACC. REBOA had an odds ratio (OR) for in-hospital mortality of 0.309 (95% confidence interval (CI) = 0.190–0.502) adjusting for trauma and injury severity score using a logistic regression model ($n = 903$). Similar associations were observed adjusting for RTS (OR = 0.224; 95% CI = 0.129–0.700) or adjusting for ISS (OR, 0.188; 95% CI, 0.116 to 0.303). In the propensity score-matched cohort ($n = 304$), REBOA was associated with lower mortality compared to ACC (OR, 0.261; 95% CI, 0.130 to 0.523). Patients with REBOA had less severe chest complications than those with ACC (Abbreviated Injury Scale thorax, 3.8 ± 0.8 vs. 4.2 ± 0.8 ; $P < 0.001$), although physiological severity and backgrounds were similar in this population.

Conclusions: Patients who underwent AO had a high mortality. REBOA might be a favorable alternative method to resuscitative ACC for severe torso trauma although some indication bias could still remain. Further studies are needed to elucidate optimal indications.

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Background

Bleeding control is a critical strategy in the management of severe trauma patients. Aortic occlusion (AO) is a standard initial procedure to control blood loss in severe torso trauma patients which buys time for a more definitive treatment. Open aortic cross-clamping (ACC), established as just such a definitive approach, has been traditionally accomplished via emergent thoracotomy or as an initial step during laparotomy [1]. However, ACC for resuscitation in critical trauma patients remains controversial because of a very high mortality rate. In actuality, open ACC might suffer from a negative perception because it might be used in patients already beyond saving, thereby driving up the mortality rate. Recently, resuscitative endovascular balloon occlusion of the aorta (REBOA) has been used as an alternative method of ACC. REBOA has been previously described as useful for hemorrhagic shock in cases of ruptured abdominal aortic aneurysm [2], gastrointestinal bleeding [3], and in postpartum hemorrhage [4]. As they are useful in solving multiple problems, endovascular approaches such as trans-catheter arterial embolization (TAE) should become more widely used in trauma settings. However, there is a dearth of clinical reports with adequate sample size and situations on which to base recommendations [5, 6]. There are a few reports to show favorable outcomes of REBOA compared with ACC [1] but no concrete indications of REBOA or ACC efficacy exist at the time of this report. To this end, our aim was to analyze the present situation of REBOA and ACC usage with nationwide trauma registry data and to then evaluate as to whether or not REBOA should be deemed a preferential alternative to resuscitative ACC.

Methods

Study designs

We conducted a retrospective cohort study using registered data from the Japan Trauma Data Bank (JTDB) to compare characteristics and outcomes between REBOA and ACC.

Data collection

Data were obtained from the JTDB, a nationwide trauma registry established in 2003 and authorized and maintained by the Japanese Association for the Surgery of Trauma and the Japanese Association for Acute Medicine to improve and assure the quality of trauma care in Japan [7]. During the study period, a total of 234 hospitals including 95% of tertiary emergency medical centers in Japan participated in the JTDB [7]. The JTDB collected variables about patients and hospitals such as patient demographics, comorbidities, injury type, mechanism, vital signs, Abbreviated Injury Scale (AIS) score, Injury Severity Score (ISS), pre-hospital treatment, in-hospital

procedures, and in-hospital and emergency department (ED) mortality [7].

REBOA has recently found use as a general technique across major emergency centers in Japan. Due to the limitations of the JTDB as being a general, total trauma registry and not a REBOA-specific database, we were unable to glean information on imaging, access, and balloon zone placement specifics. Although details of REBOA in those three areas depend mainly on local facilities and expertise, in our specific cases REBOA access is typically accomplished through a common femoral artery and the balloon insertion follows a blind approach.

Patient selection

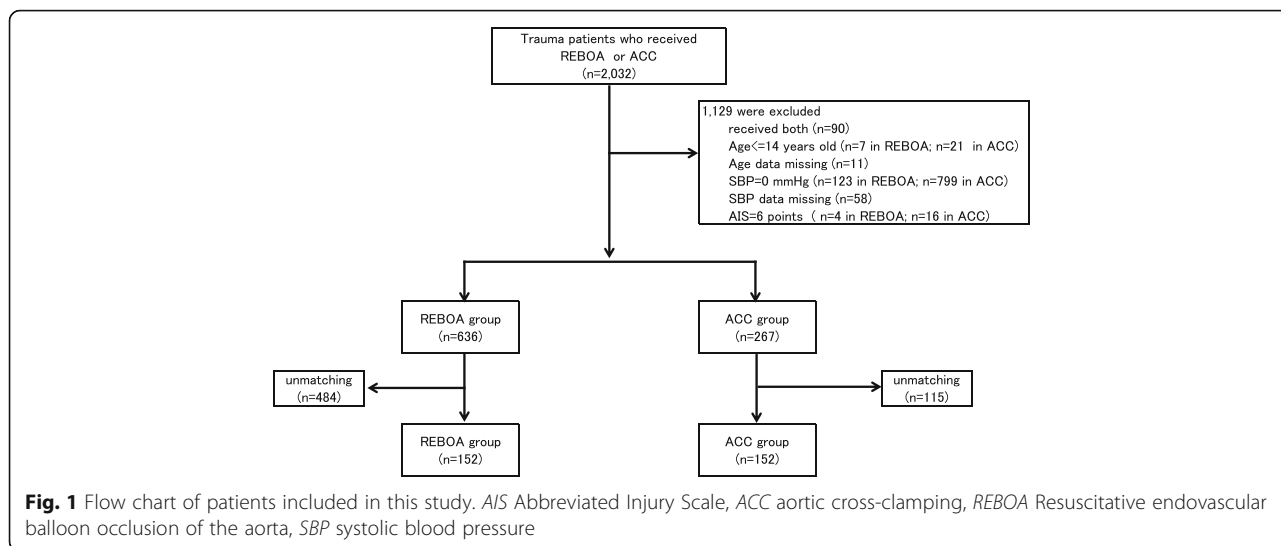
The study inclusion criteria were the presence of critical trauma and reception of either REBOA or ACC. We excluded patients who had received both REBOA and ACC. Also, we excluded subjects younger than 14 years old or those with age data missing. Patients with cardiopulmonary arrest on arrival at the ED (systolic blood pressure of 0 mm Hg or data missing on arrival) or with an AIS score of 6 (i.e., non-survivable injury) for any region were also excluded. Figure 1 shows participant selection data from this study.

Study endpoints

The primary intervention was either REBOA or ACC. Intervention strategies were solely dependent on the individual decisions of ED physicians. The primary outcome of this study was in-hospital mortality and the secondary outcome was mortality in the ED.

Statistical analysis

To report the characteristics of participants, firstly we used the Shapiro-Wilk test to identify normality of each variable. However, there was some skew in almost all the variables. We then decided whether to use the mean \pm standard deviation (SD) or median (interquartile range (IQR)) for plot figures of each variable, depending on the previous reporting style. To assess the independent effects of REBOA compared with those of ACC, outcomes were evaluated by analytical models—standard logistic regression and a logistic regression incorporating the results of propensity score matching. Since the number of survivors was very small in this study population, we chose a few covariates for the standard logistic regression analyses. Within the standard logistic regression analyses, we conducted three adjustment models: a revised trauma score (RTS)-adjusted model, an ISS-adjusted model, and a trauma and injury severity score (TRISS)-adjusted model. Together, these three standard logistic regression models, coupled with a propensity score-matched model, provide a robust method for statistically reliable analysis.



Propensity score

Because the use of REBOA or ACC was not randomly assigned, a formal causal inference is not possible. Therefore, a logistic regression analysis was used to estimate propensity scores (PSs) to predict usage of REBOA or ACC from the available predictors. These variables were age, gender, mechanism of injury, cause of injury, transport type, pre-hospital treatment, vital signs at ED, and ISS, which would reflect patient wound severity. Treatments after ED arrival (e.g., blood transfusion, cardiopulmonary resuscitation (CPR), operations) were not included in the PS derivation process because they were performed after usage of REBOA or ACC. Severity scoring systems without ISS (e.g., RTS, TRISS) were not included in the PS because in them were many of the same components that had already been included in the PS, such as vital signs. Propensity score matching extracted 1:1 matched pairs of subjects who received REBOA or ACC based on an averaged PS. The absolute standardized difference of variables for the PS estimation was used to assess the match balance. An absolute standardized difference of less than 0.2 was generally considered as an acceptable match balance between the groups.

The two-sided significance level for all tests was $P < 0.05$. All analyses were performed using SPSS software, version 21.0 (IBM, Armonk, NY, USA).

Results

A total of 159,157 trauma patients were registered in the JTDB from 1 January 2004 to 31 December 2013. Of these, 2032 patients with trauma were included in this study because they received REBOA or ACC. Cases were excluded if they received both REBOA and ACC ($n = 90$), were below the cutoff age ($n = 28$), had age data missing ($n = 11$), if they had already died ($n = 922$), had SBP data missing ($n = 58$), or if they did not have a chance to

survive (i.e., $AIS = 6$) ($n = 20$). Thus, 903 patients were included in the first round of calculations. After PS matching, 304 patients were included in a second round of analysis (Fig. 1).

Table 1 shows the characteristics of critical patients with trauma who received either REBOA or ACC. The mean age was 53.7 ± 21.2 years; 611/903 (67.7%) were male. Blunt trauma was common (838/895; 93.6%). Mean RTS was 4.94 ± 2.08 . The RTS in REBOA cases was significantly higher than in ACC cases. Median (interquartile) ISS was 34 (20). ISS in REBOA cases were also more severe than that in ACC cases. However, TRISS was higher in REBOA cases than in ACC cases. Also, 153/636 (24%) patients who were REBOA cases received TAE compared to 18/267 (6.7%) who were ACC cases. Table 2 demonstrates outcome comparisons between REBOA and ACC. In-hospital mortality was 405/607 (67%) in REBOA and 210 /233 (90%) in ACC. ED mortality was 137/625 (22%) in REBOA and 130/264 (49%) in ACC. Figure 2 shows a comparison of the mortality between REBOA and ACC ($n = 903$). Patients who underwent REBOA had a significantly lower in-hospital mortality than those who underwent ACC as shown by adjusted RTS (odds ratio (OR) = 0.224; 95% confidence interval (CI) = 0.129–0.700), ISS (OR = 0.188; 95% CI = 0.116–0.303), or TRISS (OR = 0.309; 95% CI = 0.190–0.502), respectively. After PS matching ($n = 304$), in-hospital mortality was 106/146 (73%) in REBOA and 122/134 (91%) in ACC, and ED mortality was 24/149 (16%) in REBOA and 77/150 (51%) in ACC. Thus, mortality in the REBOA patients was lower than that of ACC (OR = 0.261; 95% CI = 0.130–0.523 at discharge; OR = 0.182; 95% CI = 0.106–0.313 at ED). Table 3 lists the baseline characteristics of PS-matched patients ($n = 304$). There was no significant difference between REBOA and ACC in RTS (mean \pm SD; 4.8 ± 2.0 vs 4.7 ± 2.1 ; $P = 0.631$), ISS (median (interquartile);

Table 1 Characteristic of critical patients with trauma who had either REBOA or ACC

	REBOA (n = 636)	ACC (n = 267)	P value	Missing
Age (years)	52.5 ± 21.2	56.7 ± 21.1	0.007	0
Gender (male)	417/636 (66%)	194/267 (73%)	0.043	0
Onset year				0
2004–2008	218/636 (34%)	70/267 (26%)		
2009–2013	418/636 (66%)	197/267 (74%)		
Mechanism of injury (blunt vs. penetrating)	591/630 (94%)	247/265 (93%)	0.765	8
Cause of injury			0.754	31
Accident	429/618 (69%)	187/254 (74%)		
Suicide	127/618 (21%)	44/254 (17%)		
Assault	20/618 (32%)	6/254 (2.4%)		
Workplace injuries	39/618 (6.3%)	16/254 (6.3%)		
Other	1/618 (0.2%)	1/254 (0.4%)		
Transport type			0.008	27
Ambulance	514/617 (83%)	194/259(75%)		
Ambulance with physician	26/617 (4.2%)	24/259 (9.3%)		
Helicopter with physician	73/617 (12%)	40/259 (15%)		
Other	4/617 (0.6%)	1/259 (0.4%)		
Vital signs at prehospital				
SBP	101 (42)	105 (40)	0.42	358
HR	97 (37)	100 (43)	0.76	163
RR	24 (10)	25 (10)	0.445	232
Vital signs at emergency department				
GCS value	10(12)	5(8)	<0.001	21
SBP	89 (46)	87 (45)	<0.001	0
HR	102 (36)	106 (52)	0.181	14
RR	25 (10)	24 (15)	<0.001	99
RTS	5.2 ± 2.0	4.2 ± 2.2	<0.001	107
AIS				0
Head (n = 382)	3.6 ± 1.2	3.3 ± 1.1	0.101	
Face (n = 167)	1.6 ± 0.7	1.8 ± 1.3	0.274	
Neck (n = 14)	2.4 ± 1.3	1.5 ± 0.8	0.178	
Thorax (n = 593)	3.8 ± 0.9	4.3 ± 1.1	<0.001	
Abdomen and pelvis (n = 580)	3.6 ± 1.1	3.8 ± 1.5	0.143	
Spine (n = 187)	2.5 ± 1.1	2.6 ± 0.9	0.959	
Upper extremity (n = 209)	2.0 ± 0.6	2.1 ± 0.6	0.284	
Lower extremity (n = 558)	3.7 ± 1.3	3.7 ± 1.2	0.702	
Others (n = 39)	1.1 ± 0.4	1.2 ± 0.4	0.701	
ISS	34 (25)	34 (20)	<0.001	0
TRISS (probability of survival)	0.43 ± 0.36	0.27 ± 0.30	<0.001	12
Prehospital treatment				
Airway protection maneuver	53/636 (8.3%)	41/267 (15%)	0.003	0
Intubation	44/636 (6.9%)	35/267 (13%)	0.004	0
Intravenous fluid	55/636 (8.6%)	25/267 (9.3%)	0.703	0

Table 1 Characteristic of critical patients with trauma who had either REBOA or ACC (*Continued*)

FAST			0.013	29
Positive	359/614 (59%)	133/260(51%)		
Negative	233/614 (38%)	107/260 (41%)		
Not conducted	22/614 (3.6%)	20/260 (7.7%)		
Blood transfusion	542/636 (85%)	197/267 (74%)	<0.001	0
CPR				
ERT with CPR	71/636 (11%)	216/267 (81%)	<0.001	0
Closed CPR	141/636 (22%)	92/267 (35%)	<0.001	0
Operation at initial evaluation				
Craniotomy	19/636 (3.0%)	1/267 (0.4%)	0.012	0
Craterization	17/636 (2.7%)	3/267 (1.2%)	0.215	0
Thoracotomy	70/636 (11%)	160/267 (60%)	<0.001	0
Laparotomy	301/636 (47%)	99/267 (37%)	0.005	0
Angiography				
Chest	29/636 (4.6%)	7/267 (2.6%)	0.196	0
Abdomen	156/636 (25%)	15/267 (5.6%)	<0.001	0
Pelvis	151/636 (24%)	22/267 (8.2%)	<0.001	0
TAE (all)	153/636 (24%)	18/267 (6.7%)	<0.001	0

All categorical variables are shown as *n* (%); continuous variables are shown as mean \pm standard deviation or median (interquartile)

ACC aortic cross-clamping, AIS Abbreviated Injury Score, CPR cardiopulmonary resuscitation, ERT Emergency resuscitative thoracotomy, FAST Focused assessment with sonography for trauma, GCS Glasgow Coma Scale, HR heart rate, ISS Injury Severity Score, REBOA resuscitative endovascular balloon occlusion of the aorta, RR Respiratory rate, RTS revised trauma score, SBP systolic blood pressure, TAE trans-catheter arterial embolization, TRISS trauma and injury severity score

34 (23) vs 36 (20), $P = 0.341$), and TRISS (mean \pm SD; 0.45 ± 0.35 vs 0.39 ± 0.31 , $P = 0.115$). However, the AIS of the thorax was significantly lower in REBOA cases than in ACC cases (3.8 ± 0.8 vs 4.2 ± 0.8 , $P < 0.001$). Thoracotomy at initial evaluation was also less frequent in REBOA cases than in ACC cases. On the other hand, patients with REBOA underwent angiography of the abdomen and pelvis, including TAE, more often than those with ACC.

Table 2 Outcome comparisons between REBOA and ACC

	REBOA (<i>n</i> = 636)	ACC (<i>n</i> = 267)	<i>P</i> value
Disposition at discharge			<0.001*
Died (in-hospital mortality)	405/607 (67%)	210/233 (90%)	
Transferred	118/607 (19%)	11/233 (1.8%)	
Home	83/607 (14%)	12/233 (2.0%)	
Other	1/607 (0.1%)	0/233 (0.0%)	
Disposition at ED			<0.001*
Died (ED mortality)	137/625 (22%)	130/264 (49%)	
CU admission	472/625 (76%)	129/264 (49%)	
Ward admission	137/625 (22%)	4/264 (1.5%)	
Other	5/625 (1.8%)	1/264 (0.4%)	

The variables are shown as *n* (%)

ACC aortic cross clamping, ED emergency department, ICU intensive care unit, REBOA resuscitative endovascular balloon occlusion of the aorta

*Chi-square test

Discussion

Brief summary

This study investigated the current usage of REBOA and ACC using a large, nationwide trauma database in Japan. Mortality rates in patients requiring AO was discovered to be very high but this is attributed to the usage of ACC on patients who cannot be saved, skewing mortality out of favor with ACC. We also analyzed outcomes for patients after receiving either REBOA or ACC after adjusting for patient trauma severity. Robust analyses of the adjusted data showed that REBOA was associated with significantly reduced in-hospital mortality compared with ACC. However, due to differences in associated procedures between REBOA (e.g., increased need for angiography) and ACC (e.g., thoracotomy), there should be some consideration given to choosing either intervention.

Comparison with previous studies

To our knowledge, our current study is one of the largest cohort studies describing the use of REBOA [8]. REBOA has recently found use as a general technique across major emergency centers in Japan. The highest density of potential REBOA patients is also seen at major trauma centers in England and Wales, although the number of patients in whom REBOA was utilized is small [9]. In fact, a review of the potential use of REBOA in exsanguinating hemorrhage cases in the US suggested that this new technique should

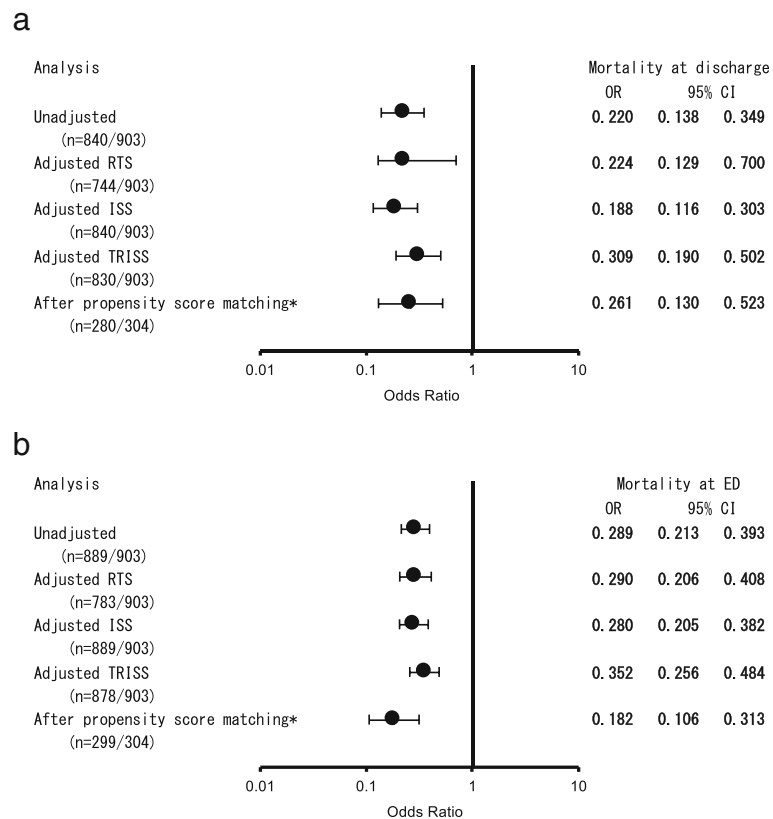


Fig. 2 Comparison of the mortality of REBOA versus ACC at discharge (a) and in the emergency department (ED) (b). *The covariates used to estimate the propensity score were age, gender, mechanism of injury, cause of injury, transport type, prehospital treatment, vital signs at ED, and Injury Severity Score (ISS). CI confidence interval, OR odds ratio, RTS revised trauma score, TRISS trauma and injury severity score

be thoroughly evaluated for broad use, but the literature currently suffers from a dearth of human studies on REBOA [10]. Although our observational study admittedly had some selection bias, we feel that our results will nonetheless become an important part of the foundation of literature supporting the evaluation of global REBOA use.

Previous studies regarding REBOA usage have been limited in size and scope, and have shown ambiguous results. For example, although previous single-center cohort studies mentioned the utility of REBOA for massive pelvic bleeding cases that could still be imaged by angiography [5, 6], another large, retrospective cohort study cautioned against REBOA usage for patients who had emergency surgery or transcatheter embolization [7, 11]. Yet another single-center cohort study also reported on the feasibility and safety of REBOA for a non-compressive torso injury (pelvic fracture or hemoperitoneum) [12], but contrasting studies also reported that REBOA usage was associated with a higher mortality compared with non-REBOA usage in JTDB [7, 11]. However, to objectively evaluate these reports, a thorough knowledge of the Japanese trauma care system is required. For example, most Japanese emergency departments see few in-house

trauma surgeries, see fewer trauma cases overall, and mostly deal with older patients and age-related maladies [13]. REBOA usage, in this context, may signal “last ditch” efforts [11]. However, our results from the same database show an incongruent outcome even though our study population and comparisons are different. Previous reports have indicated REBOA usage as a last resort in the most severe trauma cases, but only one multicenter, prospective observational study (Aortic Occlusion for Resuscitation in Trauma and Acute Care Surgery (AORTA) registry) [1] has looked at direct comparisons with ACC, which is also used in the most severe cases, and found REBOA to be beneficial. As the severity criteria for both REBOA and ACC are similar, it is reasonable to compare those outcomes directly and our reports findings strengthen the conclusion of DuBose and colleagues [1], and show a promising consistency in results.

Possible explanations and implications

AO was used on 2032 patients in our database. However, 799 patients with ACC were excluded from analysis because of pre-hospital cardiopulmonary arrest. Although we controlled for this in our study, the differences between REBOA and ACC in general need more context

Table 3 Baseline characteristic in propensity score-matched patients with severe trauma*

	REBOA (n = 152)	ACC (n = 152)	SD	P value	Missing
Age (years)	52.8 ± 21.0	54.8 ± 22.1	0.09	0.421	0
Gender (male)	111/152 (73%)	101/152(66%)	0.14	0.261	0
Mechanism of injury (blunt vs. penetrating)	142/152 (93%)	141/152(93%)	0.03	1.000	0
Cause of injury				0.726	0
Accident	104 (68%)	109 (72%)	0.07		
Suicide	28 (18%)	29 (19%)	0.02		
Assault	6 (3.9%)	5 (3.3%)	0.04		
Workplace injuries	14(92%)	9 (5.9%)	0.13		
Other	0(0%)	0 (0%)	0.00		
Transport type				0.542	0
Ambulance	124/152 (82%)	123/152(81%)	0.02		
Ambulance with physician	6/152 (3.9%)	10/152 (6.6%)	0.12		
Helicopter with physician	22/152 (15%)	19/152 (13%)	0.06		
Other	0/152 (0%)	0/152(0%)	0.00		
Vital signs at emergency department					
GCS value	8(10)	8(10)	0.02	0.909	0
SBP	77.5 (64)	73.5 (64)	0.02	0.421	0
HR	108.0 (39)	109.5(52)	0.05	0.687	0
RR	25 (10)	24 (14)	0.08	0.499	0
RTS	4.8 ± 2.0	4.7 ± 2.1		0.631	0
ISS	34 (23)	36 (20)	0.11	0.341	0
TRISS (probability of survival)	0.45 ± 0.35	0.39 ± 0.31		0.115	0
Prehospital treatment					
Airway protection maneuver	12/152 (7.9%)	11/152 (7.2%)	0.03	1.000	0
Intubation	9/152 (5.9%)	13/152 (8.6%)	0.10	0.508	0
Intravenous fluid	9/152 (5.9%)	14/152 (9.2%)	0.13	0.386	0
FAST				0.232	6
Positive	91/148 (62%)	78/150(52%)			
Negative	52/148 (35%)	64/150(43%)			
Not conducted	5/148 (3.4%)	8/150 (5.3%)			
Blood transfusion	135/152 (89%)	121/152(80%)		0.04	0
AIS					0
Head	3.8 ± 1.3	3.4 ± 1.1		0.047	
Face	1.7 ± 0.8	1.3 ± 0.5		0.036	
Neck	1.3 ± 0.6	1.3 ± 0.5		0.846	
Thorax	3.8 ± 0.8	4.2 ± 0.8		<0.001	
Abdomen and pelvis	3.5 ± 1.0	3.8 ± 1.4		0.112	
Spine	2.6 ± 0.9	2.6 ± 1.0		0.812	
Upper extremity	2.1 ± 0.7	2.1 ± 0.6		0.833	
Lower extremity	3.6 ± 1.4	3.6 ± 1.3		0.904	
Other	1.0 ± 0.0	1.3 ± 0.5		0.172	

Table 3 Baseline characteristic in propensity score-matched patients with severe trauma* (Continued)

Operation at initial evaluation				
Craniotomy	3/152 (2.0%)	0/152(0%)	0.248	0
Craterization	4/152 (2.6%)	1/152 (0.7%)	0.371	0
Thoracotomy	20/152 (13%)	92/152(61%)	<0.001	0
Laparotomy	79/152 (52%)	68/152(45%)	0.251	0
Angiography				
Chest	2/152 (1.3%)	5/152 (3.3%)	0.448	0
Abdomen	27/152 (18%)	10/152 (6.6%)	0.004	0
Pelvis	33/152 (22%)	15/152 (9.9%)	0.007	0
TAE (all)	29/152 (19%)	11/152 (7.2%)	0.004	0
CPR				
ERT with CPR	20/152 (13%)	125/152 (82%)	<0.001	0
Closed CPR	33/152 (22%)	53/152(35%)	0.015	0

All categorical variables are shown as *n* (%); continuous variables are shown as mean \pm standard deviation or median (interquartile)

ACC aortic cross-clamping, AIS Abbreviated Injury Score, CPR cardiopulmonary resuscitation, ERT Emergency resuscitative thoracotomy, FAST Focused assessment with sonography for trauma, GCS Glasgow Coma Scale, HR heart rate, ISS Injury Severity Score, REBOA resuscitative endovascular balloon occlusion of the aorta, RR Respiratory rate, RTS revised trauma score, SBP systolic blood pressure, SD standardized difference, TAE trans-catheter arterial embolization, TRISS trauma and injury severity score

*The covariates used to estimate the propensity score were age, gender, mechanism of injury, cause of injury, transport type, prehospital treatment, vital signs at the emergency department, and ISS

for accurate interpretation. In Japan, ACC currently seems to be a preferred intervention tactic in non-survivable injury cases and this differs from other countries, possibly making Japanese ACC-related mortality rates non-indicative of actual outcomes [1, 9, 13]. In addition, other patient characteristics such as better Glasgow Coma Scale (GCS) scores in cases where REBOA was used versus ACC cases (with more severe GCS scores) may also skew results. This raises the issue of snap decisions by ED physicians to choose rapid thoracotomy over REBOA because ACC would be more frequently chosen in cases with worse GCS scores. A key point to keep in mind, however is that although the probability of survival (TRISS) for REBOA was better than that of ACC, it is still no guarantee of success in severe cases (0.43 ± 0.36). Furthermore, REBOA patients who die might count as preventable, but ACC deaths with TRISS scores indicating unsurvivable injuries (0.27 ± 0.30) would be counted as non-preventable. This might not be seen as a negative even though the mortality of ACC patients was very high (90%) and might be related to more severe complications in the thorax. These issues highlight the nuances necessary to objectively interpret the data, as both REBOA and ACC have complicating factors. Survival rates at ED of 78% and 51% in REBOA and ACC, respectively, indicate that both can serve a role in trauma treatment. However, it is important to keep in mind that these procedures are not panaceas; only 14% (83/607) of REBOA patients and 2.0% (12/233) of ACC patients could leave the hospital and go home. This emphasizes the difficulty of AO in clinical practice. This is especially evident in Japan as our mortality was slightly higher than other countries [1, 9, 13]. As Japan's

prevalence of penetrating trauma is quite low (6.4%), survival probabilities may follow suit. However, taking into consideration the variability in study populations and institutional skill, a general trend in the same direction can be seen with our results versus those of other countries.

Trauma severity in ACC cases versus REBOA cases were controlled for with sensitivity analyses as seen in Fig. 2, but REBOA usage showed a clear survival benefit. PS matching was also used to control for insufficiency of adjustment and the tendency of results was the same among PS-matched patients. We found that PS matching was one of the best methods to control confounders in this prevalence and mortality. Again, direct comparisons between REBOA and ACC were conducted after PS matching because of current interest in the possibility of shifting the ACC paradigm to REBOA [10]. Table 3 shows the precision of our PS matching methodology. However, covariate differences where we did not use PS (although we did properly control for physiological severity and backgrounds) unavoidably resulted in an inability to match anatomical severity. This might be classified as an indication bias, but we feel that our analysis shows the real utility of AO.

Taken together, we feel that these results should be made part of the body of knowledge that physicians consult in the decision tree of AO. Accordingly, REBOA would conceivably be used more often as a solo abdominal trauma option even though there are no formal criteria for AO treatment utility. Still, this does not necessarily mean that all AO cases would shift from ACC to REBOA. In reality, choosing ACC for severe abdominal trauma patients who present no chest trauma is a difficult choice for ED physicians who may not have enough thoracotomy experience. This is especially

important because of recent reports on poor outcomes of emergency thoracotomies after abdominal exsanguination, adding to the reputation of ACC as a “last ditch” effort [13, 14]. However, to find the best position of an occlusion balloon with a blind approach is next to impossible when patients present with thoracic complications. This explains the higher incidence of abdominal and pelvic angiography in REBOA groups which we find to be an acceptable >trade-off for accuracy in occlusion balloon positioning. We do not doubt that REBOA will be applicable as a bridge to definitive treatment in the ED, but indications and contraindications in the light of ACC must be further refined. Finally, the most critical point to remember is that any method inducing long-lasting ischemia to at least half or more of the body has serious potential to harm the patient. To this end, the decision to use REBOA or ACC should be part of a robust clinical governance framework in order to ensure high quality patient care and maximal survival chance [9].

Limitations

Potential limitations of this study should be acknowledged. First, there remained some indication bias as previously discussed, indicating caution when interpreting results for clinical standpoints. However, we controlled for patient background using logistic regression and PS-matched analysis, when possible, and found two key points in this study. First, PS-matched analysis was one of the best methods for comparison because there was a relatively small sample size of survivors. Second, there was institutional bias although covariates were carefully selected on the basis of the assumption that none were affected directly by the intervention. This assumption could be a potential weakness and requires further study. With regard to mortality rates, a population-based study in England and Wales showed only major trauma centers had a high density of REBOA use and their rate was smaller than ours [9]. We, on the other hand, did not have institutional-level data, and therefore we could not control for it and this might account for our higher mortality rate. Although a potential weakness could be variability between physicians and institutions, AORTA registry data reveals that the general tendency of outcomes is the same [1]. Although selection bias may skew towards REBOA more than ACC in both AORTA and this study, we feel that our results are worth consideration to add to the scarce body of knowledge regarding this topic. Moreover, we did not have detailed data on REBOA or ACC such as the clamping time, the ballooning time, and the tactics of that therapy. Since the patients had the issue of ischemia/reperfusion injury, their outcome may have been influenced by time. A general assumption, however, is that clamping and ballooning times were kept as short as possible by the physicians because of the common knowledge that occlusion times should be kept to a minimum.

Conclusions

Despite any residual indication bias, REBOA might be a favorable alternative method to ACC, especially for severe trauma below the diaphragm. Further study is needed to elucidate optimal indications.

Key messages

- Patients who underwent AO had a high mortality.
- REBOA might be a favorable alternative method to ACC.

Abbreviations

ACC: Aortic cross-clamping; AIS: Abbreviated Injury Scale; AO: Aortic occlusion; AORTA: Aortic Occlusion for Resuscitation in Trauma and Acute Care Surgery; CI: Confidence interval; ED: Emergency department; GCS: Glasgow Coma Scale; IQR: Interquartile range; ISS: Injury Severity Score; JTDB: Japan Trauma Data Bank; OR: Odds ratio; PS: Propensity score; REBOA: Resuscitative endovascular balloon occlusion of the aorta; RTS: Revised trauma score; SD: Standard deviation; TAE: Trans-catheter arterial embolization; TRISS: Trauma and injury severity score

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Availability of data and materials

Not applicable

Authors' contributions

TA conceived of and designed this study, conducted data cleaning, analyzed the data, interpreted the data, drafted the manuscript, and revised the manuscript for important intellectual content. MU jointly conceived of and designed this study and interpreted the data. IN jointly conceived of and designed this study and interpreted the data. DS contributed to the acquisition of data, interpreted the data, and revised the manuscript for important intellectual content. NT interpreted the data and revised the manuscript for important intellectual content. All of the authors reviewed, discussed, and approved the final manuscript.

Competing interests

TA received a research grant from Hitachi, Ltd. previously, but declares no competing interests in relation to this research. The other authors declare that they have no competing interests.

Consent for publication

Not applicable

Ethics approval and consent to participate

We received permission to use the data from the JTDB. This study was approved by the ethics committee of Tsukuba Medical Center Hospital. The ethics committee at our institution does not require its informed consent for observational studies using anonymous data previously collected for routine operations. Also, informed consent from each patient is waived for using anonymous data according to the informed consent guidelines in Japan.

Presentation

We presented these results at the AHA Scientific Sessions Resuscitation Science Symposium on 12 November 2016. Our research was selected Best of the Best Oral Abstract Presentations and Presentation of the Best Abstract Awards for Cardiac and Trauma Resuscitation Science.

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ERRATUM

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Erratum to: Resuscitative endovascular balloon occlusion of the aorta versus aortic cross clamping among patients with critical trauma: a nationwide cohort study in Japan

Toshikazu Abe^{1,2*}, Masatoshi Uchida¹, Isao Nagata¹, Daizoh Saitoh³ and Nanako Tamiya¹

Erratum

Following publication of the original article [1], it was brought to our attention that there were a few errors in Table 2:

CU admission should read: ICU admission

11/233 (1.8%) should read: 11/233 (4.7%)

12/233 (2.0%) should read: 12/233 (5.2%)

The corrected table is presented in this erratum [Table 2].

Furthermore, the sentence "...only 14% (83/607) of REBOA patients and 2.0% (12/233) of ACC patients could leave the hospital and go home." in the Discussion section should as a consequence read: "...only 14% (83/607) of REBOA patients and 5.2% (12/233) of ACC patients could leave the hospital and go home."

This has now been corrected in this erratum.

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Table 2 Outcome comparisons between REBOA and ACC

	REBOA (n = 636)	ACC (n = 267)	P value
Disposition at discharge			<0.001*
Died (in-hospital mortality)	405/607 (67%)	210/233 (90%)	
Transferred	118/607 (19%)	11/233 (4.7%)	
Home	83/607 (14%)	12/233 (5.2%)	
Other	1/607 (0.1%)	0/233 (0.0%)	
Disposition at ED			<0.001*
Died (ED mortality)	137/625 (22%)	130/264 (49%)	
ICU admission	472/625 (76%)	129/264 (49%)	
Ward admission	137/625 (22%)	4/264 (1.5%)	
Other	5/625 (1.8%)	1/264 (0.4%)	

The variables are shown as n (%)

ACC aortic cross clamping, ED emergency department, ICU intensive care unit,

REBOA resuscitative endovascular balloon occlusion of the aorta

*Chi-square test



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Article

The relationship between raising a child with a disability and the mental health of mothers compared to raising a child without disability in Japan



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ABSTRACT

Objective: Previous studies conducted in Japan targeted only mothers who cared for children with disabilities, and lacked reference subjects, such as mothers of children without disabilities. The aim of this study was to examine the association between raising one or two children with a disability and maternal psychological distress compared to mothers of children without a disability, and to assess differences among partnered mothers living with grandparent(s), partnered mothers without grandparent(s), and single mothers.

Methods: This study utilized data from the Comprehensive Survey of Living Conditions (CSLC) in 2010. We merged the data of the children (aged six and over), mothers, and fathers. This study obtained 33,739 study subjects as a triad of a child (33,110 children without disabilities and 629 children with disabilities), mother, and father. The Japanese version of Kessler 6 (K6) was used to assess the psychological distress of mothers. Multivariate logistic regression was performed to assess the independent association of a child with a disability on maternal psychological distress after controlling for the basic characteristics of the children, mothers, and households.

Results: This study reported that raising one or two children with disabilities was significantly related to maternal psychological distress (odds ratio: 1.72 for one child, 2.85 for two children) compared to mothers of children without disability. After stratifying the analyses by family structure, significant associations remained among mothers in two-parent families but not for mothers in three-generation families and single mothers due to a small number of children with disabilities in these families.

Conclusions: This study reported the significant association between raising a child with a disability and maternal psychological distress in comparison to mothers of children without disabilities. Attention should be paid to not only single mothers, but also partnered mothers in two-parent families who have a child with a disability. It is important for health professionals to focus on the mental health of every mother of a child with a disability and to assess their needs for psychological support.

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1. Background

Raising a child with a disability is an unexpected experience for parents (Raina et al., 2005). Caring for a child with a disability, depending on the type of disability, can result in physical health problems, mental health problems, and time or financial burdens on mothers (Bourke et al., 2008; Brehaut et al., 2004; Estes et al.,

2013; Lee, 2013; Montes & Halterman, 2008a, 2008b; Nes et al., 2014; Parish, Rose, Dababnah, Yoo & Cassiman, 2012; Raina et al., 2005). In particular, maternal mental health is important for both mothers and children because poor maternal mental health is related to unfavorable parenting practices (McLennan & Kotelchuck, 2000; Minkovitz et al., 2005), child health problems (Ferro & Speechley, 2009; Schwebel & Brezausek, 2008), and poor school performance (Shen et al., 2016).

Mothers play a major role in childrearing, regardless of the presence or absence of a disability. Supporting mothers is a public issue because Japanese society faces a proliferation of nuclear and

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dual-income families. Paternal involvement in childrearing remains to a small extent in the present circumstances. For example, the parental leave obtainment rate was only 2.0% of full-time working fathers (The Ministry of Health Labour and Welfare, 2012), and fathers seldom commit to household chores and childrearing tasks (i.e., only 39 minutes per week in dual-income families) (Statistics Bureau, 2011). Therefore, mothers may experience further negative influence on their mental health in addition to ordinary childrearing tasks.

Previous studies conducted in Japan targeted only mothers who cared for children with disabilities and lacked reference subjects, such as mothers of children without disabilities (Toki et al., 2010; Yamaguchi, Takataya & Ogiwara, 2005; Yamaoka et al., 2015; Yatsugi, Suzukamo & Izumi, 2013). Furthermore, these studies did not consider the possibility of having multiple children with disabilities in a household. Comparison with general childrearing situation is needed to understand the magnitude of mental problems related to raising one or more children with disabilities and to provide specific supporting measurements in addition to general parenting support. Therefore, this study aimed to assess the relationship between raising single or multiple children with disabilities and the mental health of mothers compared mothers of children without disabilities using population-based data in Japan.

2. Method

2.1. Data source

Data for this study, were drawn from the Comprehensive Survey of Living Conditions (CSLC) (The Ministry of Health, 2011), which was conducted by the Ministry of Health, Labour and Welfare in 2010. The CSLC is a nationally representative survey of households that has been conducted every three years since 1986, with smaller annual surveys in the intervals. The CSLC employs a stratified random cluster sampling method based on the Census. The Census in 2005 surveyed 982,000 enumeration districts (EDs) throughout Japan, of which each ED included approximately 50 households. The 2010 CSLC randomly selected 5530 EDs, and all household members in the selected EDs became study subjects, except for individuals who were hospitalized or institutionalized for long periods. The enumerator collected the self-administered questionnaire by visiting all households. The 2010 CSLC surveyed 289,363 households and collected questionnaires from 229,785 households (response rate: 79.4%), which comprised 609,019 household members.

2.2. Study sample

The flow chart (Fig. 1) shows how samples were extracted to determine the study subjects. First, we extracted children who were under the age of 18 from the total CSLC sample ($n=102,668$). Second, the CSLC coded family structures as two-parent families, three-generation families, families with single mothers, and others. Other types of family structures or households without family structure data were excluded ($n=6018$) because it was impossible to determine the parent-child relationships in families with several adult couples, single adults, and children living in the same household. There were 96,650 children with three types of family structures. Third, we excluded single-father families ($n=1261$) because the outcome variable of the study was maternal psychological distress. In addition, this study aimed to compare the households of children with disabilities and those without disabilities. We used the question that inquired about disability status for children aged six and over. The content of this question is explained in the subsection “explanatory variable”. Because of the targeting age in this question, children under the age of six were excluded ($n=27,222$). Before merging each set of data for the

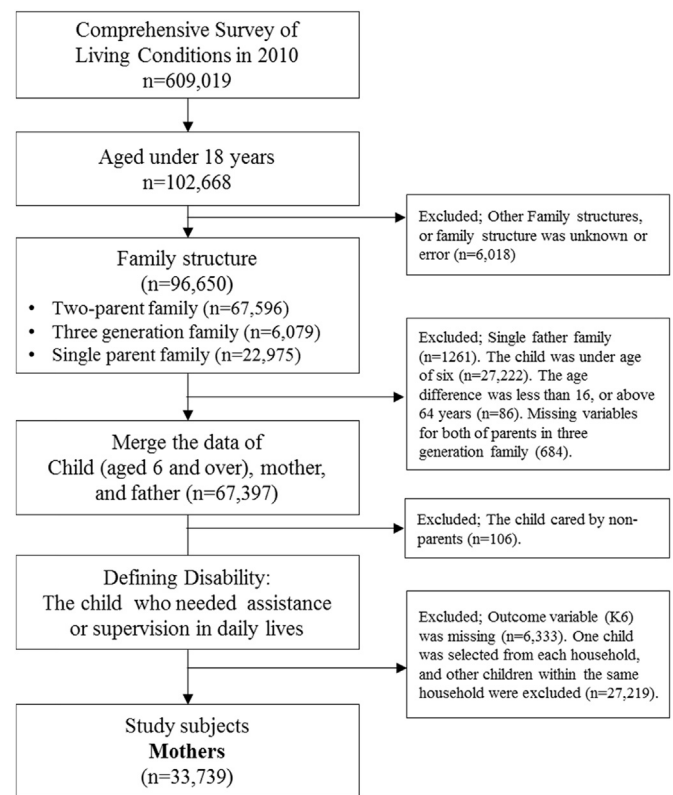


Fig. 1. Flow chart of study samples.

children, mothers, and fathers, we excluded the outliers of age differences—over the age of 64 or under the age of 16 ($n=86$)—and children without age data for both the mothers and fathers in three-generation families ($n=684$). In this manner, we merged the data of the children, mothers, and fathers ($n=67,379$). This study targeted the psychological distress level of mothers who were caring for children with or without disabilities. Therefore, we excluded children with disabilities who were not being cared for by parents ($n=106$) and mothers without outcome variables ($n=6333$). Last, we selected one child per household to eliminate the possibility of repeated measures for the same mothers and fathers, and excluded other children from the same household ($n=27,219$). In households where none of the children had disabilities, the youngest child was selected. In households where all the children had disabilities, the child with the most severe disability was selected. In households where some children did have disabilities and other children did not have disabilities, the child who was the youngest and had a more severe disability was selected. The severity of a child's disability is explained in the subsection “explanatory variable”. Finally, we obtained 33,739 mothers as a study subjects and triads of a child, mother, and father, including 33,110 children without disabilities and 629 children with disabilities.

2.3. Mental health measures

Maternal mental health was assessed by the Japanese version of the Kessler 6 (K6) scale (Furukawa et al., 2008). The K6 score, a simple screening tool with six items, ranges from 0 to 24 with a 5-point scale (0–4) for each item, and a higher total score indicates more severe mental health conditions. Non-specific psychological distress is the core dimension that people with a wide range of mental disorders have typically experienced (Kessler et al., 2002). Kessler et al. originally developed the K6 to identify non-specific psychological distress among the general population (Kessler et al.,

2002; Kessler et al., 2010). Sakurai et al. reported that the optimal cutoff point on K6 was 4/5 for screening mood and anxiety disorders in self-reported questionnaires (sensitivity 100%, specificity 68.7%) using community residents in Japan (Sakurai, Nishi, Kondo, Yanagida & Kawakami, 2011). The present study employed a cutoff point of 4 or 5 and higher in the K6 score to assess non-specific psychological distress because CSLC used a self-administered questionnaire. A K6 score of 5 and higher was coded '1' to express the presence of psychological distress and '0' otherwise.

2.4. Explanatory variables

The child's disability status was an explanatory variable in the current study. To define whether a child had a disability, we used responses to the question that inquired about disability status for children aged six and over: "Does he/she need assistance or supervision?" This question targeted children aged six and over to avoid general parenting assistance or supervision toward infants and preschoolers. Children aged six and over who received a response of "Yes" to this question were operationally categorized as a "child with disability". The number of children with a disability in the same household was used to assess the extent of association between raising them and maternal psychological distress.

In terms of the severity of disabilities, we utilized a subsequent question from the questionnaire to ask about four levels of the child's dependence in their daily life. The mildest level (Level 1) indicated that a child did have any type of disability but could go out by oneself. Level 2 indicated a child who was independent at home but needed assistance to go out. Level 3 represented a child that needed assistance in their daily life, mainly stayed in bed, and maintained a sitting position. The severest level (Level 4) indicated a child who was bedridden and needed help for all of their daily activities, such as egestion, meals, and changing clothes. The severity of disabilities was used to select one child if there were multiple children in the same household.

2.5. Covariates

Possible covariates were used from the questionnaire, such as the characteristics of a child (sex, age, outpatient visits, activity limitation), parents (age, educational attainment, and working status), and household (family structure, house ownership, number of children, population of city, and monthly household expenditure). The question for the outpatient visits was the following: "Do you currently make regular outpatient visits to hospitals, clinics, Japanese traditional massage therapists, acupuncturists, moxocautery practitioners, Judo orthopedists, or regular home visits by a physician?" If the child visits regularly to any kind of health care facility, the outpatient visits was coded as '1', or otherwise '0'. The question for activity limitation was the following: "Have you ever become bedridden or experienced the inability to perform usual activities due to health problems, such as being absent from work or school or not being able to perform house-keeping tasks at least one day during last month?" If the child had answered positively to these conditions at least one day in the last month, the activity limitation was coded as '1' and '0' otherwise. The educational attainment of parents was divided into two categories: high educational attainment indicated graduation from vocational school, technical college, university, or postgraduate school, and low educational attainment indicated graduation from primary school, junior high school, or high school. The working status of parents was coded as '1' for having any type of job and '0' for jobless. The status of having a job included a person who worked and attended school at the same time or worked for a self-owned business. Jobless status indicated a person who attended school or performed household chores without engaging in a job.

Family structure was categorized into four groups: family with two parents and child's grandparent(s), family with two parents, family with one parent and child's grandparent(s), and family with one parent (i.e., single mother family). The population of the city was also considered as one of the covariates because living in an urban city might influence maternal psychological distress, and it was divided into two categories with a threshold of 150,000. The total household monthly expenditure was categorized into either below the 25th percentile or the 25th percentile and above after dividing it by the square root of the number of family members to standardize the effect of family size. Below the 25th percentile of total household monthly expenditure was coded as '1' for lower household expenditure and '0' otherwise.

2.6. Statistical analysis

We firstly conducted univariate analysis to describe the associations between covariates and maternal psychological distress. We used Student's *t*-test for the continuous variables (age), the χ^2 test for dichotomous variables (sex, outpatient visits, activity limitation, maternal and paternal educational attainment, working status, family structure, homeownership, lower household expenditure, population of the city), and the Wilcoxon rank sum test for nonparametric variables (number of children without disability in the household).

Secondly, we conducted a multivariable logistic regression to assess the association between the presence and number of children with a disability and maternal psychological distress after controlling for the basic characteristics of the children (age, sex, number of children with a disability). Child's outpatient visits and activity limitation were not included into the multivariable logistic regression because it might mediate the dependent variable. In the next step, we entered confounding factors (maternal educational attainment and working status, house ownership, monthly expenditure, and family structure) into the model. Among family structure, partnered mothers living with the child's grandparent (s) was defined as the reference group. Because the coefficients of correlation between the age of the child and mother, child and father, and mother and father were relatively high (0.66, 0.61, and 0.82), we added only the age of the child as a confounder into the model. Paternal variables were not added to the model because there were single mothers.

Lastly, we stratified into three subgroups: partnered mothers living with grandparent(s), partnered mothers without grandparent(s), and single mothers both of with and without grandparent(s). Multivariable logistic regression was performed among partnered mothers with the confounding factors of the child, mother, household, and also father (i.e., educational attainment and working status). In the multivariable logistic regression for single mothers, we added the variable of co-residence with the child's grandparent(s) instead of paternal variables. The analyses were performed with complete cases for all variables in the models. We used STATA/MP, version 14 (Stata Corp LP, College Station, TX, USA) for all analyses.

3. Results

Table 1 shows the characteristics of the children, households, mothers, and fathers and their associations with maternal psychological distress. One-third of mothers had experienced psychological distress during the last month ($n=11,729$, 34.7%). Of the 33,739 subjects, 1.8% of mothers ($n=594$) had one child with a disability, and 0.10% of mothers ($n=35$) had two children with a disability in their household. The majority of mothers were partnered mothers in two-parent families living without the child's

Table 1Association between maternal psychological distress (K6 score \geq 5) and characteristics of child, mother, father, and household.

			Total (n=33,739)		K6 score < 5 (n=22,010)		K6 score \geq 5 (n=11,729)		p value for Chi-square test
			n	(%)	n	(%)	n	(%)	
Child (n=33,739)	Age (range: 6–17)	(mean, SD)	33,739	(10.5, 3.5)	22,010	(10.4, 3.5)	11,729	(10.6, 3.5)	< 0.001 [†]
	Sex	Male	17,223	51.0%	11,208	50.9%	6015	51.3%	0.528
		Female	16,516	49.0%	10,802	49.1%	5714	48.7%	
	Outpatient visits	No	26,091	80.7%	17,361	82.2%	8730	78.0%	< 0.001
		Yes	6227	19.3%	3758	17.8%	2469	22.1%	
	Activity limitation	No	27,826	88.8%	18,416	89.9%	9410	86.8%	< 0.001
		Yes	3510	11.2%	2078	10.1%	1432	13.2%	
Disability	No	33,110	98.1%	21,691	98.6%	11,419	97.4%	< 0.001	
	Yes	629	1.9%	319	1.4%	310	2.6%		
Mother (n=33,739)	Age (range:23–80)	(mean, SD)	33,739	(41.0, 5.8)	22,010	(41.0, 5.8)	11,729	(41.1, 5.9)	0.559 [†]
	Education attainment ^a	High	15,601	50.3%	10,446	51.5%	5155	47.9%	< 0.001
		Low	15,433	49.7%	9820	48.5%	5613	52.1%	
	Working status	Yes	23,613	70.1%	15,446	70.3%	8167	69.7%	0.319
No		10,085	29.9%	6540	29.7%	3545	30.3%		
Father (n=30,179)	Age (range: 23–71)	(mean, SD)	30,179	(43.5, 6.7)	20,054	(43.4, 6.7)	10,125	(43.7, 6.8)	< 0.001 [†]
	Education attainment ^a	High	13,817	49.4%	9407	50.6%	4410	47.0%	< 0.001
		Low	14,167	50.6%	9191	49.4%	4976	53.0%	
	Working status	Yes	29,355	98.1%	19,584	98.5%	9771	97.4%	< 0.001
No		570	1.9%	305	1.5%	265	2.6%		
Household (n=33,739)	Two parents with grandparents		7145	21.2%	4744	21.6%	2401	20.5%	< 0.001
	Two parents without grandparents		23,034	68.3%	15,310	69.6%	7724	65.9%	
	Single parent with grandparents		1119	3.3%	687	3.1%	432	3.7%	
	Single parent without grandparents		2441	7.2%	1269	5.8%	1172	10.0%	
	House ownership	No	8529	25.3%	5199	23.6%	3330	28.4%	< 0.001
		Yes	25,210	74.7%	16,811	76.4%	8399	71.6%	
	Number of children (median, IQR) (range)		33,739	(2, 1–2) (1–13)	22,010	(2, 1–2) (1–13)	11,729	(2, 1–2) (1–10)	0.012 [‡]
		Number of children with disability	None	33,110	98.1%	21,691	98.6%	11,419	97.4%
		One	594	1.8%	305	1.4%	289	2.5%	
		Two	35	0.10%	14	0.06%	21	0.18%	
Population of city	< 150,000		16,756	49.7%	10,882	49.4%	5874	50.1%	0.263
	\geq 150,000		16,983	50.3%	11,128	50.6%	5855	49.9%	
Monthly expenditure ^b	\geq 25%tile		24,580	76.2%	16,079	76.3%	8501	75.9%	0.446 [†]
	< 25%tile		7687	23.8%	4992	23.7%	2695	24.1%	

 χ^2 test.

t-test.

[‡] Wilcoxon rank sum test.^a High education attainment is for person graduated from university, college, or vocational school. Low educational attainment is for person graduated from junior high school or high school.^b Monthly expenditure was divided by the square root of number of persons in the household. Missing value: Outpatient visit (n=1421), Activity limitation (n=2403), Maternal educational attainment (n=2705), Maternal working status (n=41), Paternal educational attainment (n=2195), Paternal working status (n=254), and Monthly Expenditure (n=1472).

grandparent(s) (68.3%). The proportion of single mothers was 10.5%, including co-residence with grandparent(s) (3.3%) and without co-residence (7.2%). The proportion of mothers having a child with a disability was not significantly different among partnered mothers (1.87%) and single mothers (1.93%). However, the proportion of co-residence with grandparent(s) was significantly lower among families with one or two disabled children (16.2%) compared to families without (24.7%) (p < 0.001, these results are not presented in Table 1).

Table 2 shows the multivariable logistic regression analysis for the psychological distress of mothers. Raising a child with a disability was significantly associated with maternal psychological distress (crude odds ratio (OR) 1.78, 95% confidence interval (95% CI) 1.51–2.09 for one child with a disability; OR 2.90, 95% CI 1.47–5.70 for two children with a disability). After controlling for the characteristics of the mother and household, the adjusted OR (AOR) was still significantly associated (AOR 1.72, 95% CI 1.44–2.05 for one child with a disability; AOR 2.85, 95% CI 1.28–6.34 for two children with a disability).

Table 3 shows the stratified analyses among the three sub-groups of mothers. Among partnered mothers living with grandparent(s), the proportions of having one child or two children with disability were 87 (1.4%) and 3 (0.048%). Raising a child with a disability was not significantly associated with psychological distress (AOR, 95% CI: 1.34, 0.84–2.13). The odds ratio for having two children with a disability could not be obtained due to the small sample size of the explanatory variable. Among partnered mothers living without grandparent(s), 446 mothers had one child (2.2%) and 24 mothers had two children with a disability (0.12%). The AOR remained 1.80, with significance (95% CI: 1.47–2.21), for one child, and 2.84 for two children with a disability (95% CI: 1.14–7.07). Among single mothers, 61 mothers had one child with a disability (2.0%), and eight mothers had two children with a disability (0.26%). Raising one or two children with a disability was not significantly associated with psychological distress (AOR, 95% CI: 1.42, 0.81–2.47 for one child; 4.43, 0.51–38.7 for two children with a disability).

Table 2
The relationship between having a child with a disability and maternal psychological distress.

		All mothers n = 33,769		All mothers n = 29,868	
		OR	95%CI	OR	95%CI
Child with disability	Age	1.02	1.01–1.03	1.02	1.01–1.03
	Male	1.01	0.96–1.06	1.01	0.97–1.06
	One child with disability at home	1.78	1.51–2.09	1.72	1.44–2.05
	Two children with disability at home	2.90	1.47–5.70	2.85	1.28–6.34
Mother	Low education attainment			1.11	1.05–1.16
	Not working			1.06	1.01–1.12
Household	Number of children without disability			1.003	0.98–1.03
	Have house ownership			0.83	0.78–0.89
Family structure ^a	Lower household expenditure			0.98	0.93–1.04
	Partnered mothers without grandparents			0.97	0.91–1.03
	Single mother with grandparents			1.20	1.04–1.38
	Single mother only			1.60	1.44–1.79

The numbers of object were changed due to complete-case analyses.

^a Reference: Partnered mothers with grandparents

Table 3
The relationship between having a child with a disability and maternal psychological distress by family structure.

		Partnered mothers with grandparents n = 6214		Partnered mothers without grandparents n = 20,362		Single mothers n = 3043	
		OR	95%CI	OR	95%CI	OR	95%CI
Child with disability	Age	1.02	1.01–1.04	1.02	1.01–1.03	0.98	0.96–1.001
	Male	1.04	0.94–1.16	1.03	0.97–1.10	0.87	0.76–1.01
	One child with disability at home	1.34	0.84–2.13	1.80	1.47–2.21	1.42	0.81–2.47
	Two children with disability at home	–	–	2.84	1.14–7.07	4.43	0.51–38.7
Mother	Low education attainment	1.08	0.96–1.21	1.06	0.99–1.13	1.05	0.90–1.22
	Not working	1.10	0.97–1.26	1.03	0.97–1.10	1.71	1.38–2.12
Father	Low education attainment	1.05	0.93–1.17	1.13	1.06–1.20		
	Not working	1.55	1.09–2.19	1.75	1.41–2.16		
Household	Number of children without disability	1.02	0.96–1.08	1.01	0.98–1.03	0.94	0.86–1.03
	Have house ownership	0.78	0.56–1.08	0.87	0.82–0.93	0.67	0.56–0.80
	Lower household expenditure	0.91	0.80–1.03	0.96	0.89–1.04	1.06	0.91–1.23
	Living with grandparents					0.83	0.69–1.00

The numbers of object were changed due to complete-case analyses.

4. Discussion

To the best of our knowledge, this study was the first to report a significant association between raising a child with a disability and maternal psychological distress compared to mothers of children without disabilities based on a nationwide survey in Japan. In addition, the risk for psychological distress of mothers differed among family structures.

4.1. Raising a child with a disability compared to raising a healthy child

In Table 2, raising a child with a disability was significantly related to maternal psychological distress after controlling for confounding factors. Lee (2013) reported a literature review of 28 studies that examined the stress levels, well-being, and sleep problems of mothers of children with developmental disabilities. Five studies compared the differences between mothers of children with disabilities and children with typical development (Bourke-Taylor, Pallant, Law & Howie, 2012; Eisenhower, Baker & Blacher, 2005; Estes et al., 2009; Gray et al., 2011; Montes & Halterman, 2007). Among them, only one study, conducted by Montes and Halterman (2007), used population-based study subjects in the United States and reported that mothers of children with autism were twice as likely to experience poorer mental health

than mothers of children without autism (OR, 95%: 2.42, 1.31–4.45) after controlling for the child's social skills and demographic background. In addition, Brehaut et al. (2009), reported that the caregivers of children with health problems were more likely to experience depressive symptoms (OR, 95%: 2.48, 1.40–4.40) compared to the caregivers of healthy children according to population-based data in Canada after controlling for the characteristics of the child, mother, and household. The current study found a smaller point estimate (AOR, 95%CI: 1.72, 1.44–2.05) for one child with a disability compared to previous population-based studies. The possible reasons were that we could not sufficiently control for the children's behavior or social skills variables and lacked a clear definition of the child's disability, such as a clinical diagnosis. The survey used in this study was originally not aimed to examine child's disability and caregiving situations. Adequate variables are needed to evaluate the conditions of children and parents in a nationwide survey to target caregivers in Japan.

Having two children with a disability resulted in an elevated point estimate (AOR, 95%CI: 2.85, 1.28–6.34). The present study revealed a gradient relationship between increase numbers of children with a disability and maternal mental health compared to the mothers of children who did not need constant care in Japan. Being a single parent had an independent effect on maternal psychological distress, as shown in Table 2; however, the effect of having a child with a disability remained after adjusting for the

family composition. Therefore, it is important for health professionals to focus on the mental health of every mother of a child with a disability and to assess their needs for psychological support.

4.2. Differences according to family structure

Previous study reported that single mothers experienced poorer mental health compared to partnered mothers (Cairney, Boyle, Offord & Racine, 2003; Colton, Janzen & Laverty, 2015). It was consistent with this study shown in Table 2. Single mother was 1.60 times likely to experience psychological distress compared to partnered mothers with grandparent(s). After stratified analyses in Table 3, the mothers in two-parent families showed significant associations with having one or two children with a disability and psychological distress. However, there were no significant associations between mothers in three generation family and single mothers. For one reason, this might be due to the small number of children with disabilities. In this study, 446 mothers had one child with a disability and 24 mothers had two children with a disability among two-parent families. Among two-parent families living with child's grandparents, 87 mothers had one child with a disability, but only three mothers had two children with a disability. Among single mothers, 61 mothers had one child and eight mothers had two children with a disability. Since the stratified analyses led to small numbers of explanatory variable, we could not obtain statistical significance between maternal psychological distress and the increase numbers of children with disability in comparison with other children. Further research should include larger number of children with a disability to compare maternal mental health in detailed familial situations, such as using multiple years of data from CSLC.

In terms of family structure, this study showed similar proportions of single mother among children with a disability (11.0%) and children without a disability (10.5%). According to nationwide researches, the proportion of single parenthood was higher among children with a disability or health problems compared to other children in Canada (22.5% vs 16.3%) (Brehaut et al., 2009), UK (34.1% vs 25.6%) (Blackburn, Spencer & Read, 2010), and US (47.3–60.1% vs 41.5–49.1%) (Szilagyi et al., 2003). On the other hand, co-residence with grandparent(s) was significantly lower when a child had a disability (16.2%) compared to a child without disability (24.7%) in Japan. Traditionally, the proportion of elderly people living with their adult children is much higher than in Western countries, which is approximately 40% among households with elderly in Japan (The Ministry of Health Labour and Welfare, 2013; Kono, 2000). However, the tradition of intergenerational co-residence has been decreasing due to social and economic changes, such as the increase in women's educational attainment and employment (Kono, 2000). Therefore, it is important to assess the mental health of single or partnered mothers who have a child with a disability and do not live with children's grandparents.

4.3. Limitations and further implications

The present study had several limitations. First, this study was a cross-sectional study that did not reveal the causal mechanism between the child's disability and the mental health of mothers. Careful considerations are needed to interpret results in this study. Raina et al. (2004) suggested a multidimensional model that included not only a single relationship but also multiple relationships between caregivers' health and burden of care for children with a disability. According to that model, as a possible underlying mechanism, children's impairment is related to caregiving demands and perceptions for formal care services and the self-perception of caregivers. Further research is needed to include

variables of the children's detailed health conditions, perceptions of caregivers, service utilization, social support, and actual child-rearing situations, such as paternal involvement in child care, to examine the mechanisms of the relationships between maternal mental health and a child's disability. In terms of practical implications, health professionals should assess the health conditions of the caregivers and provide advice for the use of a formal care service to cope with their needs and to eliminate psychological distress.

Second, the definition of disability in this study assessed the care-needed status using functional impairment. It was originally developed for the elderly, closely correlated with the functional independence measure (FIM), and widely used for certification of long-term care for the elderly (Tsuboi, Murakami, Kurumadani, Shimizu, and Fujiwara (2002)). The impairment is one of important concept in the International Classification of Functioning, Disability, and Health (ICF) (World Health Organization, 2001) in despite of medical diagnosis for disorders or diseases. The variable of activity limitation used in this study represented whether the child had an experience of being bedridden or absent from school during last month. We did not employ the activity limitation to define disability status of children because it might reflect acute conditional changes rather than usual chronic conditions. Therefore, the operational definition in this study targeted broad range of non-specific disability status, such as developmental disabilities, intellectual delays, or physical impairment. One systematic research (van der Lee, Mokkink, Grootenhuis, Heymans and Offringa (2007)) reported there was no international consensus about the definition of chronic health conditions in childhood. For example, previous studies (Brehaut et al., 2009; Kohen et al., 2007;) used broad inclusion criteria using activity limitations, functional problems, clinical diagnoses, or elevated service use. The children we targeted in this study required care in addition to general parenting care. Caregivers of children with health problems experience common challenges and life experiences in despite of the specific diagnosis (Kohen et al., 2007). Further research is needed to use the various criteria as previous studies to broadly evaluate maternal mental health and children's health conditions. In addition, we excluded children younger than six because the survey asked only about children aged six and over to define the disability. Further research should examine the situation among mothers of younger children with and without disabilities.

Third, we performed a complete case analysis in multiple logistic regressions. The numbers of objects in Tables 2 and 3 decreased due to missing values. The proportions of missing values ranged from 0.1% to 8.0% for the variables of maternal and paternal educational attainment, working status, and monthly expenditure. We could not employ multiple imputation methods because of the data merging process (i.e., single mothers lacked all paternal variables). There is a possibility that bias exists because the confounders were not adequately controlled and we assessed specific characteristics which only appeared among subjects with complete answers to questions. The next analysis will consider more statistical methods, including multiple imputation and data linkage.

Lastly, we compared the differences among mothers living with a partner, a partner and grandparents, or alone. However, we excluded other types of family structures due to the inability to define the child-parent relationships in complicated families, i.e., several married couples and unmarried adults living together with several children. The socioeconomic situation and social support among them may be different from that of typical two-parent, single-parent, or three-generation families. A sophisticated strategy is needed to define the child-parent relationships within the household survey in future studies.

5. Conclusion

This study reported the significant association between raising a child with a disability and maternal psychological distress in comparison to mothers who raised a child without disability using a population-based survey in Japan. The association of raising a child with a disability and maternal mental health varied according to the number of children with a disability and the family composition. Attention should be paid to not only single mothers but also partnered mothers in two-parent families who have a child with a disability. It is important for health professionals to focus on the mental health of every mother of a child with a disability in different family structures, and to assess their needs for psychological support.

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RESEARCH ARTICLE

How Possibly Do Leisure and Social Activities Impact Mental Health of Middle-Aged Adults in Japan?: An Evidence from a National Longitudinal Survey

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Data Availability Statement: All the data underlying the findings in the study are not available upon request because of the legal restrictions by Japanese Article 33 (Provision of Questionnaire Information) of Statistics Act, by the Statistic Bureau, Ministry of Internal Affairs and Communications. The data used in this study are conducted by the Ministry of Health, Labour and Welfare (MHLW) in Japan and therefore, users of these data are strictly limited to those who have obtained official permission from the Minister of Health, Labour and Welfare. By law, it is not permitted for data-applicants to take the data out of Japan.

Abstract

Objectives

This study aimed to investigate longitudinal relations between leisure and social activities and mental health status, considering the presence or absence of other persons in the activity as an additional variable, among middle-aged adults in Japan. This study used nationally representative data in Japan with a five-year follow-up period.

Methods

This study focused on 16,642 middle-aged adults, age 50–59 at baseline, from a population-based, six-year panel survey conducted by the Japanese Ministry of Health, Labour and Welfare. To investigate the relations between two leisure activities ('hobbies or cultural activities' and 'exercise or sports') and four social activities ('community events', 'support for children', 'support for elderly individuals' and 'other social activities') at baseline and mental health status at follow-up, multiple logistic regression analysis was used. We also used multiple logistic regression analysis to investigate the association between ways of participating in these activities ('by oneself', 'with others', or 'both' (both 'by oneself' and 'with others')) at baseline and mental health status at follow-up.

Results

Involvement in both leisure activity categories, but not in social activities, was significantly and positively related to mental health status in both men and women.

Furthermore, in men, both 'hobbies or cultural activities' and 'exercise or sports' were significantly related to mental health status only when conducted 'with others'. In women, the effects of 'hobbies or cultural activities' on mental health status were no differences regardless of the ways of participating, while the result of 'exercise or sports' was same as that in men.

Those who want to use the data for any purpose should contact the Statistics and Information Department of the MHLW. Please refer to the following URL: <http://www.mhlw.go.jp/toukei/sonota/chousahyo.html>.

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Competing Interests: The authors have declared that no competing interests exist.

Conclusions

Leisure activities appear to benefit mental health status among this age group, whereas specific social activities do not. Moreover, participation in leisure activities would be effective especially if others are present. These findings should be useful for preventing the deterioration of mental health status in middle-aged adults in Japan.

Introduction

Recently, the prevalence rate of mental disorders has been increasing in Japan. The total number of people with mood disorders (including bipolar disorder) was estimated as 958,000 in 2011, of whom 426,000 people were middle-aged adults aged 40 to 64 [1]. Mental health problems are an important contributor to the risk of suicide [2], which was the third most common cause of death (after cancer and heart disease) among middle-aged Japanese adults in 2013 [3].

Growing evidence has indicated that leisure activities (e.g. hobbies, cultural activities, exercise and sports) and social activities (e.g. volunteering and community activities) benefit mental health status among middle-aged and older adults. For example, some cross-sectional and longitudinal studies have reported positive relations between certain types of hobbies or cultural activities, such as going to the cinema or reading newspapers or books, and mental health status among middle-aged and older adults [4, 5]. A cross-sectional study in Japan, Wada et al. reported that regular leisure activity was associated with a reduction in depressive symptoms among workers age 20 to 69 [6], and Wakui et al., using two-year longitudinal data, reported that doing leisure activities at least once per week was inversely related with depression among middle-aged and older caregivers [7].

With regard to exercise and sports activities, considerable evidence exists about their effects on mental health status, and some previous meta-analyses have indicated that exercise interventions were effective in sustaining good mental health status among middle-aged and older adults [8–11].

For social activities, some studies have investigated longitudinal relations between volunteering and mental health status. For example, Li and Ferraro reported that formal voluntary activity was good for mental health status among people aged 60 or older [12]. Potočnik and Sonnentag showed that volunteering improved retirees' quality of life over a period of two years [13]. In a study of middle-aged Japanese men, those who engaged in more hours of volunteer work had fewer depressive symptoms [14].

Furthermore, the presence of other persons when one is doing these activities can also help to sustain mental health status by providing social relationships. Some meta-analyses have suggested that interventions addressing social relationships can reduce depression [15, 16]. Longitudinal studies with large populations have shown similar findings. One 10-year follow-up study reported that lack of social relationships was a major risk factor for depression among American adults age 25 to 75 [17]. In an 18-year follow-up study, participation in group leisure or social activities was found to benefit the mental health status of older adults [18]. These findings suggest the possibility that doing activities with other persons may have additional positive effects that are not achieved if one engages in leisure activities alone.

However, the effects of leisure and social activities on mental health status among middle-aged adults are still unclear in Japan. No study considering a broad range of leisure and social activities has been conducted, nor has any study investigated whether causal relations between these activities and mental health status are affected by the presence of other persons.

Thus, this study aimed to investigate longitudinal relations between leisure and social activities and mental health status among middle-aged adults, using nationally representative data in Japan, while also considering, as an additional variable, the presence or absence of other persons in those activities.

Methods

Study population and procedure

This study used panel data extracted from a nationwide, population-based survey, the 'Longitudinal Survey of Middle-aged and Elderly Persons (LSMEP)' conducted once a year on the first Wednesday of November as of 2005 by the Ministry of Health, Labour and Welfare (MHLW) in Japan. Respondents to the survey were extracted randomly through a stratified two-stage sampling. First, 2,515 districts were selected at random from the entire 5,280 districts surveyed by a population-based 'Comprehensive Survey of the Living Conditions of People on Health and Welfare' conducted by the MHLW in 2004. Second, 40,877 residents were chosen randomly from those aged 50 to 59 living in each selected district, in proportion to the population size.

In 2005, the first year of the survey, the questionnaires were drop off to the respondents' homes by enumerators. Then, the enumerators collected the self-completed questionnaire several days later. As of 2006, the method had changed from a "drop-off" to mail survey and so the questionnaire was mailed only to those who had responded to the first survey in 2005. 'LSMEP' has not recruited new samples since the first year of survey.

We used data from the first and sixth surveys in 2005–2010. Of the 40,877 people who received a self-administered questionnaire, 34,240 responded to the survey in 2005 (response rate: 83.8%) and these respondents were followed up thereafter. In 2010, the number of respondents decreased to 26,220 (response rate: 64.1%). Out of these, we excluded respondents who had missing values in K6 scale and those who had bad mental health status (K6 total score of 5 points or above). Furthermore, respondents who had some difficulties in activities of daily living were also excluded because they could potentially not do some leisure or social activities, especially exercise or sports. Finally this study used 16,642 respondents (valid response rate was 63.5%).

We obtained an official permission to use 'LSMEP' by the MHLW on the basis of Article 32 of the Statistics Act. An ethical review of 'LSMEP' was not required, based on the 'Ethical Guidelines for Epidemiological Research' of the Japanese government [19].

Measurements

Mental health status. Mental health status was assessed using the Japanese version of the Kessler 6 (K6) scale [20], a screening scale for psychological distress that can effectively discriminate between cases and non-cases of Diagnostic and Statistical Manual of Mental Disorders (DSM-IV) disorders [21]. Respondents answered six items on a 5-point Likert scale, and responses on each item were transformed to scores ranging from 0 to 4 points. A higher total score corresponds to a poorer mental health condition. All respondents were split into two groups, 'good mental health status' (scores below 5 points) or 'bad mental health status' (5 points or above); the 5-point mark has been identified as the optimal cut-off point for screening mood and anxiety disorders in Japan (100% sensitivity and 68.7% specificity), and it has been used in previous Japanese studies [22, 23]. The Japanese version of the K6 has been validated [20], and the internal consistency reliability (Cronbach's alpha) of the scale in this study was 0.88.

Leisure and social activities. The respondents were asked whether they participated in two types of leisure activities ('hobbies or cultural activities' and 'exercise or sports') and four types of social activities ('community events', 'support for children', 'support for elderly individuals' and 'other social activities') within the past one year from the date of the survey. Those who answered 'yes' to each of these question were categorized as 'active', and those who answered 'no' were categorized 'inactive'. Those who participated in each of these activities were also asked to indicate in what way they 'mainly' participated in the activity ('by oneself', 'with families or friends', 'with co-workers (including former co-workers)', 'in a neighbourhood community association' or 'in a non-profit organization or corporation in the public interest'). For the purposes of this study, respondents were categorized into three groups: 'by oneself', 'with others' or 'both' (both 'by oneself' and 'with others').

Demographic and socioeconomic status. Demographic and socioeconomic status included age (calculated from the month and year of birth), gender, living arrangement (spouse, child or children, father, mother, father-in-law and mother-in-law), job status (employed or unemployed), personal income and family care provision.

Chronic diseases. Respondents answered the presence of chronic diseases (diabetes, heart diseases, cerebral stroke, high blood pressure, hyperlipidemia and cancer). They were rated on a dichotomized scale (yes or no).

Health behaviour. Health behaviour included smoking status (smoker or non-smoker) and drinking alcohol status (drinker or nondrinker).

Statistical analysis

We used the multiple imputation by chained equations to handle missing data in this study. Analysis of imputed datasets reduces the potential bias introduced by missing data. This method assumes that data are missing at random, whereby any systematic differences between the missing and observed values can be explained by differences in observed data [24]. Missing values were imputed according to a model consisting of other all variables, and we used multiple imputation to create and analyse 10 multiply imputed datasets. Imputed data were analysed by gender.

At first, in order to investigate the relations between leisure and social activities in the baseline survey and mental health status in the follow-up survey, two kinds of multiple logistic regression models were applied as follows. Model 1 included the six types of leisure and social activities as independent variables, separately ('hobbies or cultural activities', 'exercise or sports', 'community events', 'support for children', 'support for elderly individuals' and 'other social activities'); Model 2 included the two types of leisure activities ('hobbies or cultural activities' and 'exercise or sports') and a summary index which indicates the involvement in at least one of the four social activities ('community events', 'support for children', 'support for elderly individuals' and 'other social activities') as independent factors.

Furthermore, we used a multiple logistic regression analysis to investigate the association between the ways of participating in those leisure and social activities ('inactive', 'by oneself', 'with others' or 'both') in the baseline survey and mental health status in the follow-up survey.

These multiple logistic regression analyses were adjusted for demographic and socioeconomic status, physical health condition, health behaviour and mental health status at the baseline. The level of significance for all analyses was set at $p < 0.05$. All statistical analyses were performed using IBM SPSS version 23.0.

Results

Descriptive statistics of the characteristics are shown in [Table 1](#). The K6 score increased significantly from the baseline to the follow-up survey periods in both men and women (using paired t-test: $p < 0.001$).

One thousand three hundred fifty three (16.6%) of men, and 1,677 (19.8%) of women were categorized into the group of bad mental health status in the follow-up surveys. The proportion of people who had bad mental health status in women was larger than that in men.

Table 1. Characteristics of respondents after multiple imputation of missing values.

	Men (n = 8175)		Women (n = 8467)		p
	Mean (SE)	n (%)	Mean (SE)	n (%)	
Demographic and socioeconomic status					
Age (years)	54.76	(0.03)	54.73	(0.03)	0.446 ^a
Living arrangement					
Spouse (Presence)		7193 (88.0)		7189 (84.9)	<0.001 ^b
Child(ren) (Presence)		5206 (63.7)		5213 (61.6)	0.005 ^b
Father (Presence)		874 (10.7)		271 (3.2)	<0.001 ^b
Mother (Presence)		1948 (23.8)		680 (8.0)	<0.001 ^b
Father-in-law (Presence)		208 (2.5)		526 (6.2)	<0.001 ^b
Mother-in-law (Presence)		464 (5.7)		1317 (15.5)	<0.001 ^b
Job status (Employment)		7858 (96.1)		6051 (71.5)	<0.001 ^b
Personal income (thousand yen)	52.13	(0.80)	30.65	(0.60)	<0.001 ^a
Family care provision (Yes)		435 (5.3)		759 (9.0)	<0.001 ^a
Chronic diseases					
Diabetes (Presence)		662 (8.1)		331 (3.9)	<0.001 ^b
Heart diseases (Presence)		238 (2.9)		100 (1.2)	<0.001 ^b
Cerebral stroke (Presence)		72 (0.9)		50 (0.6)	0.028 ^b
High blood pressure (Presence)		1504 (18.4)		1247 (14.7)	<0.001 ^b
Hyperlipidemia (Presence)		739 (9.0)		725 (8.6)	0.277 ^b
Cancer (Presence)		81 (1.0)		126 (1.5)	0.004 ^b
Health behaviour					
Smoking status (Smoker)		3774 (46.2)		886 (10.5)	<0.001 ^b
Drinking alcohol status (Drinker)		2033 (24.9)		5874 (69.4)	<0.001 ^b
Leisure and social activities					
Hobbies or cultural activities (Active)		4784 (58.5)		5533 (65.3)	<0.001 ^b
Exercise or sports (Active)		3971 (48.6)		3835 (45.3)	<0.001 ^b
Community events (Active)		2490 (30.5)		2494 (29.5)	0.158 ^b
Support for children (Active)		436 (5.3)		558 (6.6)	0.001 ^b
Support for elderly individuals (Active)		476 (5.8)		761 (9.0)	<0.001 ^b
Other social activities (Active)		909 (11.1)		1038 (12.3)	0.022 ^b
Mental health status					
Baseline	0.98	(0.02)	1.14	(0.02)	<0.001 ^a
Follow-up	2.06	(0.03)	2.43	(0.04)	<0.001 ^a
Bad mental health at follow-up		1353 (16.6)		1677 (19.8)	<0.001 ^b

^a Independent t-test

^b Chi-square test.

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Table 2. Multiple logistic regression analyses for the relations between leisure and social activities and mental health status at the follow-up period.

		Model 1 ^a			Model 2 ^a		
		AOR	95% CI	p	AOR	95% CI	p
Men							
Hobbies or cultural activities	Active (ref. Inactive)	0.85	0.74–0.98	0.023	0.84	0.73–0.98	0.028
Exercise or sports	Active (ref. Inactive)	0.85	0.74–0.98	0.029	0.86	0.74–0.99	0.034
Community events	Active (ref. Inactive)	1.01	0.87–1.18	0.855			
Support for children	Active (ref. Inactive)	0.89	0.60–1.31	0.550			
Support for elderly individuals	Active (ref. Inactive)	1.32	0.94–1.87	0.110			
Other social activities	Active (ref. Inactive)	0.85	0.65–1.13	0.263			
Social activities	Active (ref. Inactive)				0.98	0.86–1.12	0.740
Women							
Hobbies or cultural activities	Active (ref. Inactive)	0.72	0.63–0.83	0.000	0.71	0.61–0.84	0.000
Exercise or sports	Active (ref. Inactive)	0.88	0.77–1.00	0.042	0.88	0.78–1.01	0.069
Community events	Active (ref. Inactive)	0.96	0.81–1.13	0.578			
Support for children	Active (ref. Inactive)	1.18	0.86–1.62	0.294			
Support for elderly individuals	Active (ref. Inactive)	1.16	0.86–1.58	0.320			
Other social activities	Active (ref. Inactive)	0.88	0.63–1.22	0.420			
Social activities	Active (ref. Inactive)				1.01	0.90–1.14	0.852

^a Adjusted for demographic and socioeconomic status, physical health condition, chronic diseases and mental health status at the baseline.

AOR: Adjusted odds ratio; CI: Confidence interval.

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The number of respondents who participated in each of leisure and social activities at the baseline was as follows: 4,784 (58.5%) of men and 5,533 (65.3%) of women in ‘hobbies or cultural activities’, 3,971 (48.6%) of men and 3,855 (45.3%) of women in ‘exercise or sports’, 2,490 (30.5%) of men and 2,494 (29.5%) of women in ‘community events’, 436 (5.3%) of men and 558 (6.6%) of women in ‘support for children’, 476 (5.8%) of men and 761 (9.0%) of women in ‘support for elderly individuals’ and 909 (11.1%) of men and 1038 (12.3%) of women in ‘other social activities’. The proportion of people who participated in ‘hobbies or cultural activities’, ‘support for children’, ‘support for elderly individuals’ and ‘other social activities’ in women was larger than that in men, whereas that of people who participated in ‘exercise or sports’ in men larger than that in women.

Table 2 shows the results of multiple logistic regression analyses. In regard to men, the result of Model 1 showed that ‘hobbies or cultural activities’ (OR 0.85, 95% CI 0.74–0.98, $p < 0.05$) and ‘exercise or sports’ (OR 0.85, 95% CI 0.74–0.98, $p < 0.05$) were significantly related to mental health status at the follow-up period. Model 2 also showed a similar result to Model 1, such that ‘hobbies or cultural activities’ (OR 0.84, 95% CI 0.73–0.98, $p < 0.05$) and ‘exercise or sports’ (OR 0.86, 95% CI 0.74–0.99, $p < 0.05$) had a significant relation with mental health status in the follow-up survey.

In regard to women, the result of Model 1 showed that ‘hobbies or cultural activities’ (OR 0.72, 95% CI 0.63–0.83, $p < 0.001$) and ‘exercise or sports’ (OR 0.88, 95% CI 0.77–1.00, $p < 0.05$) significantly associated with mental health status, as was the case with men. By contrast, Model 2 showed that ‘hobbies or cultural activities’ were significantly related only to mental health status at the follow-up period (OR 0.71, 95% CI 0.61–0.84, $p < 0.001$).

Furthermore, the relations between the ways of participating in ‘hobbies or cultural activities’ or ‘exercise or sports’ and mental health status were investigated in both men and women.

Table 3 shows the results when the ‘in active’ category as reference was selected. Regarding men, both ‘hobbies or cultural activities’ and ‘exercise or sports’ were significantly related to mental health status only when conducted ‘with others’ (‘hobbies or cultural activities’: OR 0.83, 95% CI 0.71–0.97, $p < 0.05$; ‘exercise or sports’: OR 0.84, 95% CI 0.73–0.98, $p < 0.05$). Regarding women, ‘exercise or sports’ was significantly related to mental health status only when conducted ‘with others’ (OR 0.86, 95% CI 0.75–0.99, $p < 0.05$), whereas significant ORs for ‘hobbies or cultural activities’ were observed in all of three categories (‘by oneself’: OR 0.74, 95% CI 0.62–0.90, $p < 0.01$; ‘with others’: OR 0.76, 95% CI 0.67–0.87, $p < 0.001$; ‘Both’: OR 0.55, 95% CI 0.33–0.93, $p < 0.05$).

Additionally, when the ‘with others’ category as reference was selected, the significant OR was observed only in the ‘inactive’ category (‘hobbies or cultural activities’: OR 1.21, 95% CI 1.03–1.41, $p < 0.05$ in men, OR 1.32, 95% CI 1.16–1.50, $p < 0.01$ in women; ‘exercise or sports’ OR 1.18, 95% CI 1.02–1.38, $p < 0.05$ in men, OR 1.16, 95% CI 1.01–1.34, $p < 0.05$ in women).

Discussion

The main objective of this study was to investigate relations between leisure and social activities and mental health status among middle-aged adults in Japan using nationally representative data. The results show that participation in leisure activities such as ‘hobbies or cultural activities’ and ‘exercise or sports’ at the baseline would be positively related to mental health status at the period after five-year follow-up in both men and women. Therefore, that is consistent to some previous cross-sectional and longitudinal studies which have reported relations between involvement in hobbies or cultural activities and mental health status in middle-aged and older adults [4–7]. Moreover, some meta-analyses have found physical activity interventions to be effective in sustaining mental health status within the same age cohort [8–11]. Leisure activities could play a role in benefitting overall well-being and providing a buffer against stress. This benefit may occur by promoting a variety of social and physical resources that enable individuals to feel refreshed and to cope adequately with stress [25, 26].

In contrast, this study observed no benefits of the participation in social activities at the baseline on mental health status in the follow-up survey in both men and women. Several previous studies reported the longitudinal relations between social activities and mental health status. Li and Ferraro found that volunteer work had beneficial effects on mental health status among older adults [12]. A previous study in Japan suggested that volunteer work was associated with reduced depressive symptoms among adults in later middle age, even after controlling for pre-existing depressive symptoms, socioeconomic factors and physical health [14].

Table 3. Multiple logistic regression analyses for the relations between the way of participation and mental health status at the follow-up periods.

		Men				Women			
		n	AOR ^a	95% CI	p	n	AOR ^a	95% CI	p
Hobbies or cultural activities (ref: Inactive)	By oneself	1350	0.96	0.81–1.15	0.683	1107	0.74	0.62–0.90	0.002
	With others	3317	0.83	0.71–0.97	0.018	4291	0.76	0.67–0.87	0.000
	Both	116	0.85	0.39–1.86	0.689	135	0.55	0.33–0.93	0.024
Exercise or sports (ref: Inactive)	By oneself	1283	0.93	0.77–1.11	0.408	1310	0.89	0.75–1.05	0.158
	With others	2560	0.84	0.73–0.98	0.030	2405	0.86	0.75–0.99	0.039
	Both	126	0.79	0.33–1.89	0.596	120	0.97	0.59–1.56	0.887

^a Adjusted for demographic and socioeconomic status, physical health condition, health behaviour and mental health status at baseline.

AOR: Adjusted odds ratio, CI: confidence interval.

However, these studies did not control the influence of activities other than social activities. Our findings showed no longitudinal relation between social activities and mental health status when the effects of leisure activities were considered.

One previous study reported that social activities were associated with longitudinal changes in mental health status even after considering the influence of leisure activities [27]; however, respondents in that study were much older than those in our study. Because the perceived value of life tends to decrease gradually with age [28], maintaining social activities may be especially important to sustain good mental health status among older adults. However, because our respondents were still relatively young in their fifties, and 96.4% of men and 71.5% of women had a job at the baseline period, almost all of them had other forms of regular social involvement. This could be one reason why our results indicate that leisure activities contribute to mental health status in middle-aged adults, whereas specific social activities do not.

Furthermore, this study investigated the relations between the presence of other persons in leisure activities and mental health status. Our results showed that hobbies or cultural activities in men and exercise or sports in both men and women would reduce such risk only when conducted with others, whereas hobbies or cultural activities in women might have effects on mental health regardless of the presence of others.

These results suggested that social relationships through leisure activities would be the key factor of preventing the deterioration of mental health status regardless of gender differences. Some previous studies have suggested that the improvements in mental health status following exercise or sports could be partially because of the social relations that can be experienced through participating in these activities with others [29, 30]. However, in this study, no relation existed between doing exercise or sports by oneself and mental health status. The previous study in Japan suggested that, even when exercise was performed once a week or more, incident of functional disability might be better prevented if the person participated in a sports organization than if they did not [31]. Our result suggests that the psychological effects of social relations may be especially needed in not only the case of exercise or sports, but also the case of hobbies or cultural activities. On the other hand, we found no significant difference between 'by oneself' or 'both' category and 'with others' category. Thus, the effect of the presence of others may need to be carefully considered.

This is the first study to show a longitudinal relation between leisure and social activities and mental health status among middle-aged adults in Japan. The study has several particular strengths. First, it used a good set of nationally representative data. Second, unlike previous studies of middle-aged adults in Japan, our study encompassed a wide range of leisure and social activities. Finally, our study indicated further details about the appropriate way of doing leisure activities to maintain good mental health status while approaching old age (i.e. whether it is effective to do these activities alone or only with others).

The study also has several limitations. First of all, this study might not completely identify the pure effects of the participation into leisure and social activities on mental health status, because the data in the baseline period must suffer from reversed causality problem between the participation and mental health status. In another word, those who are in better mental health status are more likely to involve into various social activities. Second, although the multiple imputation was used to try to reduce impact of missing variables, our study would still have some selection bias. About 6,000 people did not respond a questionnaire in the baseline survey, and almost 8,000 respondents were dropped out in the follow-up study. Thus, generalization of the results in our study should be done carefully. Third, the participation in leisure and social activities was indicated on a dichotomized scale, and thus, the frequency or variety of participation was not assessed. Fourth, questionnaires about leisure and social activities were self-reported and retrospective, rendering them somewhat inaccurate. Finally, 'LSMEP'

excluded patients in hospitals and clinics and residents of long-term elderly care facilities. These people might have a higher-than-average rate of bad mental health status and might not have been likely to engage in leisure and social activities. Therefore, possibly, the positive relations between leisure and social activities and mental health status were underestimated.

In conclusion, this study indicates that leisure activities might contribute to good mental health status among middle-aged adults in Japan, whereas social activities would provide no mental health status benefit. Moreover, participation in leisure activities would be effective especially if others are present. These findings may be useful for preventing the deterioration of mental health status in middle-aged adults in Japan.

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Author Contributions

Conceived and designed the experiments: FT. Analyzed the data: FT. Wrote the paper: FT HN NT TM. Reviewing of the manuscript: HN NT TM.

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