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CONTENTS

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INFLUENCE OF ERAS PROTOCOL ON POSTOPERATIVE OUTCOMES AFTER ELECTIVE COLORECTAL RESECTION SURGERY: A PROSPECTIVE COHORT STUDY - TWO YEARS SINGLE CENTER EXPERIENCE

Duje Apostolski[1], Matea Babić[2], Ivana Grgić[3], Božena Ivančev[4], Zdravko Perko[3]

ABSTRACT

Background: ERAS (Enhanced Recovery After Surgery) protocol is a multimodal pathway of perioperative surgical care consisting of evidence-based procedures. ERAS protocol is hard to comply with, because medical staff often opposes well established practice.

Methods: We analyzed length of hospital stay, postoperative complications, time until first stool passage and introduction of normal nutrition in patients undergoing elective colorectal resection surgery in University Hospital Center Split from October 2016. to October 2018. Patients were divided into 4 groups considering operation type (open/laparoscopic) and application of ERAS protocol (good/poor). Application of 60% or more ERAS steps was considered as well performed protocol.

Results: Groups Laparoscopy/ERAS and Open/ERAS had shorter postoperative hospital stay (Median, IQR; days) than groups Laparoscopy/non-ERAS and Open/non-ERAS (LE 5, 4-8 , OE 6, 5-9 vs LNE 7, 5-8,5 , ONE 7, 6-12). Similar difference was shown in times until first stool passage. Patients operated laparoscopically had shorter times until normal food tolerance (Median, IQR; days): LE 3, 2-3, LNE 3, 2-4 than patients who underwent open surgery (OE 3, 3-4, ONE 4, 3-5). In addition, laparoscopically operated patients had lower overall morbidity (P<0.001). Incidence of unplanned operations and hospital readmissions did not differ significantly among groups.

Conclusion: Well-performed ERAS protocol can improve length of hospital stay and time until first stool passage in both open and laparoscopic types of operation. Optimal combination for colorectal resection is laparoscopic surgery with ERAS protocol. If open surgery is done, it should be preferably applied with ERAS protocol as well.

Keywords:

colorectal cancer, colorectal surgery, ERAS protocol

INTRODUCTION

ERAS (Enhanced Recovery After Surgery) is a multimodal, evidence based protocol of perioperative care. The goal of this modality, introduced by Kehlet in early 1990´s, is to standardize perioperative care and combine many scientifically proven steps in order to reduce the length of perioperative stay, surgical stress and number of complications. This way, patient is back in the centre of care rather than individual decisions of a surgeon and other medical staff [1-27]. ERAS protocol in University Hospital Center Split was first introduced in October 2016.

MATHERIALS AND METHODS

Study population and exclusion criteria.

We collected data from a longitudinal, prospective cohort of patients at Department of surgery in University Hospital Center Split in two years period following introduction of ERAS protocol (October 2016. - October 2018.). Only the patients that fulfilled following criteria were included: 1) Pathohistological evidence of malignant disease before hospital admission, 2) Elective colorectal resection surgery was indicated as curative therapy of choice, 3) Patients were part of ERAS protocol with predefined discharge criteria.

The exclusion criteria were following: 1) History of major abdominal operation, 2) History of any operation 30 days before colorectal resection, 3) ASA score >3, 4) Disseminated (metastatic) disease. Every patient was followed three months after day of hospital discharge. All patients were informed about ERAS protocol and agreed to participate in study.

Group formation

Group formation was based on compliance of ERAS protocol (60% or more steps applied were considered as well performed ERAS protocol) and type of operation (open or laparoscopic). Consequently, we divided patients into four groups: Open/non-ERAS (open operations and poorly performed ERAS) or „ONE", Open/

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ERAS (open operations and well performed ERAS) or „OE", Laparoscopy/non-ERAS (laparoscopic operations and poorly performed ERAS) or „LNE" and Laparoscopy/ERAS (laparoscopic operations and well performed ERAS) or „LE" (Figure 1).

Baseline characteristics

For every patient basic set of information (age, gender, BMI, body fat percentage, haemoglobin levels, ASA stage, consumption of tobacco and alcohol, comorbidities), diagnostic workup (cancer site and staging,) and operation data (type, duration, presence of stoma, amount of fluid given during operation) were collected.

ERAS steps

We followed application of 20 ERAS steps in total (8 preoprative, 4 intraoperative and 8 postoperative). ERAS step was considered applied only when it fulfilled strict criteria. All criteria were based on prominent clinical studies and recommendations as shown in Table 1.

Outcomes

Primary outcomes were length of postoperative hospital stay, time of return of bowel function (day of first stool passage and introduction of normal nutrition). All patients were discharged when they complied with these hospital discharge criteria: (1) tolerance of oral intake of fluids and food, (2) passage of first stool, (3) absence of nausea, (4) pain that can be controlled with oral analgesics and (5) patient´s consent.

Secondary outcomes were incidence of major and minor complications, unplanned reoperations, readmissions and in-hospital mortality.

Statistical analysis

For continuous data we tested normality of distribution (Shapiro-Wilk test) and homogeneity of variance (Levene´s test). We expressed our data as arithmetic mean ± standard deviation, median with interquartile range, minimum and maximum value where appropriate. In inferential statistics we used Analysis of Variance (ANOVA-test) and Kruskal Wallis with Mann-Whitney U test. For categorical data we used Chi-squared ($\chi 2$) test. Significance level was set at 5% ($P<0.05$). Statistics were made using software IBM SPSS Statistics 25.0., Chicago, Illinois, USA.

RESULTS

Baseline characteristics

We compared baseline characteristics among groups as seen in Table 2. There were statistically significant differences in haemoglobin and body fat percentage values. In surgical aspects there were significant differences in types of resection and presence of stoma between open and laparoscopic groups. When we compared types of surgical resections and stoma presence in open and laparoscopic groups separately they did not differ significantly.

ERAS steps compliance

We noted 20 ERAS steps in total and compared number of applied steps in each group. In „Open ERAS" group 13,26±1,35 out of the 20 steps and in „Laparoscopic ERAS" group 14,15±1,57 per person was applied. In groups with poorly performed ERAS some of the steps were also applied. In „Open non-ERAS" group 10,04± 1,11 and in „Laparoscopic non-ERAS" 8,79±1,85 was applied successfully (Table 3).

Primary outcomes

Length of postoperative hospital stay was significantly shorter in patients operated laparoscopically with well performed ERAS (LE) comparing to other three groups. On the other hand, patients that underwent open surgery with poorly performed ERAS (ONE) had statistically significant longer postoperative stay than patients in other three groups. Group Open/ERAS (OE) had shorter postoperative stay than Laparoscopy/non-ERAS (LNE), but without statistical significance. Similarly, group Laparoscopy/ERAS (LE) had significantly shorter time until first stool passage than other three groups. Group Open /ERAS (OE) had shorter time until first stool passage than both groups with poorly performed ERAS, but only difference in comparison to group Open/non-ERAS (ONE) was statistically significant. When analysing time until normal food introduction, patients operated laparoscopically (groups LE and LNE) tolerated this kind of food earlier. However, the difference between Laparoscopy/non-ERAS (LNE) and Open/ERAS (OE) was not statistically significant. Open/non-ERAS (ONE) group had significantly longer time until normal food introduction in comparison with other three groups (Table 4).

Secondary outcomes

There was significant difference in overall morbidity and major complications when comparing laparoscopic (LNE and LE) and open (OE and ONE) groups ($P<0,001$) as illustrated in Table 5. Open/ERAS (OE) group showed lower overall morbidity and less major complications when comparing to Open/non-ERAS (ONE) group, but without statistical significance. Similarly, „Laparoscopic ERAS" showed better overall morbidity with less major complications than „Laparoscopic non-ERAS group", but also without statistical significance.

DISCUSSION

Our study showed that combination of laparoscopic surgery and ERAS protocol (group LE) enables significantly faster postoperative recovery (length of postoperative hospital stay, time until first stool passage and induction of normal nutrition) than all three other combinations. Group Open/ERAS (OE) had significantly faster postoperative recovery than group Open/non-ERAS (ONE) and very similar results as group Laparoscopy/non-ERAS (LNE). More precisely, group OE had shorter length of postoperative stay and time until

first stool passage than group LNE, but without statistical significance. Patients in group Open/non-ERAS (ONE) had significantly prolonged postoperative recovery in comparison to all three other groups. Morbidity was significantly lower in patients operated laparoscopically (groups LE and LNE). There was no statistically significant difference in unplanned reoperations, readmissions and in-hospital mortality among groups. In this category ERAS protocol had smaller influence than in postoperative recovery. However, group Open/ERAS (OE) had lower overall morbidity and lower incidence of unplanned reoperations and readmissions in comparison to group Open/non-ERAS (ONE), but without statistical significance. Length of postoperative hospital stay as well as overall morbidity is similar to results given in prominent meta-analyses [27-29]. However, incidence of complications shown in our study is mostly higher than results published in meta-analyses. It can be explained by non-existing upper age limit in our study. Length of postoperative hospital stay was almost equal to prominent LAFA-study by Vlug et al. [30]. Although, when comparing times until first stool passage and normal food tolerance, LAFA study showed much better results. This could be partially explained with fact that we included patients with planned stoma formation (colostoma and ileostoma). Designs of ERAS studies differ dramatically. Therefore, comparison among papers is extremely difficult [27]. Some studies only include operations of the colon without rectum [31-36] or only laparoscopically operated patients [31, 37-44] Also, number and type of ERAS steps applied is different in almost every study [27]. Systematic review of Messenger et al. precisely described mentioned variability by publishing range of 6 to 21 steps applied among 34 analyzed studies [45]. Moreover, it is rarely seen that authors describe exact criteria that they used when deciding which ERAS step was applied. We explained every ERAS step individually with adequate reference, hoping that future studies will also provide this kind of information. Strength of this study is thoroughly described compliance for every ERAS step. This set of information could help discovering which ERAS step is more important in the future. Studies rarely report this result which was mentioned in systematic review by Messenger et al. They noted that only one of all analyzed RCTs reported this kind of data [45]. We conducted audit in accordance to systematic reviews and meta-analysis who constantly pointed out weaknesses of analyzed studies [27-29, 45]. In order to enable easier comparison in the future we also followed audit guidelines of ERAS® society [46]. Limitation of this study is it´s sample size. Moreover, groups are unbalanced in number and in some basic characteristics. Group Laparoscopy/ERAS (LE) had significantly higher level of preoperative haemoglobin and body fat percentage than group Open/non-ERAS (ONE). Body fat percentage is easily explained with higher percentage of females but higher haemoglobin levels remain unexplained. Some types of operations were more common in laparoscopic and some in open type of surgery. It is very hard to say which operation is more severe so the influence of this kind of data is unknown. Except types of operation, presence of stoma was also significantly different when comparing laparoscopic and open surgery. Stomas were more common in laparoscopic surgery. Presence of stoma is linked with worse outcomes, especially regarding stool passage, which could explain why patients in group OE needed less time to achieve this goal than patients in group LNE [47]. Unbalanced group sizes and usage of nonparametric tests in our inferential statistics have lowered the strength and therefore significance in given results could be exaggerated. On the other hand, still relatively low overall compliance and small difference in compliances between ERAS and non-ERAS groups could underestimate it [48,49]. In our study, all patients were part of ERAS protocol, planned to achieve as many steps as possible. That explains smaller difference in compliance between well and poorly performed ERAS groups. We can say that type of operation and ERAS compliance are two most important predictors of surgical outcomes. At the end, we can state that ERAS protocol is a type of perioperative care that will lead to better outcomes in almost every parameter analyzed. Some of the results did not show statistical significance, but we should consider that even unsignificant differences can provide substantial clinical benefit.

CONCLUSION

Our study showed that well performed ERAS protocol can reduce length of postoperative hospital stay and time until first stool passage. The best combination to use when performing colorectal surgery is laparoscopy with well performed ERAS protocol. ERAS is also recommended type of perioperative care in combination with open surgery.

CONFLICT OF INTEREST:

The authors declare that there is no conflict of interest.

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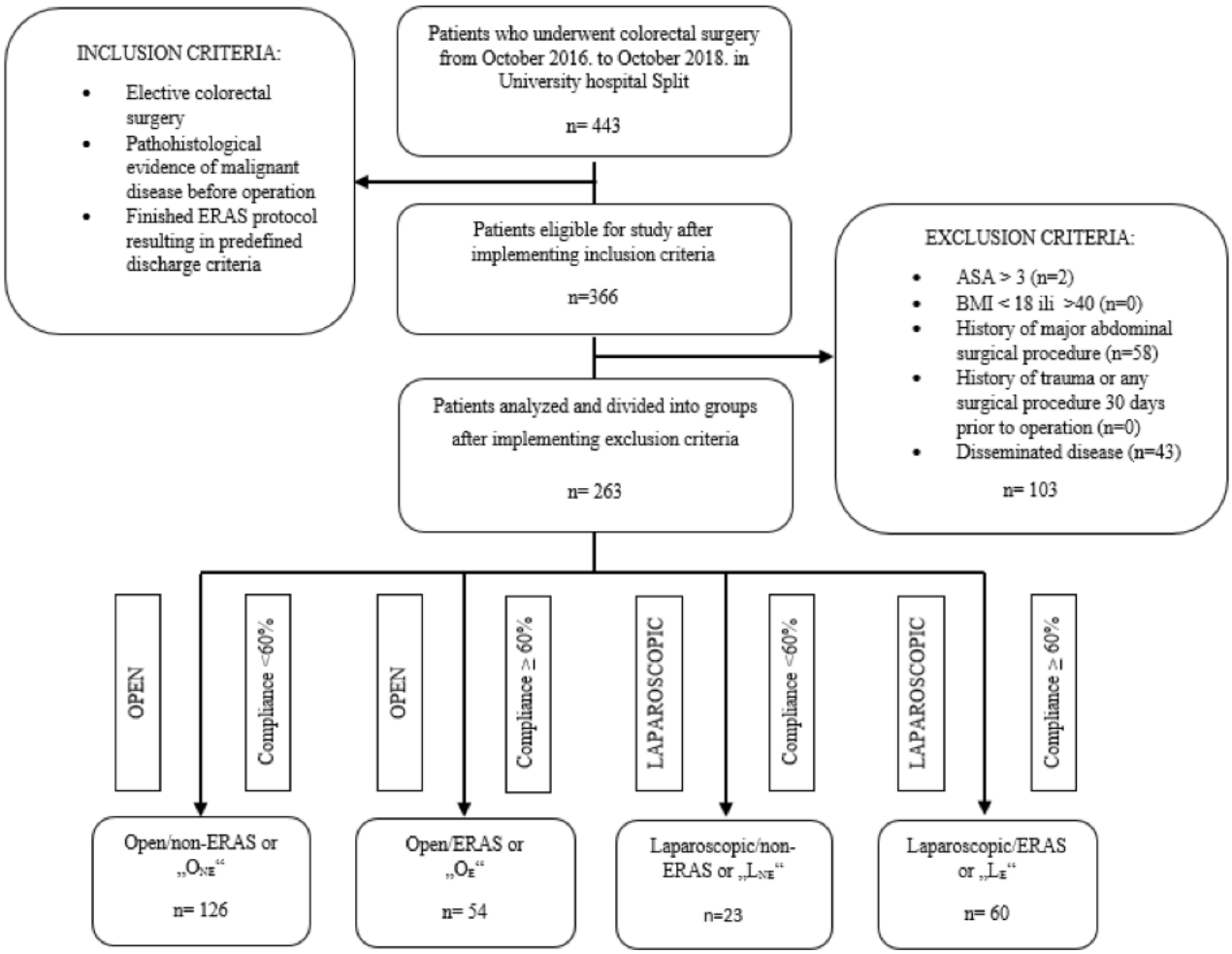

Figure 1. Study flow

ERAS STEP	EVIDENCE BASED CRITERIA
1. Preoperative counselling	Patient is familiar with ERAS steps, expectations, type of operation and risks (3,4).
2. Preoperative nutritional screening	Calculating NRS 2002 score at least two weeks before day of operation. Patients with 3 points or more are scheduled for nutritional intervention with high-protein drinks (5).
3. Bowel preparation	For operations with total mesorectal excision polyethylene glycol is used afternoon before operation and enema morning before operation. For other operations no bowel preparation is used (6,7).
4. Preoperative fasting treatment	Patients are allowed to take solid food up to 6 hours and clear fluids up to 2 hours before induction of anaesthesia (8,9).
5. Preoperative carbohydrate treatment	Patients should ingest >100g of carbohydrates afternoon before and >50g morning before the operation (10).
6. Antibiotic prophylaxis	First generation cephalosporin (cefazolin) and metronidazole are given intravenously 60 minutes before skin incision (11,12).
7. Antithrombotic prophylaxis	Usage of compression socks or intermittent pneumatic compression with low-molecular heparin (13,14).
8. Preanesthetic medication	Routine usage of enteral sedative drugs is avoided. If needed, usage of 5 mg diazepam intravenously is allowed (15).
9. Anaesthetic protocol	Combination of general and regional anesthetics with rapid awakening is used (15).
10. Prevention of intraoperative hypothermia	Patients are warmed under a blanket during procedure with temperature maintained at 34°C (16).
11. Intraoperative fluid management	1-4 mL/kg/h of crystalloid fluid is used for open and laparoscopic surgery (17).
12. Application of nasogastric tube	Routine application as ileus and vomiting prevention should be avoided (18-20).
13. Abdominal drainage	All abdominal drains are removed in first three days after operation.
14. PONVa prophylaxis	For patients with low/mild risk dexamethasone (4-8 mg) with or without metoclopramide (10 mg) is administered. For patients with high risk ondansetron/granisetron is administered (22,23).
15. Postoperative analgesia	Avoidance of opioid analgesics and usage of NSAIDs, paracetamole, metamizole and epidural analgesia (15, 22).
16. Signs of ileus check	Signs of ileus are checked every day.
17. Intake of fluid on the day of operation	Patient should drink (not more than 800 mL) of fluid afternoon after the procedure (23).
18. Early ambulation	Patients are mobilized to sit at the end of the bed or get up and walk short distances (15, 24).
19. High protein ONSb intake	Patients need to take high-protein drinks first four days after the operation (15,25).
20. Urinary drainage management	Urinary catheter is removed in first two days after the operation (26).

[a] *Prophylaxis Of Nausea and Vomiting*
[b] *Oral Nutrition Supplement*

Table 1. ERAS steps and criteria

	Laparoscopy and ERAS (L_E) N=60	Open and ERAS (O_E) N=54	Laparoscopy non-ERAS (L_{NE}) N=23	Open non-ERAS (O_{NE}) N=126	P
Age (AM±SD[a], year)	66,63	67,23	68,17	68,86	0,451*
Gender (M/F)	31/29	35/19	14/9	91/35	0,051†
BMI[b] (AM±SD, kg/m^2)	26,89	25,96	27,05	26,55	0,584*
Body fat (AM±SD, %)	26,77	24,81	26,35	23,43	0,030*
Preoperative haemoglobin (AM±SD, g/L)	131,25	121,74	130,35	125,98	0,037*
Duration of procedure median (IQR[c], min)	135 (117,5-165)	100 (70-130)	140 (120-155)	100 (80-122,5)	<0,001‡
Fluids given during procedure median (IQR, mL)	1000 (800-1250)	1000 (800-1300)	1050 (1000-1600)	1100 (1000-1500)	0,110‡
ASA[d], N (%)					0,576†
I	2 (3,33)	2 (3,7)	0 (0)	3 (2,38)	
II	44 (73,3)	34 (62,96)	18 (78,26)	79 (62,70)	
III	14 (23,33)	18 (33,33)	5 (21,74)	44 (34,92)	
Comorbidities[e]					
Total, N	54	42	22	118	0,734†
Patients with one or more comorbidities, N (%)	39 (65,00)	32 (59,26)	17 (73,91)	82 (65,08)	0,668†
Tobacco, N (%)	8 (14,55)	6 (11,32)	4 (19,05)	24 (19,51)	0,535†
Alcohol, N (%)	1 (1,82)	2 (4,08)	1 (4,55)	5 (4,07)	0,862†
Type of colectomy, N (%)					
Ileocecal resection	2 (3,33)	1 (1,85)	0 (0)	1 (0,79)	
Right hemicolectomy	0 (0)	24 (44,44)	0 (0)	45 (35,71)	
Transversotomy	0 (0)	1 (1,85)	0 (0)	2 (1,59)	
Left hemicolectomy	0 (0)	2 (3,70)	0 (0)	8 (6,35)	
Sigmoidectomy	4 (6,67)	7 (12,96)	1 (4,35)	13 (10,32)	
Hartmann resection	5 (8,33)	8 (13,33)	2 (3,33)	4 (3,17)	
Lower anterior resection	38 (63,33)	9 (16,67)	15 (65,22)	41 (32,54)	
Quenn-Milles (Abdominoperineal resection)	9 (15,00)	2 (3,70)	5 (21,74)	9 (7,14)	
En bloc resections	0 (0)	0 (0)	0 (0)	1 (0,79)	
Total/subtotal colectomy	0 (0)	0 (0)	0 (0)	1 (0,79)	
Other[f]	2 (3,33)	0 (0)	0 (0)	1 (0,79)	
Presence of stoma, N (%)	23 (38,33)	11 (20,37)	7 (30,43)	20 (15,87)	0,006†
T-Stage, N (%)					0,284†
Tis	0 (0)	0 (0)	1 (5,26)	0 (0)	
T_1	7 (14,29)	2 (3,92)	2 (10,53)	6 (5,13)	
T_2	13 (26,53)	10 (19,61)	3 (15,79)	22 (18,80)	
T_3	22 (44,90)	28 (54,90)	9 (47,37)	72 (61,54)	
T_4	7 (14,29)	11 (21,57)	4 (21,05)	17 (14,53)	
N-Stage, N (%)					0,613†
N_0	33 (67,35)	32 (62,75)	13 (68,42)	83 (70,94)	
N_1	9 (18,37)	14 (27,45)	5 (26,32)	27 (23,08)	
N_2	7 (14,29)	5 (9,80)	1 (5,26)	7 (5,98)	

[a] *Arithmetic Mean±Standard Deviation*
[b] *Body Mass Index*
[c] *Interquartile range*
[d] *American Society of Anesthesiologists*
[e] *Hypertension, diabetes type II, COPD, liver cirrhosis, heart failure, history of myocardial infarction and/or CVI, other neurological diseases e.g. epilepsy, mb. Parkinson*
[f] *Transanal total mesorectal excision (TaTME), segmental resections*
** Analysis of variance (ANOVA-test)*
† Chi-squared test
‡ Kruskal-Wallis test

Table 2. Baseline characteristics and surgical aspects per group

ERAS STEPS, N (%)	Laparoscopy and ERAS (L_E) N=60	Open and ERAS (O_E) N=54	Laparoscopy non-ERAS (L_{NE}) N=23	Open non-ERAS (O_{NE}) N=126
Preoperative counselling	49 (81,67)	36 (66,67)	11 (47,83)	45 (35,71)
Preoperative nutritional screening and intervention	29 (48,33)	21 (38,89)	1 (4,35)	17 (13,49)
Bowel preparation	55 (91,67)	45 (83,33)	11 (47,83)	58 (46,03)
Preoperative fasting treatment	58 (96,67)	52 (96,30)	13 (56,52)	62 (49,21)
Preoperative carbohydrate treatment	47 (78,33)	47 (87,04)	12 (53,17)	64 (50,79)
Antibiotic prophylaxis	60 (100)	54 (100)	23 (100)	124 (98,41)
Antithrombotic prophylaxis	59 (98,33)	54 (100)	20 (86,96)	113 (89,68)
Preanesthetic medication	5 (8,33)	3 (5,55)	1 (4,35)	11 (8,73)
Anaesthetic protocol	35 (58,33)	39 (72,22)	12 (53.17)	62 (49,21)
Prevention of intraoperative hypothermia	23 (38,33)	19 (35,19)	4 (17,39)	25 (19,84)
Intraoperative fluid management	0 (0)	0 (0)	0 (0)	0 (0)
Application of nasogastric tube	59 (98,33)	53 (98,15)	22 (95,65)	113 (89,68)
Abdominal drainage	39 (65,00)	26 (48,15)	4 (17,39)	9 (7,14)
PONV prophylaxis	28 (46,67)	15 (27,78)	4 (17,39)	13 (10,32)
Postoperative analgesia	49 (81,67)	47 (87,04)	16 (69,57)	64 (50,79)
Signs of ileus check	59 (98,33)	54 (100)	23 (100)	125 (99,21)
Intake of fluid on the day of operation	37 (61,67)	31 (57,41)	10 (43,48)	49 (38,89)
Early ambulation	53 (88,33)	34 (62,96)	13 (56,52)	41 (32,54)
High protein ONS intake	54 (90,00)	49 (90,74)	19 (82,61)	91 (72,22)
Urinary drainage management	51 (85,00)	37 (68,52)	12 (52,17)	21 (16,67)
TOTAL, AM±SD	14,15±1,57	13,26±1,35	10,04±1,11	8,79±1,85

Table 3. Protocol compliance

Length of postoperative hospital stay, days				L_E	O_E	L_{NE}	O_{NE}
GROUP	Median (IQR)[a]	Min[b]	Max[c]	P<0,001*			
Laparoscopy and ERAS (L_E) N=60	5 (4-6)	2	23	/	0,001	0,001	<0,001
Open and ERAS (O_E) N=54	6 (5-9)	3	16	0,001	/	0,374	<0,001
Laparoscopy non-ERAS (L_{NE}) N=23	7 (5-8,5)	5	19	0,001	0,374	/	0,034
Open non-ERAS (O_{NE}) N=126	7 (6-12)	4	25	<0,001	<0,001	0,034	/
Day of first stool passage				L_E	O_E	L_{NE}	O_{NE}
GROUP	Median (IQR)	Min	Max	P<0.001*			
Laparoscopy and ERAS (L_E) N=60	3 (2-4)	1	10	/	0,004	<0,001	<0,001
Open and ERAS (O_E) N=54	4 (3-5)	1	9	0,004	/	0,077	0,011
Laparoscopy non-ERAS (L_{NE}) N=23	5 (4-6,5)	2	8	<0,001	0,077	/	0,269
Open non-ERAS (O_{NE}) N=126	4 (3-5)	1	14	<0,001	0,011	0,269	/
Day of normal food introduction				L_E	O_E	L_{NE}	O_{NE}
GROUP	Median (IQR)	Min	Max	P<0,001*			
Laparoscopy and ERAS (L_E) N=60	3 (2-3)	1	5	/	<0,001	0,070	<0,001
Open and ERAS (O_E) N=54	3 (3-4)	2	7	<0,001	/	0,077	<0,001
Laparoscopy non-ERAS (L_{NE}) N=23	3 (2-4)	1	5	0,070	0,077	/	<0,001
Open non-ERAS (O_{NE}) N=126	4 (3-5)	2	14	<0,001	<0,001	<0,001	/

[a] *Median (Interquartile range)*

[b] *Minimal value*

[c] *Maximal value*

** Kruskal-Wallis test. Groups were individually compared by Mann-Whitney U test*

Table 4. Primary outcomes

	Laparoscopy and ERAS (LE) N=60	Open and ERAS (OE) N=54	Laparoscopy non-ERAS (LNE) N=23	Open non-ERAS (ONE) N=126	P*
Overall morbidity < 30 days, N (%)	20 (33,33)	38 (70,37)	9 (39,13)	92 (73,02)	<0,001
Patients with one or more major complication, N (%)	4 (6,67)	15 (27,78)	3 (13,04)	51 (40,48)	<0,001
Total no. of major complications	5	15	4	59	
Pathological ileus	3	15	3	45	
Anastomotic leakage	1	0	0	5	
Anastomotic fistula	1	0	0	2	
Wound dehiscence	0	0	0	2	
Peritonitis	0	0	0	1	
Intraabdominal abscess	0	0	0	2	
Haemoperitoneum	0	0	0	1	
Othera	0	0	1	1	
Patients with one or more minor complication, N (%)	16 (26,67)	23 (42,59)	6 (26,09)	41 (32,54)	0,278
Total no. of minor complications	17	28	6	46	
Wound infection	6	13	3	23	
Wound haematoma	0	1	0	1	
Urinary tract infection	2	4	0	5	
Pneumonia	0	1	0	1	
Nausea and vomiting	7	6	2	13	
Otherb	2	3	1	3	
Unplanned reoperations, N (%)	3 (5,00)	2 (3,70)	1 (4,35)	13 (10,32)	0,375
Total no. of readmissions, N (%)	3 (5,00)	1 (1,85)	0 (0)	6 (4,76)	0,568
< 1 month	3	1	0	5	
< 3 months	0	0	0	1	
In-hospital mortality, N (%)	0 (0)	0 (0)	0 (0)	0 (0)	

[a] *Necrosis of skin around stoma*
[b] *Blood in stool*
* *Chi-squared test*

Table 5. Secondary outcomes

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TREATMENT OF PRIMARY SPONTANEOUS PNEUMOTHORAX IN PEDIATRIC PATIENTS: 15-YEAR EXPERIENCE AT A SINGLE-INSTITUTION

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ABSTRACT

Background: Primary spontaneous pneumothorax (PSP) is a relatively uncommon condition in children. Due to the lack of pediatric-specific guidelines the treatment strategy varies among different centers. This study demonstrates a single-institution experience in the treatment of primary spontaneous pneumothorax in pediatric patients.

Materials and methods: Retrospective review of 49 patients with the diagnosis of primary spontaneous pneumothorax between 2003 and 2018 who were treated conservatively or invasively at the surgical department of Children's Hospital Zagreb.

Results: During the period of 15 years there were 49 patients noted with spontaneous pneumothorax at the surgical department of Children's Hospital Zagreb. The patient age ranged from 11 to 18 years (mean 15.28). 36 patients were male and 13 female, with a male to female ratio of 2.77:1. Pneumothorax occurred on the left side in 31 patients, on the right side in 15 patients, and on both sides in 3 patients. Clinical presentation in all patients was sudden, powerful chest pain. Other symptoms include dyspnea, cough. In all patients, a plain x-ray was made, and later the vast majority of patients underwent computed tomography (CT) scan. The CT scans detected 13 cases of apical bullae, 2 large bullae, 1 bulla in 6. segment and 1 parenchymal inflammation. Eight patients with stable clinical presentation and small pneumothorax underwent hospital observation. Eighteen patients were successfully managed with chest tube drainage without recurrence. Video-assisted thoracoscopic surgery (VATS) was performed on 19 patients with only two recurrences. Open thoracotomy was performed on 4 patients.

Conclusion: Due to the variation of diagnostic and therapeutic approaches from different centers, the creation of guidelines and standardized practice for the pediatric patient is necessary.

Keywords:

Primary spontaneous pneumothorax, Pediatric, Video-assisted thoracoscopic surgery

INTRODUCTION

Pneumothorax is defined as a collection of air in the pleural space with secondary lung collapse [1]. Pneumothorax is divided into two main groups: spontaneous and nonspontaneous. Spontaneous pneumothorax is classified into primary spontaneous pneumothorax (PSP), which occurs in healthy patients without underlying lung disease, and secondary spontaneous pneumothorax (SSP) which occurs with preexisting lung diseases such as cystic fibrosis, asthma, congenital cystic adenomatoid malformation, connective tissue disease, malignancies, and other disorders [2,3]. Nonspontaneous pneumothorax may be iatrogenic or caused by trauma [4].

Primary spontaneous pneumothorax is a relatively uncommon condition in the pediatric population, with an incidence of 3.4 per 100,000 children [4]. The disease most commonly occurs in tall, thin adolescent males between 15-17 age, with a male to female ratio of 4:1 [5,6]. There are no large studies showing a combined ratio PSP to SSP [5]. Smaller studies on 162 patients suggest that PSP is more common than SSP, with the presentation of PSP in 120 and SSP in 42 patients [7]. Also in the same studies small PSP (66%) were more common than large ones (34%) [7].

The size of the pneumothorax is divided into small and large pneumothorax. According to the British Thoracic Society (BTS) small pneumothorax is defined as the presence of a visible rim less than 2 cm between the lung margin and the chest wall (at the level of the hilum), where in the large pneumothorax rim is more than 2 cm [8].

The first line in the diagnostic approach is chest X-ray, but the use of computed tomography (CT) scan in the initial presentation of PSP is controversial. Some centers perform routine CT scan in patients with PSP to detect abnormal lung findings, while in other CT is performed in recurrent PSP or in a patient with an atypical presentation [2,3,6,9,10,11].

The management strategy varies between different centers due to the lack of evidence-based guidelines [3,6,9,12]. Treatment of the spontaneous pneumothorax varies from a conservative observation approach to the tube thoracostomy, video-assisted thoracoscopic surgery (VATS) and thoracotomy. Most often the first line of treatment in

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small pneumothorax is intensive observation while in large pneumothorax the tube thoracostomy with observation is indicated [2,3,8,6,9]. The nonoperative approach was associated with higher recurrency rate > 40% [2,6,9,13]. The first method of choice in surgical intervention is VATS [2,3,8]. Video-assisted thoracoscopic surgery has proven to be safe and effective compared to open thoracotomy, with decreased postoperative pain and morbidity. Low recurrence rates (10%–13%) were conceted to VATS [2,6,9]. Open thoracotomy remains a method of choice in the case of giants bullas and blebs that are not positioned in the apical part of the lung [2,14].

MATERIAL AND METHODS

Paper is a retrospective review of all patients (49 patients) with a diagnosis of spontaneous pneumothorax between the years 2003. and 2018. who were treated conservatively or invasively at the surgical department of Children's Hospital Zagreb.

The data was analyzed using descriptive statistics to demonstrate demographic and baseline patient characteristics (age, sex), initial clinical presentation, hospital course, procedures, imaging, outcomes, length of hospitalization and follow up period. Results are expressed as the means, medians, ranges, percentages and proportions.

All patients under 18 years old with confirmed diagnoses of PSP between 2003-2018 were included. Patients with secondary spontaneous and nonspontaneous pneumothorax were excluded. PSP was classified as small if the visible rim was less then 2cm between the lung margin and the chest wall on the x-ray scan, where the large PSP is more than 2cm [8]. Depending on the side of PSP we distinguish left, right and simultaneous bilateral in which bilateral PSP includes patients with PSP on one side and second presentation on the contralateral side, and patients with a simultaneous presentation on both sides. Recurrent PSP patients are described as the occurrence of new ipsilateral PSP. Indication for CT scan was confirmed PSP with x-ray imaging [2,7,12]. CT scan was noted as positive if CT scan has abnormal findings.

The patient was described as undergoing hospital observation if they did not undergo any procedure or operation, and only observation of vital signs, oxygen saturation, and control plan x-ray was done. Hospital observation was indicated in patients with small PSP and minimal symptoms [2,8,9].

Patients were described as undergoing thoracostomy if they underwent thoracostomy tube placement, aspiration, and observation without any additional intervention. Tube thoracostomy was indicated in unstable symptomatic patients and in large PSP [2,8,9]. The size of the tube and location of thoracostomy was choosed depending on the age and size of the patient. In patients younger than 14 years old small chest drain (No.14) was inserted, while in 14 years and older patients larger tube were used (No.20) [2]. In all patients, thoracostomy was done with low-pressure suction with pressures between -10 and -20cm H2O [2]. The most common placement of the drain was between 4th and 6th intercostal space [2, 8,9].

Patients were described as undergoing operative management if they underwent VATS or open thoracotomy. VATS were indicated in the case of a persistent air leak (rang 5–6 days) after chest tube drainage, recurrence and in all patients with apical bullae [2, 8, 9]. VATS was performed with a stapler wedge resection of the apical part of the lung with an endostapler device (articulating endoscopic linear cutter with 45mm staple line) and mechanical pleurodesis or pleurectomy [2]. In children with giants bullas and blebs that are not positioned in the apical part of the lung, open thoracotomy with resection of bullas was indicated. The incision was adjusted to the localization and size of the lesion. Upper posterolateral open thoracotomy was most commonly applied [2,14].

RESULTS

There were 49 patients with spontaneous pneumothorax, and recurrence was noted in 14 patients. The patient age ranged from 11 to 18 years (mean 15.28) with the majority between 15 and 17 years (81.6%). Thirty-six patients were male and 13 female with a male to female ratio of 2.77:1. Pneumothorax occurred on the left side in 31 patients, on the right side in 15 cases and it was bilateral in 3 patients. The follow-up period was from 4 months to 5 years (median 3.6 years). Length of hospital stay ranged from 4 - 29 days with a median of 11 days. Clinical presentation in all patients was sudden, powerful chest pain. Other symptoms included dyspnea and cough. In all patients, we made a plain X-ray, followed by CT scan. Chest CT scans were indicated in all patients, but were performed on 35 patients (71%). In 13 cases CT has shown apical bulla, 2 large bullae in the lower part of the lung, 1 bullae in 6. segment, and 1 parenchymal inflammation.

Eight patients with stable clinical presentation and small pneumothorax underwent hospital observation. Eighteen patients with unstable symptomatic presentation and with large PSP underwent thoracostomy. The mean duration of the chest drainage was 5 days. In 18 patients with tube drainage, the resolution of the pneumothorax and air leak with re-expansion of the lung were followed (Figure1.). VATS was performed in 19 patients. Three patients had pneumothorax on both sides. One case of simultaneous presentation of small PSP on both sides underwent hospital observation. Two had pneumothorax on both sides, but not simultaneously, and 2 separate VATS procedures were done in each case (Table 1).

We had only 2 recurrences after VATS, in one case with small pneumothorax recurrence, the intensive

observation was indicated and in other case open thoracotomy was indicated. There was no complication related to the VATS approach.

Four patients underwent open thoracotomy, from which one after VATS recurrence and other 3 had pneumothorax with lung collapse. After a CT scan investigation in patient with lung collaps, lung bulla was found. A fourteen-year-old boy had 1,5cm bulla in 6. lung segment, a 17-year-old boy had a bulla of 9 cm diameter, and a 12-year-old girl had a bulla of 5 cm diameter. In these children, VATS wasn't performed because the location of pneumothorax was not on the apical part of the lung and we considered that bullas were too large for endostapler wedge resection. There was no complication related to the open thoracotomy approach.

DISCUSSION

Spontaneous pneumothorax is a very serious medical condition, and intensive care with hospitalization is necessary for all patients [2]. The generally accepted cause of PSP is the rupture of subpleural bulla [10,7]. Some studies suggest that bullas are cause of PSP in 60% [7]. The demographic and baseline characteristics of PSP patients were similar to other studies with characteristic male to female prevalence and with similar age interval presentations with the majority between 15-17 years [5].

Due to the lack of pediatric-specific guidelines, the diagnostic approach and treatment strategy varies from different centers. The initial treatment of PSP is based on clinical presentation and the degree of lung collapse on imaging. Management recommendations of PSP in the pediatric population are based on adult British Thoracic Society (BTS) guidelines [7].

Indication for CT scan in the first episode remained controversial. While some studies suggest CT scan at initial presentation [2,7,12,13], others are reserving it for atypical presentation and recurrences [10]. In our center, a CT scan was indicated at the initial presentation of PSP with aim to identify the presence or absence of abnormal pulmonary findings [2,7]. Studies have suggested that only a few pediatric cases of PSP were without identifiable pathology and that in pediatric patients there is a higher rate of bulla than it is in adults [7,15]. Early detection of the bullae or blebs can improve the operative management of PSP and eventually reduce the recurrence rate [7]. Other studies detected that CT has low sensitivity (36%) for the detection of blebs and therefore limited diagnostic value, that why CT scan is an option only in recurrent and atypical patients with the aim to avoid potentially unnecessary ionizing radiation [10,12].

The majority of surgeons treat the first episode of PSP nonoperatively. Patient with small pneumothorax and stabile clinical presentation require only an observation and short-interval follow-up, while those who are unstable or those with a large pneumothorax requires chest tube drainage with suction and observation [8,9,2]. The nonoperative approach was associated with higher recurrency rate > 40% [6, 9].

Regardless of the imaging, surgeons most often relay on clinical presentation, post drainage persistent air leak, incomplete lung re-expansion, and recurrence rate as the main indication for surgical intervention. Majority of surgeons recommend VATS intervention in persistent air leak in initial hospitalization, but number of days before recommending operation varies from 3 to 5 days. Also VATS is mainly indicated at the first episode of PSP with bullas in CT scan, and in patients with recurrence [8,9,2]. Nowadays surgical management of PSP is apical wedge resection of blebs/bullae and pleurodesis or pleurectomy typically performed using VATS. Video-assisted thoracoscopic surgery has been a safe and effective way of treating PSP, it has many advantages over open thoracotomy [8,9,2], and it's associated with low recurrence rates (10%–13%) [2,9]. Our studies also detected a lower recurrence rate in the VATS approach.

Open thoracotomy is performed in the case of giant bullae and bullae that are not positioned in the apical part of the lung [2]. Open thoracotomy was done in 4 patients.

In Children's Hospital Zagreb over a period of 15 years there were 49 patients with PSP. In 8 patients with stable clinical presentation and small pneumothorax, only short-interval follow-up was done. 18 patients were successfully managed with chest tube drainage without recurrence. Chest-tube suction was recommended for at least 5 days. VATS ware performed on 19 patients with only two recurrences. Open thoracotomy was done in 4 patients. VATS is the gold standard in surgical management of PSP due to its many advantages compear to open thoracotomy.

CONCLUSIONS

The use of computed tomography at the initial presentation remains controversial due to the lack of reliable results. While in management of patients with initial presentation of small stable PSP there is consensus, management of patients with large unstable PSP varies between different centers. Due to the variation of diagnostic and therapeutic approaches at the initial presentation from different centers, the creation of guidelines and standardized practice for the pediatric patient are necessary.

CONFLICT OF INTEREST:

The authors declare that there is no conflict of interest.

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FIGURES AND TABLES

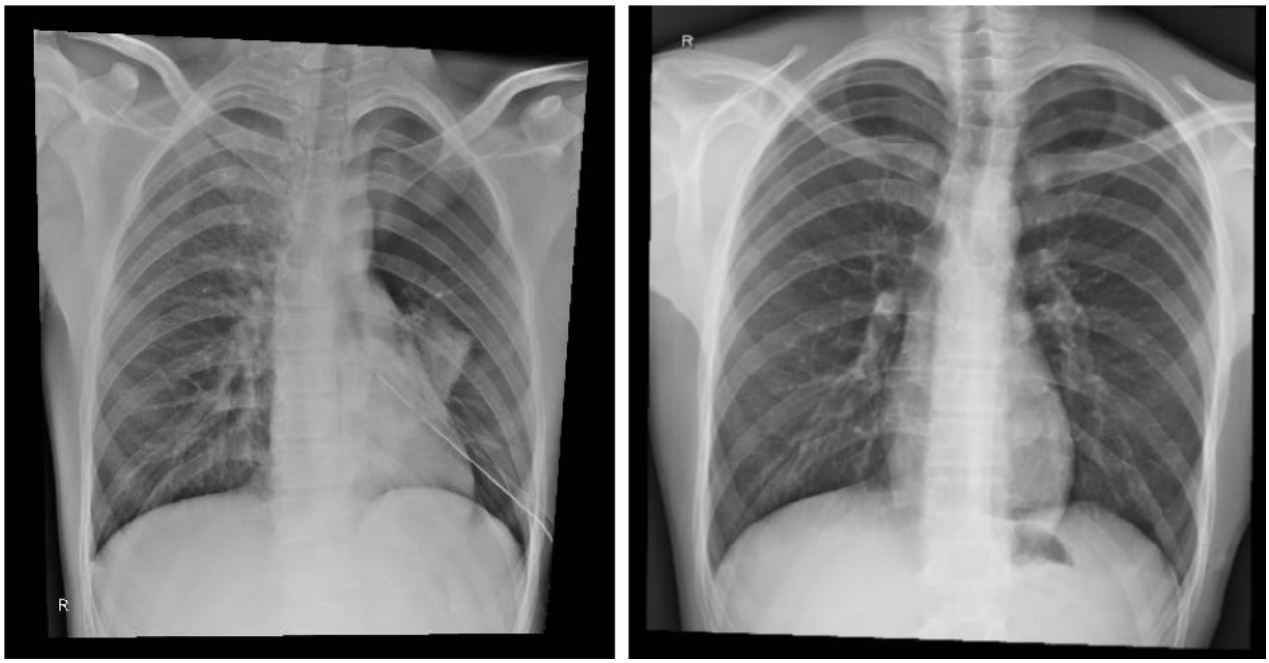

Figure 1. Left- chest tube drainage of pneumothorax. Right- post drainage resolution of the pneumothorax and air leak with re-expansion of the lung.

Table 1

Patient demographics, clinical characteristics, and management	
Patient demographics	All patients (n=49)
Gender (M/F)	36/13
Age (mean, range), years	15.28 (11-18)
Clinical characteristics	
PSP location	31 left 15 right 3 bilateral
Management	
CT scen:	35
-apical bullae	13
-large bullae	2
-not apical bullae	1
-parenhymal inflammation	1
Hospital observation	8
Tube drainage	18
VATS	19
Open thoracotomy	4

PSP, primary spontaneus pneumothorax, CT, computed tomography, VATS, ideo-assisted thoracoscopic surgery

Acta Chir Croat 2020; 17: 23-26

PELVIC INFLAMMATORY DISEASE AND SURGICAL TREATMENT OVER A TEN YEARS PERIOD; SINGLE INSTITUTION EXPERIENCE

Klara Matek[1], Oliver Vasilj[2], Lucija Brkić[1]

ABSTRACT

Background: In our study, we examined the frequency of surgical procedures due to the acute presentation of the pelvic inflammatory disease (PID) in regard to all gynecological procedures in a single institution. We also wanted to present patients' age range and type of surgical approach as well as surgical extension.

Materials and methods: This retrospective study investigated a total of 10,175 surgical procedures, out of which 103 were due to acute PID. The study included 101 patients. We obtained data from surgical procedures performed from September 2009 to September 2019.

Results: In the observed ten years period, the frequency of surgical procedures due to the acute PID was 1.01%. It was found that women were mostly older than 25 (85.15%). Laparoscopy was performed in 52.43% of cases and laparotomy in 37.86% of cases. Laparotomy incisions were transverse in 26.67% of cases and vertical in 42.22% of cases. The most radical procedures were performed in 17.48% of cases in which patients underwent a subtotal or total hysterectomy with unilateral or bilateral adnexectomy.

Conclusion: In conclusion, although rare, surgical procedures in PID are relevant because they are, according to our data, more common in patients older than 25, and surgical procedures tend to be more extensive.

Keywords:
pelvic inflammatory disease, surgical treatment, surgical approach

INTRODUCTION

Pelvic inflammatory disease pertains to an acute infection of the upper genital tract structures in women, affecting any or all of the following: uterus, fallopian tubes, ovaries and even adjacent pelvic structures. It is mostly caused by untreated sexually transmitted diseases which ascend from the lower genital tract. According to the literature available, it is typically found in sexually active women younger than 25 who do not use contraception and have multiple sex partners, whereas it rarely occurs in older women [1–3].

The term PID implicates a broad spectrum of clinical presentations, although in many women it can be asymptomatic as well. Consequently, it is difficult to diagnose, which is why the Center for disease Control and Prevention (CDC) has defined a minimum and additional criteria to help in making a more accurate diagnosis. The treatment is based on antibiotic therapy, however in some cases surgical intervention is required [3].

The actual worldwide incidence and prevalence of PID are difficult to determine due to the typically unclear clinical presentation and the lack of objective diagnostic criteria [4]. In this study, we wanted to show the frequency of surgical procedures due to the acute presentation of pelvic inflammatory disease in regard to all gynecological procedures, as well as patients' age range, type of surgical approach and surgical extension at a single institution.

MATERIALS AND METHODS

The data for this descriptive study were collected from gynecological surgical records of women treated between September 1, 2009, and September 1, 2019, at the Department of Obstetrics and Gynecology in Clinical Hospital "Sveti Duh", Croatia. In this study, we investigated a total of 10,175 surgical procedures, out of which 103 were due to acute PID. To point out, the number of patients included in this study was 101, given that one patient underwent two reoperations in one year. The data provided information on age, preoperative diagnoses, surgical approach and type of surgical incision. The collected data were imported and analyzed retrospectively in the computer software Microsoft Excel 2013.

RESULTS

In the observed ten year period in 10,175 cases there were 103 surgical procedures due to acute PID, which is 1.01% out of all surgical procedures in the Clinic. The highest percentage was in 2013, 10 surgical procedures out of 699 cases (1.43%), whereas the lowest was

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in 2009, 1 surgical procedure at 371 cases (0.27%). However, it needs to be addressed that the data from 2009 were limited to the period between September 1 and December 31. The yearly frequency of surgical procedures is presented in the Table 1.

While analyzing the age of the 101 patients it was evident that a majority of 86 patients were older than 25 (85.15%), whereas only 15 of them (14.85%) were younger than 25. The oldest patient was 63 and the youngest 18 years old. This is shown in Figure 1.

In regard to the type of surgical approach used in the treatment of PID, in 54 (52.43%) laparoscopy was performed, while 39 (37.86%) had the laparotomy. Furthermore, laparoscopic procedures were converted to laparotomy in 6 (5.83%) patients. Culdocentesis was performed in 4 (3.88%) patients.

Considering all laparotomy cases, including those where conversion was performed, we compared types of surgical incision. A total of 45 procedures were analyzed and we noticed vertical incision in 42.22% of cases, whereas 26.67% of cases had Pfannenstiel incision. There was also one case where the Pfannenstiel incision was converted to a vertical incision. Owing to the limitations of used database, in one-third of the observed cases the type of surgical incision that was used remained unknown.

The cases also have differences as regards the range of infection and affected structures. Analyzing a total of 103 cases, a right-sided infection occurred in 25.24% of cases and a left-sided infection occurred in 14.56% of cases. However, the infection was mostly bilateral, in 47.57% of cases. The affected side remained unknown in 12.62% of cases. We also noticed that appendectomy was performed in 21.36% of cases. Finally, more radical procedures were performed in 17.48% of cases in which patients underwent a subtotal or total hysterectomy with unilateral or bilateral adnexectomy. In our analysis, we included one case of total hysterectomy with unilateral ovariectomy without salpingectomy. All patients were older than the age of 40, except one (29 years old) who underwent three surgical procedures due to complications. Furthermore, in 11 out of 18 cases additional genital tract pathology, e.g. ovarian cyst, myoma or tumor, was present.

DISCUSSION

Approximately one-third of patients with a moderate or severe clinical presentation of PID responds poorly to antibiotic therapy and need to undergo a surgical procedure [5]. However, there is no specific international data available for PID incidence and prevalence worldwide, which is why it is hard to determine the real percentage of surgically treated patients [4]. In this study, we wanted to show the frequency of surgically treated PID patients in regard to all gynecological surgical procedures in our Department, which turned out to be relatively constant in a ten years period (Table 1).

Analyzing patients' age range, our results showed that surgically treated women with PID were mostly over 25 years of age. These results may seem surprising considering that PID typically occurs in women younger than 25 and rarely in older women [1,2]. However, this study includes only surgically treated patients' age and cannot be reflected in all PID patients. This result may be explained by the fact that in older, especially postmenopausal women, the clinical presentation is usually severe, with the more common formation of tuboovarian abscesses or additional intraabdominal pathology, which can all require surgery [6].

Laparoscopy was performed in slightly over half of cases, which is not surprising considering its advantages compared to laparotomy, such as less invasiveness, smaller incisions and shorter operation duration with less postoperative pain [5]. However, despite its many advantages, in more severe cases laparoscopy is not fully adequate. In those cases, patients need to undergo open abdominal surgery, which explains the relatively high percentage of laparotomy cases reported in this research (37.86%) [7]. Sometimes, intraoperative conversion of surgical procedures is needed due to complications or extensive adhesions [8]. In our study it was performed in 6% of cases. In 4% of cases transvaginal drainage from the pouch of Douglas, or by its other name, culdocentesis, was performed [7].

Laparotomy, or open abdominal surgery starts with an incision of abdominal wall [7]. In this study, we observed different types of incision and noticed that in about 42% of cases a midline laparotomy with a vertical incision was performed. The incision can extend from the xiphoid to the pubic symphysis. On the other hand, in 27% of cases, a transversal or Pfannenstiel incision was performed, which is placed in the curving interspinous skin crease [9]. However, in one-third of cases, this information remains unknown, which is why the real percentage of different types of incision is hard to determine.

As results showed, in almost half of cases infection was bilateral, which coincides with a fact that surgical treatment is reserved for severe cases. In around 20% of cases, the appendix was removed, with or without removal of genital tract structures. However, there was no information on whether the appendix was removed due to infection or unaffected as a measure of prevention [7].

Finally, in almost 18% of cases, the most radical surgical procedures included the partial or total removal of the uterus and adnexa. Almost every patient was older than 40 and most of them had an additional genital tract pathology. This result can be compared to a study

wherein 28.6% of surgically treated patients underwent a hysterectomy, out of which 95% were older than 40 and 57% had comorbidities [5]. Although the exact indications for such radical procedures remain unknown owing to the limitations of the used database, we can assume its relation to the fact that desire for future fertility is considered while making a decision about undergoing extensive surgical treatment [7].

CONCLUSION

Assuming that the incidence of sexually transmitted diseases is increasing, while bacteria are becoming resistant to a large number of antibiotics, we can expect that surgically treated PID will become more common. Indications for surgical procedures are becoming more important to recognize as, according to our data, patients treated surgically are older than 25 and procedures tend to be more extensive.

CONFLICT OF INTEREST:

The authors declare that there is no conflict of interest.

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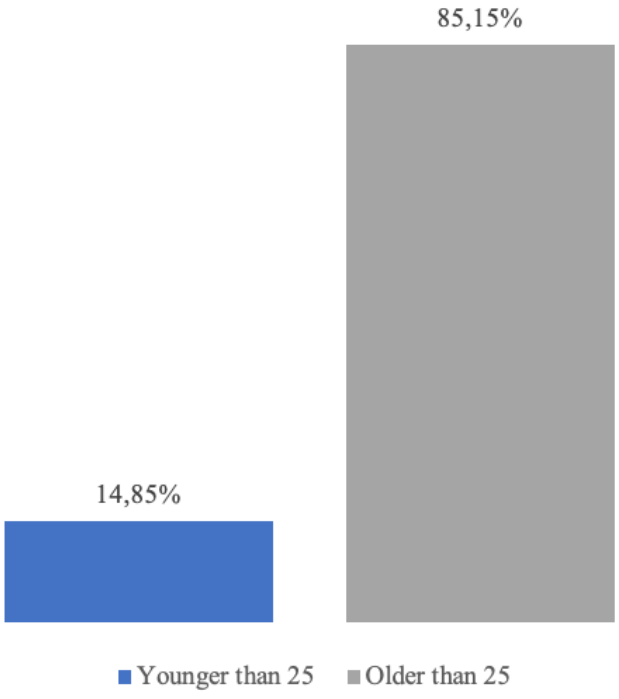

Figure 1. Graphical representation of patients' age

Year	All surgical procedures	PID surgical procedures	Percentage (%)
2009*	371	1	0,27
2010	932	11	1,18
2011	1006	11	1,09
2012	957	8	0,84
2013	699	10	1,43
2014	926	11	1,19
2015	1012	13	1,28
2016	1084	9	0,83
2017	1089	7	0,64
2018	1184	10	0,84
2019**	915	12	1,31
Total	**10175**	**103**	**1,01**

**September 1 - December 31*

***January 1 - September 1*

Table 1. Frequency of surgical procedures in a ten years period

Acta Chir Croat 2020; 17: 27-30

MENSTRUAL CYCLE RELATED PNEUMOTHORAX: CASE REPORT AND REVIEW OF THE LITERATURE

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ABSTRACT

Background: Catamenial pneumothorax is the most common form of thoracic endometriosis syndrome. It occurs around the beginning of a menstrual cycle. Although the mechanism of catamenial pneumothorax is not definitely clear, endometriosis plays an important role in it. Video-assisted thoracic surgery is standard procedure for the treatment of recurrent pneumothorax in general.

Case study: We report on a case of catamenial pneumothorax in women with a history of recurring spontaneous pneumotoraces associated with diaphragmatic endometrial implants who is involved in the *IVF* procedure.

Conclusion: Combination of video-assisted thoracoscopic surgery (VATS) and gonadotropin-releasing-hormone analogue gives the best results, to reduce the risk of pneumothorax to recur. Treatment of catamenial pneumothorax is complex and should include thoracic surgeon and gynecologist as soon as the diagnosis is definitive.

Keywords:
catamenial pneumothorax, video-assisted thoracic surgery, pelvic endometriosis, thoracic syndrome

INTRODUCTION

Catamenial pneumothorax (CP), derived from the Greek word *"katamenios"* which suggests its monthly occurrence, is a clinical condition defined as pneumothorax occurring within 72 hours of menstruation [1,2]. According to Mehta *et al,* CP is one of the most underdiagnosed etiological feature of primary spontaneous pneumothorax(PSP) found in young healthy menstruating women who addressed more than one case of clinical manifestation of dyspnoea and chest pain followed by or in coexistence with menses [3]. Although underdiagnosed, only 3% to 6% of spontaneous pneumothorax cases meet the definition of catamenial pneumothorax, establishing an indication for surgical treatment in only as 1/3 of the affected women as it was stated by Barbosa *et al.* [4].

Generally, CP was considered a rare phenomenon in history of its tracking with a previously reported prevalence of 0.9 – 5.6% of all cases of PSP in women, but with the progress in radiological diagnostics and better understanding of the condition, the awareness has been raised and the number of diagnosed cases has grown to 18 -30% [1,4].

The mean age of onset is 32-35 years in ovulatory women, while the women under hormonal therapy (such as contraceptive medication), the pregnant ones and women in menarche are generally not subject to it [1,3,5,6]. The localization of lesion found according to a review by Takahashi *et al.* is predominantly on the right side of the chest (85-95%), even though it may also occur on the left side (4.8%) or bilaterally (3.5%) [7].

There are three main criteria that determine the diagnosis of CP. Along with previously referred occurrence of the pneumothorax meticulously linked to ongoing menstruation, two subsequent criteria are clarified as: absence of any primary lung disease where pneumotoraces initiated secondary to a known underlying lung disease has not been classified as catamenial and at least one recurrent event (minimum of two episodes of pneumothorax) to achieve the definition criteria. [3-5,8].

While reevaluating the diagnosis of the CP before the surgical treatment, chest radiography, less often computed tomography (CT), and rarely magnetic resonance imaging (MRI) are performed. The fact radiography is routinely performed; there are no specific imaging diagnostic criteria [3,9].

When choosing the best treatment method for catamenial pneumothorax, several techniques have been proposed in literature. Nonetheless, video-assisted thoracoscopic surgery (VATS) has found to be the treatment of choice [1-10]. If possible, it is advisable to perform VATS during menstruation, because it allows maximum visibility of the potential endometriotic implants [9].

Thoracotomy is indicated almost exclusively in cases of recurrence after a previous procedure [11]. The use of video-assisted mini-thoracotomy (VAMT) has been suggested if the procedure involves extensive lesions within the diaphragm [8].

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CASE STUDY

A 33-year-old woman was admitted at thoracic surgery department for second onset of right pneumothorax in two months. After evaluation video-assisted thoracoscopic surgery (VATS) procedure was indicated and performed in a standard mode. Catamenial pneumothorax was suspected by clinical signs. Elective thoracic surgery procedures are not conducted during menstruation, so we operated after cessation of menstrual bleeding. The diagnosis of catamenial pneumothorax is rarely confirmed by histopathology as in our case. Three port VATS was performed, apical segment of the upper lobe resected and pleurodesis achieved by termocoagulation of the upper half of parietal pleura. During procedure typical lesions of the diaphragm ware found (Figure 1). Additional investigation reviled that both onsets of pneumothorax occurred on first day of menstrual bleeding. CA 125 level was 49.9 kIU/L (normal <35 kIU/L). Ultrasonography (US) examination showed a typical endometrial cystic formation measuring 25 mm on the right ovary. Gonadotropin-releasing-hormone analogue therapy was recommended but patient was trying to get pregnant after a series of miscarriage and rejected therapy. Two months after VATS procedure she had symptoms of pneumothorax and x-ray revealed partial subpulmonal pneumothorax that did not required treatment (Figure 2). Positron emission tomography scan was performed and suspected endometriosis at the abdominal side of the diaphragm was found. In the follow up period she had one more episode of pneumothorax like symptoms at the beginning of menstrual bleeding. Patient is currently in the *in vitro* fertilization procedure.

DISCUSSION

We report on a case of catamenial pneumothorax in women with a history of recurring spontaneous pneumotoraces associated with diaphragmatic endometrial implants who is involved in the *IVF* procedure.

Given the fact that pneumothorax is reported in concordance with menstruation, followed by the coexisting endometrial extrauterine tissue findings, suggests the presence of an undeniable connection between these two entities [1-12]. First association of CP with endometriosis was acknowledged by the Maurer *et al.*, who found erosive epiphrenic implants in patients suffering from recurrent spontaneous pneumotoraces whose appearance coincide with the beginning of menstruation [8]. Endometriosis, described as any endometrial tissue found outside of the uterine cavity, affects 5% to 15% of women in reproductive age [1-5,7-13]. Although it appears to be one of the most common benign gynecological proliferations, this disease remains poorly understood. Studies reviewed by Mehedintu *et al.* implicate there is no relationship between the extent of the disease and its symptomatology [14].

According to the previously mentioned, it is not the quantity of endometrial foci rather the localization that results with recurring pneumothorax. Several theories concerning the endometriosis related CP has been proposed. They all conflate around the idea of increased fallopian tube permeability in peri menstrual period combined with the fenestration in diaphragmic wall due to a congenital defect or more often seen endometrial metastatic lesions found on the surface of diaphragm that can damage the soft tissue of diaphragm and open the pathway to lung pleura [1,7,8,14-17]. In some cases the endometrial cells continue their migration through the diaphragm and form endometrial nodules on the surface of the pleura [8]. The reason why women's period represents the inevitable risk factor for a threatening pneumothorax lies in the fact that endometrial tissue attached to the pleura becomes physiologically active [7-13]. Influenced by gonadotropins released during menstruation, endometrial cells free the prostaglandin molecules which then corrode the small pulmonal vessel and alveolar septs culminating with occurrence of pneumothorax [18]. Accompanied by hematochezia, hemothorax and radiologically apparent endometrial nodules on the pleura it refers to a specific entity called thoracic endometriosis syndrome (TES). It's occurrence corelates with existence of pelvic endometriosis in around 50 – 70%, where the thoracic endometriosis occurred approximately 5 years later [3,7,14-18]. As previously detailed, difference in the mean age occurrence between pelvic and thoracic endometriosis may be explained with the time necessary for endometrial tissue to migrate through the right diaphragm [16].

Catamenial pneumothorax is found to be associated with both thoracic and pelvic endometriosis, although endometrial character of the disease cannot be confirmed histologically in every case [7,9]. From the clinical point of view, CP displayed to be leading cause of primary spontaneous recurrent pneumothorax with prevalence of almost 30% in emergency room [1-6]. When reviewing the literature upon the data on statistical correlation between catamenial pneumothorax and underlying endometriosis, the findings showed to be following.

From the prospective research of Rousset- Jablonski *et al.* where 156 women were surgically treated for spontaneous pneumothorax, we discovered that histologically documented thoracic endometriosis was found in 23.1% (36/156) of all patients (including catamenial and non-CP patients), being 6.5 times more frequent in patients with catamenial pneumothorax. As expected, pelvic endometriosis was found in about 50% of thoracic endometriosis cases, with wide variation among studies suggesting even higher rate of occurrence. In the analysis of 110 thoracic endometriosis cases by Joseph *et al.*, pelvic endometriosis was found

in 84% (51/61) of investigated patients with thoracic endometriosis [1].

The overall small number of reported cases suffering from thoracic endometriosis comes from the fact that not only it is a rare condition; endometriosis in the visceral pleura might be overlooked during surgery because endometrial tissue disappears at certain points during the menstrual cycle [7].

When investigating the possible causes of PSP during the first encounter with the patient, it is of crucial importance not only to ask for the last period commencement then to search for the pelvic pain and infertility history where pelvic exam including pelvic imaging is recommended [14,16-20]. In addition to previously mentioned, of great diagnostic value is the measurement of cancer antigen (CA)125 in cases of CP. As observed in research of Tsunezuka *et al.* in the female patients diagnosed with endometriosis related pneumothorax after VATS procedure, serum CA125 level was very high before the surgery compared to the control group of disease-free females [21].

As illustrated in this case report, our patient has had a history of miscarriage and was in the process of in vitro fertilization during hospitalization. After she presented with pneumothorax we opted for VATS, the video assisted thoracic surgery, where the suspected endometrial tissue was found on the inner wall of the diaphragm. Despite the fact that the procedure has been successfully carried out several episodes of reoccurring pneumothorax were documented. Their reoccurrence originates due to rejected analogues hormonal therapy recommended for lowering the recurrence risk [22].

The foundation of treatment for endometriosis-related pneumothorax, even before undergoing the VATS procedure, is the suppression of ovarian oestrogen secretion. In the long history of CP management commonly applied medications were oral contraceptives, progesterone agents, danazol or gonadotropin-releasing hormone (GnRH) agonists [23-25]. Although the preceding two have been used most frequently they all share similar mechanism. Women's self-regulated GnRH receptors are suppressed with GnRH analogues causing down-regulation and creating reversible hypogonatrophic hypogonadism. In previous years, hormone therapy has been the backbone treatment of CP. However through years, VATS was introduced as a method of choice and hormone therapy took place as maintenance therapy preventing the recurrence of the pneumothorax. The reason for that lays in the fact that hormonal therapy needed to be maintained for prolonged period of 6 months causing loss of trabecular bone density, hot flashes and vaginal dryness which presented as large drawback for its utilization [25-27].

In regard with our patient's wishes to conceive, estrogen suppression was contraindicated.

CONCLUSION

Leakage of air in pleural space due to the rupture of alveolar septs influenced by women's menstrual cycle, which is shown to be recurrent in its nature, is defined as catamenial pneumothorax. Because of the high recurrence rate despite the therapy, a need evolved to determine the underlying cause to make the treatment more effective and to lower the recurrence to a minimum. Combination of VATS and gonadotropin-releasing-hormone analogue gives the best results, to reduce the risk of pneumothorax to recur. Treatment of catamenial pneumothorax is complex and should include thoracic surgeon and gynecologist as soon as the diagnosis is definitive.

CONFLICT OF INTEREST:

The authors declare that there is no conflict of interest.

The patient gave her informed consent prior to her inclusion in case report.

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FIGURES

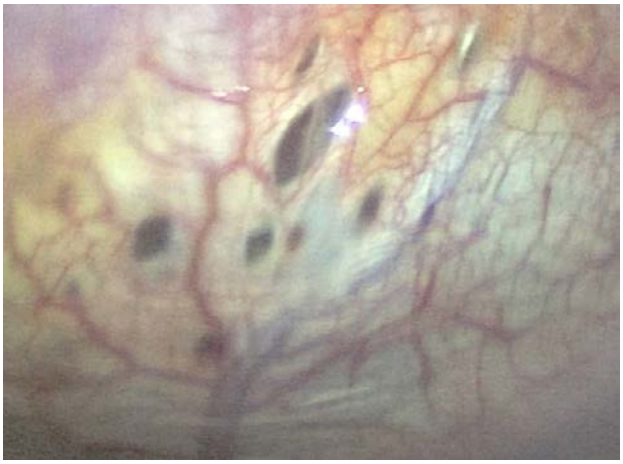

Figure1. Intraoperative finding of diaphragmal lesion.

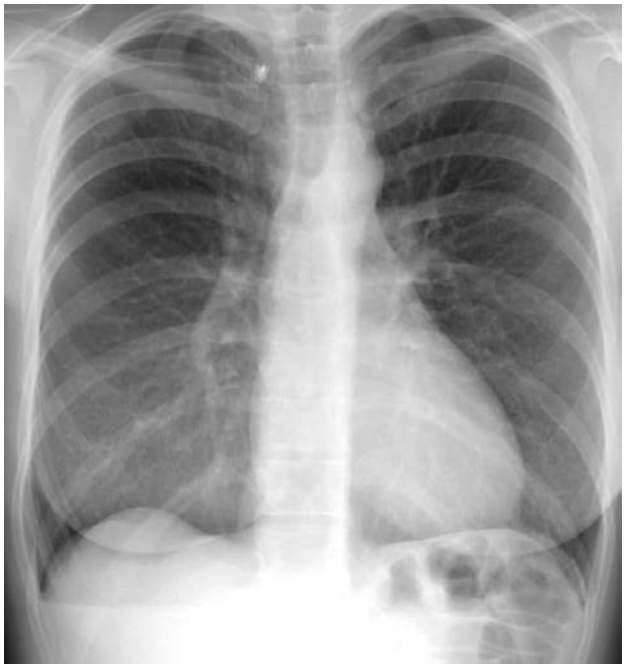

Figure2. Chest x-ray shows subpulmonal pneumothorax.

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DOUBLE SCHWANNOMAS OF THE ULNAR NERVE: CASE REPORT

Filip Mustac[1], Mia Poturica[2], Kresimir Bulic[3]

ABSTRACT

Background: Schwannomas are benign peripheral nerve sheath tumors, mainly solitary, well defined and with low incidence of malignant transformation. The incidence of schwannomas in hand tumors is 5%. Schwannomas are initially asymptomatic, but later due to their growth they can compress surrounding tissue and cause pain.

Case study: We present a case of a 68-year-old patient with a double schwannoma of the right ulnar nerve. The MRI showed a soft tissue mass which was suspected to be a schwannoma and the diagnosis was confirmed by pathohistology. Enucleation of both tumors with nerve preservation was performed and the patient is without any postoperative neurological deficit.

Conclusion: Intra-capsular enucleation with nerve preservation is a treatment option which may significantly lower postoperative nerve damage wth an acceptable risk of local recurrence.

Keywords:
schwannoma, magnetic resonance imaging, ulnar nerve

INTRODUCTION

Schwannoma is a benign nerve sheath tumor which rarely turns malignant. The first schwannoma was described by Verocay in 1908. They are most commonly solitary and grow slowly. They are typically 1.5 to 3cm in diameter [1,2]. They can affect any nerve in the body, where among the hand tumors the incidence of schwannoma is 5%. After surgical removal, the incidence of recurrence is very rare. When looking among all nerves, the one most commonly affected by schwannoma is the *median nerve* [3]. In general, the schwannoma occurs more often in the nerves of the upper extremities than lower. They most commonly occur between the ages of 30 and 50. Magnetic Resonance Imaging (MRI) is the gold standard in peripheral nervous system tumor diagnosis [1]. When doing the differential diagnosis, one must consider neurofibroma, as well as the malignant peripheral nerve tumors; where the final diagnosis does require a pathohistological finding. The choice therapy is tumor resection with as much nerve preservation as possible in order to preserve function [3].

CASE STUDY

A year ago, sixty-eight-year-old woman noticed a lump on the hypothenar muscle of her right fist that was progressively getting larger and was painful on touch. Physical examination determined a lump size 1x1cm, of hard consistency, movable in respect to surface, and without skin changes. Pressure and percussion of the lump caused throbbing pain in the fourth and fifth finger. The patient had a pulpable lump on the ulnar side of the distal forearm which was asymptomatic and looked like a ganglion cyst. As it seemed that this could be a tumorous form either originating from the ulnar nerve or pressing on it, the patient was sent for an MRI of the right wrist (Figure 1). An oval and sharply delineated form in the subcutaneous fat tissue of the palm at the level of the 5th carpometacarpal joint was visible before and after the intraveneous contrast application. It measured at 0.8x0.7x0.4cm and appeared to be well vascularized. It was located on the *abductor muscle of digiti minimi* and 2mm from the *ulnar nerve*. There were no signs of infiltration of the surrounding tissue. According to the MRI characteristics, it resembled a neuroma. The exploration of the formation was done under general anesthesia and with tourniquet. The suspected tumor of the ulnar nerve in the Guyun channel was confirmed, and the formation in the distal forearm was also in the continuum with the ulnar nerve (Figure 2). An intra-capsular enucleation of both formations was performed along the line of the direct seam of one of the fascicles which was cut during the enucleation of the distal tumor.

Histologically, both nodes were made of long bundles of spindle cells whose nuclei were partially collapsing. There was a spindle capsule on the top. Pathology results determined a diagnosis of schwannoma.

DISCUSSION:

This case was about schwannoma of the ulnar nerve. The gradual growth of the tumor leads to the compression of the nerve which may result in pain; although when it comes to schwannoma most commonly there is

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DOI: *10.5281/zenodo.4010883*

only a painless soft tissue growth. Very rarely, it may be manifested as a focal neuronal outbreak [3]. Due to this inprecise clinical picture there is a bigger problem with differential diagnosis, especially neurofibromas, as well as other soft tissue formations. The appearance of multiple soft tissue structures, especially in younger patients with loaded family history, should raise red flags regarding neurofibromatosis. If the patient has multiple schwannomas, but no tumor of the vestibular nerve, this could be an indication of possible schwannomatosis which is the third type of neurofibromatosis [4]. Electromyography is not of much use, except to show which nerves or plexuses are affected. Computer Tomography (CT) in the diagnosis of the soft tissue tumor structures is not the best choice becuase the density of the possible structures is very similar to the muscle density. CT could be useful if there is tumor calcification. MRI can detect the origination site, the position in relation to the nerve (whether it is intrinsic or extrinsic), as well as the actual structural characteristics such as irregular edges, bleeding, or necrosis [5]. Even though the MRI is very good at showing the structure, the final diagnosis can only be made through a pathohistological examination.

Pathohistological examination is used to determine the type of tumor and whether the tumor is benign or malignant. When it comes to schwannoma there are two histological regions Antoni A and Antoni B. Antoni A region consists of spiral bound spindles of Schwann cells forming palisades and producing Verocay bodies. As opposed to the hypercellular Antoni A regions, Antoni B regions are hypocellular and the Schwann cells do not have a specific arrangement within the loose connective tissue. In most tumors either Antoni A or Antoni B regions dominate. When diagnosing peripheral nerve tumors, it is very useful to perfom immunohistochemical analysis which determines antibodies for S-100, CD31, CD34 and GFAP proteins [1].

Microsurgical treatment is the therapy of choice. In most cases improvement in peripheral nerve function has been achieved by enucleation, however there is a possibility of iatrogenic nerve damage. There are two enucleation techniques – extra-capsular and intra-capsular. In one study the advantages of intra-capsular enucleation of schwannoma over the extra-capsular were examined. The study had 35 patients, and 36 schwannomas. Twenty schwannomas were treated via extra-capsular enucleation, while 16 were treated via the intra-capsular. The postoperative neurological deficits were classified as light, heavy, and transient. Of the total number of patients 22 had no neurological deficit or they developed a deficit which subsided in six months. Ten patients developed a new neurological deficit which required more than six months to subside. Four patients developed a motor deficiency which continued through the publication of the study; where all patients with a motor deficiency had been treated with extra-capsular enucleation. The study showed that intra-capsular enucleation is a safer approach to treatment than extra-capsular as there is a much lower risk of nerve injury [5]. In this case we decided on the intra-capsular enucleation as it carries a lower risk of nerve injury, especially as the patient had a tumor on two levels and where the final cummulative nerve damage could significantly compromise functioning. After the surgery, the patient did not have any obvious neurological incidents, had a two-point discretion of 4 mm on the 4th and 5th fingertip and hand muscle strength of 5/5 innervated by an ulnar nerve.

CONCLUSIONS

Schwannoma is a tumor of the nerve sheath which occurs with the frequency of 5% among tomors of the hand. Intra-capsular enucleation with nerve preservation is a treatment option which may significantly lower postoperative nerve damage wth an acceptable risk of local recurrence.

CONFLICT OF INTEREST:

The authors declare that there is no conflict of interest.

The patient gave her informed consent prior to her inclusion in case report.

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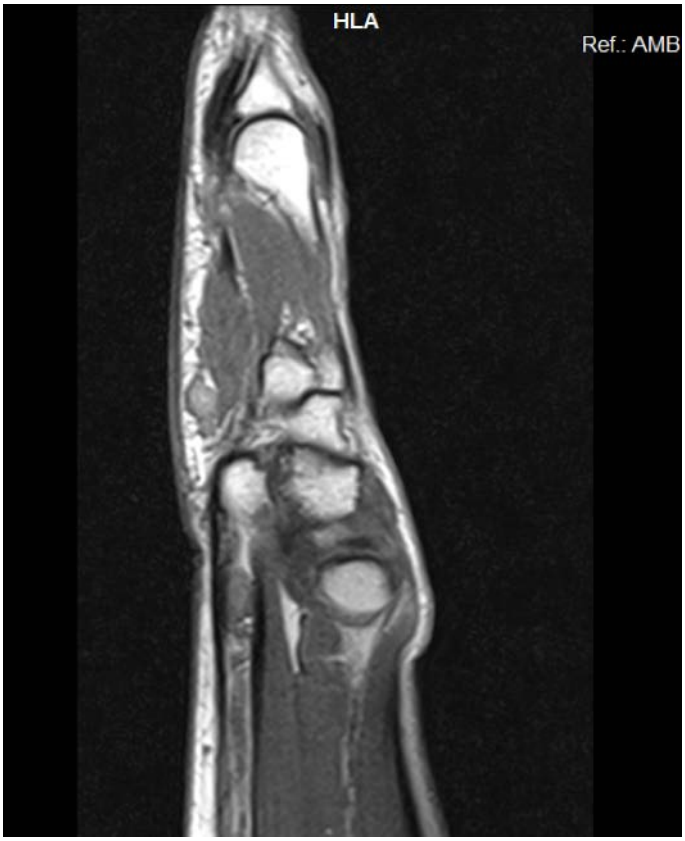

Figure 1. Oval and sharply constrained formation in subcutaneous fat tissue on the palm at the height of the 5th carpometacarpal joint.

Figure 2. Intraoperative picture of the right fist with changes in the ulnar nerve in the distal forearm and in the Guyon channel area

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MULTIPLE ESOPHAGEAL LEIOMYOMAS: CASE REPORT

Željko Djaković[1], Dinko Stančić-Rokotov[1], Ivka Djaković[2]

ABSTRACT

Background: Primary intramural benign tumors of the esophagus are rare. Leiomyomas are the most common benign esophageal neoplasms. Multiple esophageal leiomyomas are very rare, with only a few reports on more than ten coexisting lesions.

Case study: A male patient presented with progressing dysphagia and a tumor of the esophageal wall, over 10 cm in length, confirmed by magnetic resonance imaging and endoscopic ultrasound examination. There were no changes of the esophageal mucosa. Multiple fine needle aspirations were performed with inconclusive finding. Surgical exploration through right thoracotomy revealed multiple extramucosal tumors from 5 to 25 millimeters in size. A total of 16 tumors were removed by enucleation without opening the esophageal mucosa. Postoperative period was uneventful.

Conclusion: Exact preoperative diagnosis of esophageal submucosal tumors may be difficult to establish without open biopsy. Removal by enucleation is the treatment of choice.

Keywords:
multiple leiomyomas, diagnostic imaging, dysphagia, esophageal tumor

INTRODUCTION

Primary intramural benign tumors of the esophagus account for 2% of all esophageal tumors. Leiomyomas are the most common benign esophageal neoplasms. Incidence in autopsy is from 0.005 to 5%. Multiple esophageal leiomyomas are extremely rare. There are only a few reports on more than ten coexisting lesions. [1,2]

CASE STUDY

A 62-year-old male patient presented with progressing dysphagia and a tumor of the esophageal wall, over 10 cm in length, confirmed by magnetic resonance imaging (Figure 1) and endoscopic ultrasound examination. There were no changes of the esophageal mucosa. Multiple fine needle aspirations were performed with inconclusive finding. After complete diagnostic work-up the diagnosis of a solitary process was established. However, surgical exploration through right thoracotomy revealed multiple extramucosal tumors from 5 to 25 millimeters in size (Figure 2.A/B). A total of 16 tumors (Figure 2.C) were removed by enucleation without opening the esophageal mucosa. Histopathological diagnosis was Leiomyomatosis oesophagi. Patient was discharged on the seventh postoperative day. Postoperative period was uneventful. There were no swallowing problems since the release. No signs of extramural compression were found at esophagoscopy 40 days after surgery.

DISCUSSION

Diagnostic algorithm for esophageal submucosal tumors includes computed tomography or magnetic resonance imaging, esophagoscopy, endoscopic ultrasound and needle biopsy [3-5]. Leiomyomas should be considered whenever there is no mucosal involvement. Surgery should be performed if symptoms are present, the tumor grows during follow-up, or coexisting malignancy is suspected [1,4]. Surgical options include esophagectomy or extramucosal enucleation [2]. Multiple leiomyomatosis can mimic malignant disease and sometimes diagnosis is established at the operation.

CONCLUSION

Although multiple esophageal leiomyomas are extremely rare they should be taken in to consideration, especially if there is no mucosal involvement. Exact preoperative diagnosis may be difficult to establish without open biopsy. Removal by enucleation is the treatment of choice.

CONFLICT OF INTEREST:

The authors declare that there is no conflict of interest.

The patient gave his informed consent prior to his inclusion in case report.

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FIGURES

Figure 1. MRI showing a tumor of the esophagus over 10 cm in length.

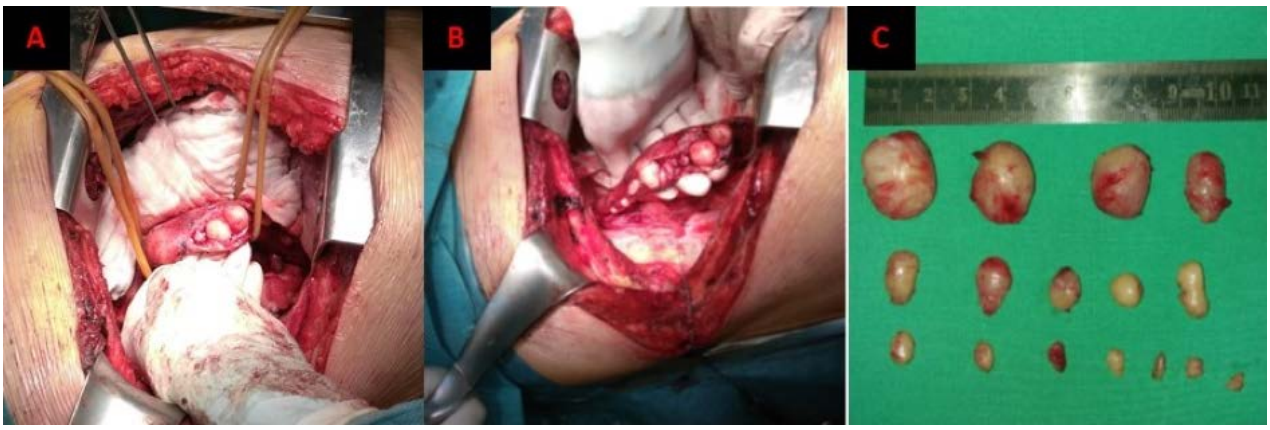

Figure 2.

A, B) Opening of the muscular layer of the esophagus revealed multiple solid leiomyomas

C) A total of 16 enucleated tumors measuring from 5 to 25 mm.

Acta Chir Croat 2020; 17: 37-39

A PATIENT WITH THREE AORTOENTERIC FISTULAS IN A PERIOD OF FIVE YEARS: CASE REPORT

Josip Figl[1], Andrea Crkvenac Gregorek[1], Predrag Pavić[1,2], Irena Šnajdar[1], Marko Mance[1]

ABSTRACT

Background: Aortoenteric fistula (AEF) is a pathological communication between the aorta and gastrointestinal tract that presents a life-threatening condition. It can be primary or secondary, based on the underlying cause of fistula development.

Case study: We present a 67-year-old female patient who suffered from three secondary AEFs in a period of five years. After two abdominal surgeries for gastric ulcer and colorectal adenocarcinoma (TNM stage II), the patient had an open abdominal aortic aneurysm reconstruction. For each AEF presentation, opened surgical reconstruction was performed.

Conclusion: Morbidity and mortality rates after AEF surgery are high despite advances in surgical techniques and materials. Three times recurrent AEF in a single patient with 5-years survival after initial reconstructive surgery is rare event.

Keywords:

Aortoenteric fistula, aortic surgery (AEF), aortic reconstruction complication, abdominal aortic aneurysm (AAA)

INTRODUCTION

Aortoenteric fistula (AEF) is a pathological communication between the aorta and gastrointestinal tract. It can be primary or secondary, based on the underlying cause of fistula development [1]. A primary AEF occurs naturally, without any prior aortic reconstruction. It can develop due to compression of the AAA on the duodenum, local inflammation or aortic infection (peptic ulcer perforation into the aorta or tumor erosion of the aorta) [1].

A secondary AEF forms following some surgical intervention such as an aortic reconstruction. The AEF usually affects the distal third or fourth region of the duodenum, but can also affect the proximal jejunum [2]. In the majority of cases, the initial event leading to this condition is a prosthesis-infection, leading to a weakening of the suture line. This subsequently leads to a false aneurysm and AEF formation [3]. In a small number of cases continuous pulsatile pressure of the aortic graft to duodenal compression or duodenal injury leads to AEF formation [4].

Diagnostic options in detecting this condition include esophagogastroduodenoscopy (EGDS), multislice computed tomographic angiography (MSCTA) and scintigraphy with radiographically marked red blood cells: all of which are seldom positive (MSCTA in 11%) [5,6]. Due to these low rates of true positive findings, clinical examination and physicians' opinion remains the most important diagnostic tool.

AEF presents a rare and particularly complicated problem in vascular surgery. When left untreated, the outcome is usually fatal. Surgical repair is fraught with complications, morbidity and mortality rates remain high despite advances in surgical techniques and materials [1].

A recurrence of AEF following surgery in the same patient is an extremely rare clinical finding.

CASE STUDY

A 67-years old female patient came to the emergency department because he collapsed and had signs of melena. Her blood pressure initially was 95/70 mm Hg, heart rate 90 bpm and hemoglobin concentration of 65 g/l. She had a history of stomach ulcer surgery, right hemicolectomy due to colorectal adenocarcinoma (TNM stage II) and chronic renal failure. Also, surgical reconstruction of the abdominal aorta using a Dacron graft (Vascutec Gelsoft Plus, Terumo, Tokyo, Japan) due to an abdominal aortic aneurysm (AAA) was performed in another institution six years prior to admission. An emergency EGDS was performed which did not show any active or recent bleeding. There were no signs of a visible aortic graft through the bowel or other signs of bowel perforation. MSCTA showed periaortic hematoma and active extravasation from the aorta directly into the bowel which was adherent to the previously reconstructed aorta – an AEF (Figure 1). Emergency surgery was performed during which the rupture of the proximal and distal aortic anastomosis was discovered and an AEF with the proximal jejunum. Due to the patient's hemodynamic instability, only re-suturing of the proximal and distal aortic anastomosis of the existing aortic graft was performed. The bowel perforation was directly sutured in two layers.

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DOI: *10.5281/zenodo.4010898*

Following surgery, the first twelve postoperative days went uneventful. On the 13th day the patient had recurrent stomach pain and hematochezia, with consequent blood loss resulting in a hemoglobin count of 68 g/L . A MSCTA was ordered which showed the possibility of active extravasation in the previously operated area. The patient underwent to a second surgical procedure during which active bleeding and anastomotic disruption was ruled out.

After six weeks of antibiotic treatment with teicoplanin, meropenem, imipenem, cilastatin and fluconazole, the patient was discharged.

That same year the patient was re-operated in another hospital due to a new AEF formation. During that procedure the old prosthesis was removed and replaced with a new Dacron prosthesis (Interguard Silver, Maquet, Rastatt, Germany).

Four years after the second procedure due to AEF, the patient presented with hematemesis and hematochezia (lasting one day) in the emergency department. An MSCTA revealed a newly formed AEF with active bleeding into the bowel. Surgery was indicated and the patient lost consciousness just before entering the operating theatre. Her condition of hypovolemic shock was accompanied with decreased blood pressure (BP 70/40 mm/Hg), tachycardia (HR 123 bpm) and consequent cardiac arrest. Cardiopulmonary resuscitation was initiated and a heart rhythm was restored. During the surgery, removal of the previous graft was performed and a new aortic reconstruction (Silver Graft, B. Braun, Berlin, Germany) and bowel suturing were completed successfully. Despite all efforts, the patient died two hours after surgery in the intensive care unit due to cardiopulmonary failure.

DISCUSSION

AEFs are rare, but when they present are very severe and urgent conditions, usually demanding prompt diagnostics and treatment. The diagnostic capabilities are not very specific and it is very difficult to diagnose an AEF with any imaging methods available or with EGDS. It is therefore usually left to clinical suspicion and physician experience that guides the surgeon towards the diagnosis and indication for emergency surgical treatment. Secondary AEFs are seen much more frequently than the primary cases [7]. In the case of a secondary AEF, diagnosis is often easier because it is usually seen in a patient who previously underwent an aortic reconstruction. These patients commonly present with melena and "herald bleeding".

However, recurring AEF's following previous surgical reconstruction is a rare occurrence. The patient we presented in this paper presented with her first secondary AEF one year after the initial reconstruction of the AAA. Nevertheless, she later presented with two more AEFs which resulted in three AEF surgeries. She survived five years after the initial AEF reconstruction which is an excellent result.

There are many methods of AEF reconstruction such as local graft repair, excision of the graft without the reconstruction, in situ graft replacement and extra-anatomic revascularization with graft excision or delayed excision after two to three days. All the mentioned methods are accompanied with a high mortality rate and it is still doubtful which is the best choice [8]. In this patient, several methods were performed.

The first surgery included re-suturing the old prosthesis. This method was chosen due to the poor hemodynamic instability of the patient. Although this method has been associated with worst outcomes, high rates of reinfections and high mortality rates, in this situation, this was the only possible option to save the patient [9-12].

The second AEF surgery was done in another hospital almost one year after the first. In this procedure the old graft was removed and replaced with a new prosthesis.

Five years after the initial AEF reconstruction surgery, the patient developed a third AEF. At this point the patient was in a very poor clinical condition. Despite the difficulty to operate this patient due to the previous abdominal surgeries, the reconstruction was successful and a new graft was placed. The patient died couple hours following surgery.

CONCLUSIONS

AEF presents as a rare and extremely complicated condition in vascular surgery. There are vast arrays of surgical procedures available which can lead to many outcomes depending on the patient. Using the appropriate surgery in critically ill patients is imperative and it is the goal of the surgeon to give them the best chance of survival,

Unfortunately, after multiple efforts our patient eventually died as an indirect result of her AEF. This patient was our first to have survived two previous AEF surgeries and the only patient who had developed a total of three in her lifetime.

CONFLICT OF INTEREST:

The authors declare that there is no conflict of interest.

The patient gave her informed consent prior to her inclusion in case report.

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FIGURES

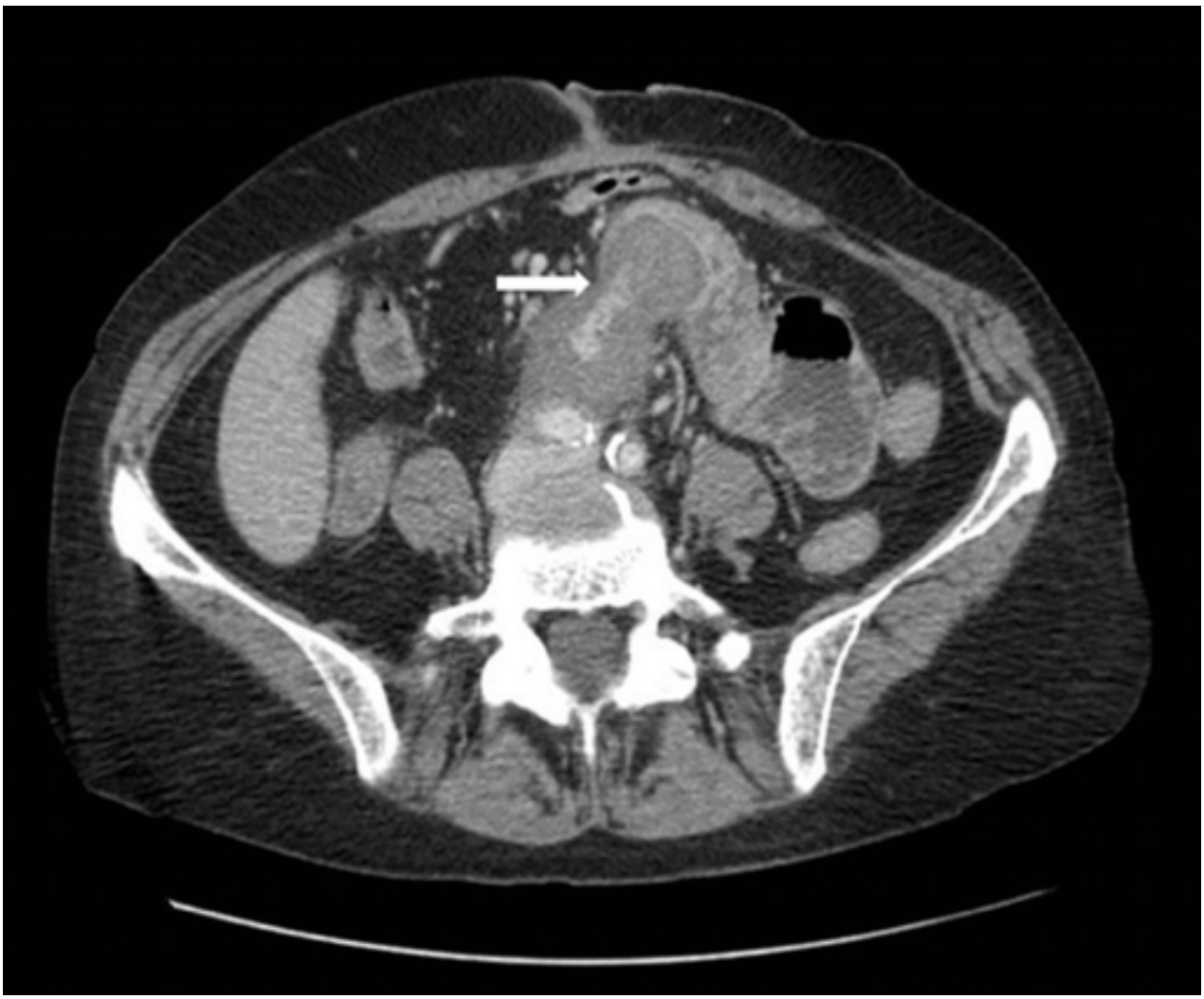

Figure 1. Arrow in the image shows the position of aortoenteric fistula (AEF)

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