

Enterothorax After Hepatic Surgery: A Single-Center Experience

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Abstract

Background Enterothorax (ET) is a rare complication after hepatic surgery. The literature in this field is limited and mainly based on case reports. The aim of this study was to review our department's experience.

Patients and methods We retrospectively analyzed 602 patients who underwent hepatic resection between November 2008 and December 2016. Major hepatic surgery ($n = 321$) was defined as right or extended right hepatectomy ($n = 227$), left or extended left hepatectomy ($n = 63$), trisegmentectomy ($n = 13$), and living donor liver transplantation ($n = 18$). ET cases were identified by analyzing clinical courses and radiological imaging.

Results ET was observed in five out of 602 patients (0.8%). All patients developed the complication after major hepatic surgery (five out of 321, 1.6%). ET exclusively occurred after right ($n = 3$) or extended right hepatectomy ($n = 2$). Median time to diagnosis was 22 months. Radiological imaging showed herniation of small ($n = 2$), large bowel ($n = 2$), or omental fat ($n = 1$) with a median diaphragmatic defect of 3.9 cm. Two patients presented with acute incarceration and underwent emergency surgery, one patient reported recurrent pain and underwent elective repair, and two patients refused surgery. Follow-up imaging in two operated patients showed no recurrence of ET after 36 and 8 months.

Conclusions Patients after right hepatectomy have a substantial risk of ET. Acute right upper quadrant pain and/or dyspnea after hepatectomy should be investigated with adequate radiological imaging. Elective surgical repair of ET is recommended to avoid emergency surgery in case of incarceration.

Introduction

Enterothorax (ET) is defined as a prolapse of abdominal viscera to the thoracic cavity [1]. In contrast to diaphragmatic hernia, no peritoneal sac is present and abdominal

organs are directly penetrating into the chest. Generally, congenital ET [2] as a distinct entity can be distinguished from ET secondary to traumatic or iatrogenic diaphragmatic defects [3]. While posttraumatic ET is a well-described complication of blunt or penetrating trauma [4], much less is known about ET after elective abdominal surgery. Recently, an increase in ET in the era of minimally invasive surgery has been suggested, probably caused by less adhesion formation after laparoscopy compared with the open transabdominal approach [5]. Iatrogenic ET has been reported after esophagectomy and total gastrectomy [6–8], but also following radio frequency ablation of liver tumors [9] and major liver resection [10–13]. In retrospective cohort studies, the published

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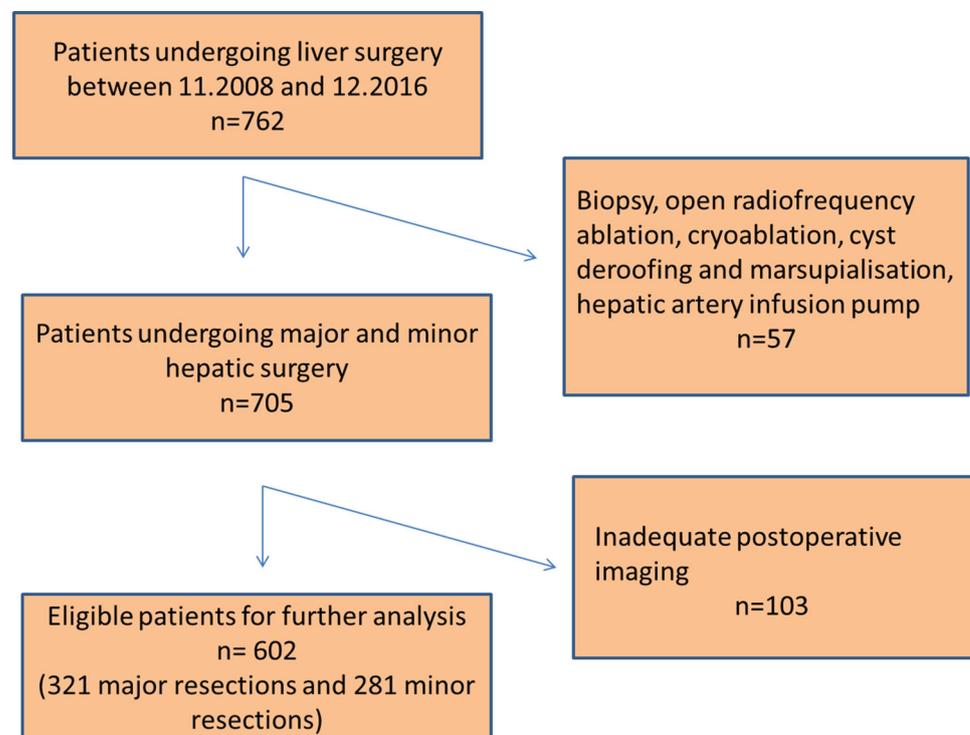
incidence of ET after major hepatic resection varies between 0.6 and 1.0% [10, 14]. Clinically, patients often present as an emergency with acute symptoms of bowel obstruction, recurrent upper abdominal pain, or respiratory distress [10]. Prompt detection and therapy of ET has been shown to correlate with improved prognosis [10]. In this setting, it was challenging to review our department's experience with ET after major hepatic surgery.

Patients and methods

We retrospectively analyzed all patients ($n = 762$) who underwent hepatic surgery between November 2008 and December 2016 at the University Hospital of Zurich, Switzerland. After exclusion of minor hepatic procedures (cyst deroofing, cryoablation, radio frequency ablation, and hepatic artery infusion pumps, $n = 57$) and of patients without adequate radiological (CT scan or MRI) follow-up (benign pathology and living liver donors, $n = 103$), 602 patients were eligible for analysis (Fig. 1). The study cohort consisted of patients after major liver resection (≥ 3 segments, $n = 321$), wedge resections or segmentectomies ($n = 234$), and bisegmentectomies ($n = 47$). In detail, major liver resection was defined according to the Brisbane 2000 terminology [15] as right hepatectomy (RH) ($n = 166$), extended RH ($n = 61$), left hepatectomy (LH) ($n = 39$), extended LH ($n = 24$), trisegmentectomy

($n = 13$), and living liver donor transplantation ($n = 18$). Nineteen patients underwent additional diaphragmatic resection during major hepatectomy. Thirty-six resections were performed laparoscopically (6%); all of them were minor resections (21 atypical or wedge resections, six segmentectomies, and nine bisegmentectomies). Sixty-one patients underwent concomitant thermal ablation during liver surgery (radio frequency ablation, microwave ablation or nanoknife). Postoperative radiological imaging (CT or MRI scans) of all eligible patients was reviewed by an experienced radiologist (C.R.). Enterothorax was defined as a defect in the diaphragm with herniation of abdominal viscera to the thoracic cavity. When a disruption of the posterolateral aspect of the diaphragm was seen and the defect was present also on preoperative exams, it was considered a Bochdalek hernia [16]. Records of patients who developed ET were reviewed with respect to demographic characteristics, symptoms, diagnostic workup, surgical procedures, postoperative course and long-term outcome. Baseline characteristics were summarized as mean (standard deviation) or median [range] for continuous variables and frequencies and percentages for categorical variables, respectively. Institutional review board approval was obtained for this study (BASEC-Number 2017-01878).

Fig. 1 Flow diagram representing the all eligible patients ($n = 762$) and the patients included in the retrospective analysis ($n = 602$)



Systematic literature search

We performed a systematic literature search with the key words «enterothorax and liver resection» and «diaphragmatic hernia and liver resection» on March 8, 2018. Two authors (G.M. and C.K.) independently screened MEDLINE, Cochrane, and PubMed databases. The literature search was restricted to abstracts and papers published in the English language. Inclusion criteria were each type of publication describing an ET in adults after any type of liver resection for any reason, including living donor hepatectomy and living donor transplantation.

Results

Own experience

After a mean radiological follow-up of 22.7 months (± 22 months), the rate of ET after any type of liver resection was 0.8% (5/602). ET was exclusively diagnosed after RH ($n = 3$) and extended RH ($n = 2$), resulting in an incidence of 1.6% after major liver resection (5/321) and 2.2% (5/227) after (extended) RH. Patient characteristics and details of surgical procedures are shown in Table 1. The indications for initial surgery in patients who developed ET were echinococcosis ($n = 2$), cholangiocellular carcinoma ($n = 1$), liver metastatic colorectal cancer ($n = 1$), and giant hemangioma ($n = 1$). None of the patients with ET had an additional diaphragmatic resection or simultaneous thermal ablation at initial hepatic surgery. Five patients were diagnosed with Bochdalek hernia (two bilateral, two right, and one left); none of them developed ET at follow-up. Likewise, none of the patients undergoing laparoscopic liver resection was diagnosed with ET at FU.

Details of patients with ET are summarized in Table 2. Four cases were diagnosed by means of CT scan (Figs. 2, 3) and one by MRI. Median follow-up until diagnosis of ET was 22 months (range 5–96 months). The mean pre-operative BMI of patients with ET was 28.2 kg/m² (± 3.2 kg/m²), and the median size of the diaphragmatic defect was 3.9 cm (2.5–5.5 cm) with herniation of small bowel ($n = 2$), colon ($n = 2$), or omental fat ($n = 1$) to the right chest. In one patient with minor symptoms, ET was diagnosed during regular follow-up, one patient complained of chronic right upper quadrant abdominal pain and underwent surgery in an elective setting, and two patients presented with acute incarceration and underwent immediate surgical repair, whereas one patient was completely asymptomatic and refused surgery. Open revisional surgery was performed in all three cases, and the diaphragmatic defect was closed with non-absorbable sutures (Fig. 4). Reoperated patients had a BMI of 23.7 kg/m², 26 kg/m²,

Table 1 Patients characteristics

Patients characteristics ($n = 602$)	Number (%)	Mean (\pm SD)
Age (years)		58.5 (± 14)
Gender		
Male	393 (65.3)	
Female	209 (34.7)	
Etiology		
Primary liver malignancy	167 (27.7)	
Metastatic liver disease	301 (50)	
Benign liver pathology	50 (8.3)	
Gallbladder cancer	26 (4.3)	
Echinococcosis	37 (6.2)	
Healthy living donor	9 (1.5)	
Cirrhosis	12 (2)	
Operation		
Major liver resection	321 (53.3)	
RH	166 (51.7)	
Extended RH	61 (19)	
LH	39 (12.2)	
Extended LH	24 (7.5)	
Trisegmentectomy	13 (4)	
Living donor transplantation	18 (5.6)	
Minor liver resection	281 (46.7)	
Wedge resection or segmentectomy	234 (83.3)	
Bisegmentectomy	47 (16.7)	
Number of laparoscopic resections	36 (6)	
Concomitant diaphragmatic resection	21 (3.5)	
Concomitant thermal ablation	61 (10.1)	
RFA	25 (41)	
MWA	24 (39.4)	
Nanoknife	12 (19.6)	
Patients undergoing more than one liver resection	47 (7.8)	
Follow-up (months)		22.7 (± 22)

RFA, radio frequency ablation; MWA, microwave ablation

and 37.8 kg/m², respectively. Radiological follow-up (MRI and CT scan) was available in two patients after 36 and 8 months without evidence of recurrence. One asymptomatic patient with prolapsed omental fat refused to undergo the advised revisional intervention; follow-up imaging showed neither progression of ET nor evidence of an enlarged diaphragmatic defect after 34.5 months.

Review of the literature

The literature search identified ten publications totaling 22 patients with ET after liver resection or living donor transplant (Table 3). In detail, there was one review of the literature [17], three retrospective cohort studies

Table 2 Characteristics of patients with enterothorax ($n = 5$)

No.	Age at primary operation	Gender	Pathology	Tumor diameter (cm)	Primary surgery	Resection of diaphragm	Ascites	Thermal ablation	Time to event (month)	Symptoms	Imaging	Site of hernia	Size of hernia (cor × sag)	Surgical technique	Herniated organ
1.	60	Female	CCC	5.5	Extended RH* with segment 1 resection	No	Yes	No	5	Acute small bowel obstruction	CT	Right thoracic	2.2 × 2.5	Transabdominal primary repair (sutures)	Small bowel
2.	32	Male	Echinococcosis	16.5	RH*	No	No	No	15	Right upper abdominal pain	CT	Right thoracic	5.4 × 5.5	Transabdominal primary repair (sutures)	Small bowel
3.	44	Female	Giant hemangioma	20	RH*	No	No	No	96	Acute bowel obstruction	CT	Right thoracic	3.5 × 3.3	Transabdominal primary repair (sutures)	Colon
4.	60	Female	IPN liver segment V and echinococcosis VII–VIII	3.5 IPN; 1.5 cysts	RH*	No	No	No	22	Mild symptoms	MRI	Right thoracic	3.5 × 3.9	No surgical revision	Colon
5.	53	Male	Hepatic metastasized colorectal cancer	Multiple lesions	Extended RH*	No	No	No	27.5	Asymptomatic	CT	Right thoracic	3.9 × 3.9	No surgical revision	Omental fat

*RH, right hemihepatectomy; IPN, intraductal papillary neoplasm; CCC, cholangiocellular carcinoma

Fig. 2 CT scan showing a herniated small bowel loop (arrowheads) 5 months after right hemihepatectomy in the coronal (a) and sagittal (b) plane

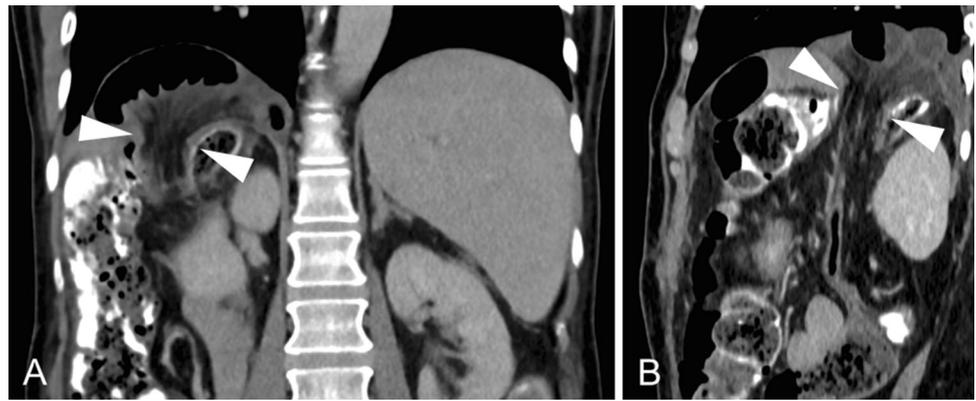


Fig. 3 (a) CT scan showing a prolapse of the right hemicolon through a diaphragmatic defect (arrowheads). (b) The defect can be located in the dorsal diaphragm in the sagittal view

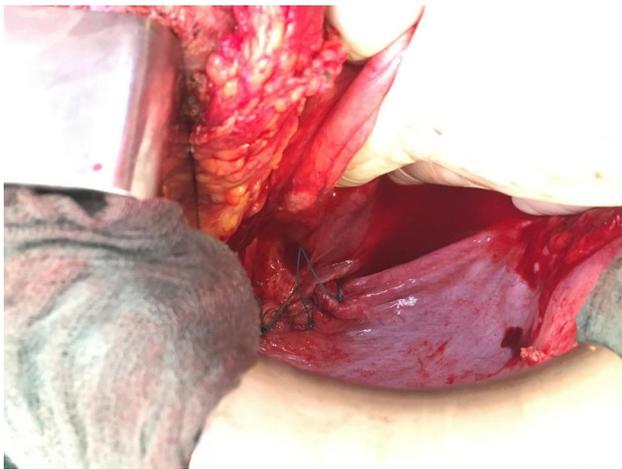
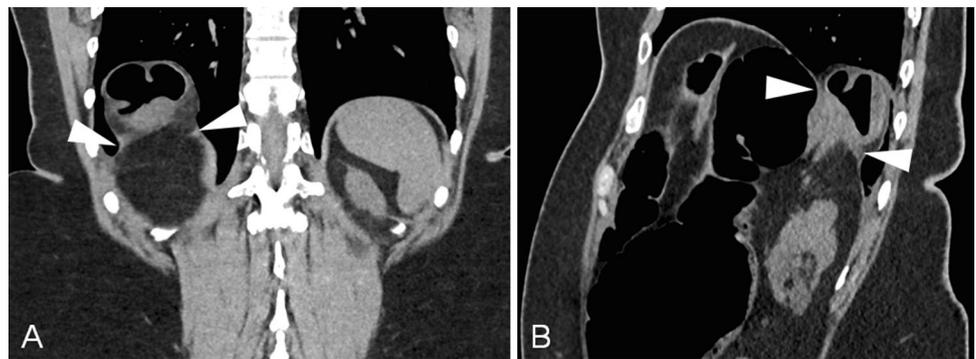


Fig. 4 Direct single-suture closure of the diaphragmatic defect

[10, 14, 18], five case reports [11–13, 19, 20], and one letter to the editor [21]. One case report was excluded because the main text was written in Japanese [22]. Patients with ET presented at a mean age of 48.5y, and the median time from initial surgery to diagnosis of ET was 20 months. The incidence after RH ($n = 14$) was almost

five times higher than after LH ($n = 3$). Eleven out of 22 cases (50%) were observed after living donor hepatectomy (RH $n = 8$, LH $n = 1$, not described $n = 2$), two cases after living donor transplant, five after oncological resections, and four after hepatectomy for benign pathology.

All except two asymptomatic patients presented with right upper quadrant abdominal pain plus symptoms of bowel obstruction. CT scan was the diagnostic modality in all cases. Herniated viscera (described in 11 patients) were colon ($n = 7$), small bowel ($n = 3$), and stomach ($n = 1$). In one patient, reoperation was performed via thoracotomy, and all other cases underwent transabdominal surgery. Non-absorbable suture material was used to close the diaphragmatic defect in all patients, and an additional non-absorbable mesh was used in five cases. In two patients with intestinal obstruction, small bowel resection was performed.

Discussion

ET is a rare complication after liver surgery. The total incidence of 1.6% after major liver resection and 2.2% after (extended) RH in this retrospective study compares well with the literature [10]. Interestingly, all five patients

Table 3 Literature on enterothorax after liver resection in adults

Author and year [reference number]	Type of publication	Number of patients analyzed	Number of patients with ET and main pathology
Livingstone 2016 [17]	Literature review and case report	–	2 after living donor right hepatectomy
Tabrizian 2012 [10]	Retrospective cohort study	993	10 (5 after malignant disease, 4 after benign disease, 1 after living donor transplant)
Kousoulas 2011 [18]	Retrospective cohort study	87	2 living donor hepatectomy
Yaprak 2011 [14]	Retrospective cohort study	181	1 after right donor hepatectomy
Hawxby 2006 [11]	Case report	–	1 after right donor hepatectomy
Vernadakis 2012 [12]	Case report	–	1 after right donor hepatectomy
Jeng 2015 [13]	Case report	–	1 after right donor hepatectomy
Dieter 2011 [20]	Case report	–	2 after living donor right hepatectomy
Perwaiz 2010 [19]	Case report	–	1 after living donor liver transplant
Mizuno 2014 [22]	Letter to the editor	–	1 after living donor left hepatectomy

of this series developed ET following RH or extended RH. The increased risk for ET after major right hepatic resection was confirmed by our review of the literature, showing an almost five times higher rate after RH compared with LH. At the time of diagnosis, the majority of our patients were symptomatic with chronic right upper abdominal pain or signs of acute intestinal obstruction after a median period of 22 months from the primary operation, which is similar to other research [10].

Known risk factors for ET after liver surgery are major resections for large hepatic lesions, RH or extended RH [10], direct trauma or thermal injury to the diaphragm, poor nutritional status, postoperative ascites, high pressure gradient between abdomen and thorax, and fragile diaphragmatic tissue [9, 11, 23, 24]. Likewise, all our patients developed ET after RH or extended RH, and two out of five patients with ET underwent surgery because of a large hepatic lesion. Mean preoperative BMI was 28.2 kg/m², indicating that overweight may play a role as a risk factor for ET. Postoperative mild ascites was present in one patient with ET only, and none underwent simultaneous thermal ablation during hepatic surgery. 7.8% of our patients underwent more than one resection, and none developed ET.

Tabrizian et al. found that patients requiring a simultaneous diaphragmatic resection are at increased risk of ET at follow-up (5.4 vs. 0.7%, $p = 0.011$) [10]. In contrast, none of our ET patients underwent diaphragmatic resection during initial hepatic surgery. In conclusion, the extent and type of the initial liver resection seems to be a critical factor for the development of ET.

During surgical mobilization of the liver, small diaphragmatic lesions may remain unnoticed. Because of the strong contractile force of the diaphragm [25], these microlesions may enlarge with time, leading to a transmural diaphragmatic defect and ET. The sinewy central

part of the diaphragm (pars tendinea) is strong and resistant to traction forces, whereas on the right side, small muscular lesions are more likely to enlarge with time. Accordingly, ET occurs more frequently after right hepatectomy. In contrast, large diaphragmatic defects, such as resections, are usually meticulously closed during initial surgery, which may explain the absence of ET after simultaneous diaphragmatic resection in our series.

The existing literature suggests that living liver donors are at increased risk for ET [11–13, 18]. Kousoulas et al. [18] analyzed 87 (RH $n = 36$, LH $n = 47$) living donor transplantations. Three donors (3.5%)—all after RH—were diagnosed with ET. In contrast, none of the nine liver donors in our study (RH $n = 7$, LH $n = 2$) developed ET. However, we do not perform routine radiological follow-up in liver donors, and consequently, some cases might have been missed. In a recent review of the literature, the incidence of ET in living liver donors was 2.3% [17]. Obstruction or strangulation was present in 45% of ET cases, necessitating bowel resection in a substantial proportion of patients. Consequently, the authors suggested radiological screening with conventional chest radiography in living liver donors to improve early diagnosis of ET. In our experience, however, conventional chest radiography has shown a limited sensitivity for the detection of diaphragmatic defects, and therefore, we have excluded all patients without cross-sectional follow-up imaging. In malignant cases, standard oncological aftercare including CT scan or MRI is usually adequate to detect ET at an early asymptomatic stage. However, as cross-sectional imaging is usually not performed in benign cases at follow-up; we are convinced that a specific radiological surveillance program may be useful, particularly in high-risk situations such as RH, obesity, or particularly frail diaphragmatic tissue. Indication and timing of surgery in asymptomatic patients with radiologically proven ET is another

controversial issue. In our experience, the high proportion of emergency interventions in this subset of patients clearly suggests a low threshold for early elective surgical repair.

Although ET is rare after major liver surgery, it is a well-known complication after esophagectomy [5, 26–30], especially after minimally invasive approach [8, 31–33]. During esophagectomy, the esophageal hiatus is left open to provide adequate passage of the conduit to the chest. This causes an increased risk of herniation of abdominal contents into the thoracic cavity [26–28]. The increased incidence of ET after minimally invasive esophagectomy is most likely caused by reduced adhesion formation [8, 31, 32]. In a meta-analysis comparing the incidence of symptomatic ET after minimally invasive and open esophagectomy [33], an almost fivefold increase after the minimally invasive approach was found [33]. Similarly, laparoscopic and robotic-assisted liver resections have become popular in many centers for liver surgery during the last 20 years [34–36]. With greater use of minimally invasive techniques, the incidence of ET is expected to increase in the near future and this complication will gain more clinical relevance. In our cohort, only 6% of operations were performed laparoscopically. However, all minimally invasive procedures were minor resections, and none of the patients developed ET. Future studies including minimally invasive major liver resections will show whether the incidence of ET is indeed higher than that currently observed in patients after open surgery.

The management of ET should be prompt at onset of symptoms and strictly aimed at repairing the defect before prolapsed viscera become strangulated. All patients of our series presenting with acute intestinal obstruction underwent immediate surgical repair without the need for bowel resection. Tabrizian et al. described primary closure of the diaphragmatic defect using non-absorbable sutures via an abdominal approach for patients with small-sized (<5 cm) and moderate-sized (5–10 cm) hernias [10]. In contrast, for larger (>10 cm) defects, additional use of a surgical mesh is recommended. In our study, all defects were smaller than 5 cm, and single non-resorbable sutures were applied accordingly.

In summary, our study confirms that patients after right (extended) hepatectomy are at increased risk of ET. Patients with the previous liver resection presenting to the emergency with right hypochondrial or thoracic pain should immediately be scheduled for cross-sectional radiological imaging to exclude this complication, as delayed therapy often results in intestinal resection and increased perioperative morbidity.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Informed consent Informed consent was obtained from all individual participants included in the study.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards. Institutional review board approval was obtained for this study (BASEC-Number 2017-01878).

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