UDK 617 – 089

ISSN (print) 1845 – 2760
ISSN (online) 1848 – 5367

Acta Chirurgica Croatica

OFFICIAL JOURNAL OF THE CROATIAN SOCIETY OF SURGERY

www.acc.hkd.com.hr

Vol. 16 No 1 (p 1–40) Zagreb 2019

UDK 617 – 089

ISSN (print) 1845 – 2760
ISSN (online) 1848 – 5367

Acta Chirurgica Croatica

OFFICIAL JOURNAL OF THE CROATIAN SOCIETY OF SURGERY

www.acc.hkd.com.hr

Vol. 16 No 1 (p 1–40) Zagreb 2019

IMPRESSUM

Publisher

Croatian Society of Surgery – Croatian Medical Association
Department of Surgery, University Hospital Center Zagreb

Graphic design and print
BTravel d.o.o., Zagreb

Editorial Office
Acta Chirurgica Croatica
Department of Surgery, University Hospital Center Zagreb
Kišpatićeva 12
10 000 Zagreb
E-mail: editor@acc.hkd.com.hr

Circulation
300

Official Journal of
Croatian Society of Surgery
Croatian Society of Pediatric Surgeons
Croatian Society for Endoscopic Surgery

Acta Chir Croat 2019; 16: 3-4

INSTRUCTIONS FOR AUTHORS

Legal Requirements

Acta Chirurgica Croatica publishes original research articles, case reports, reviews and short communications, in Croatian or English, on current developments in surgical practice and research. Manuscripts submitted for publication must contain a statement to the effect that all human studies have been reviewed by the appropriate ethics committee and have therefore been performed in accordance with the ethical standards laid down in an appropriate version of the 1964 Declaration of Helsinki. It should also be clearly stated in the text that all persons gave their informed consent prior to their inclusion in the study. Details that might disclose the identity of the subjects under study should be omitted.

Conflict of Interest

Authors must indicate whether or not there is a financial relationship between them and the organization that sponsored the research. This note should be added in a separate section previous to the reference list. If no conflict exists, authors should state: "The authors declare that there is no conflict of interest". The editor in chief reserves the right to reject manuscripts that do not comply with the above stated requirements. The author will be held responsible for false statements or for failure to fulfil the requirements. Every paper is assessed by an independent reviewer on whose recommendation the editor's decision on acceptance or rejection will be based.

Editorial Procedure

Authors are requested to submit their articles by e-mail to the Editorial Office:

petra.matjasec@kbc-zagreb.hr

For all business communication concerning the journal please use the above e-mail address or:

Acta Chirurgica Croatica
UHC Zagreb
Department of Surgery
Kišpatićeva 12
10 000 Zagreb, Croatia

After the article has been accepted and published, the publisher will supply the corresponding author with two complimentary copies of the relevant issue.

Manuscript Preparation

Arrange the elements of the manuscript in the following order: (1) Title, (2) Abstract, (3) Keywords, (4) Introduction, (5) Material and methods, (6) Results, (7) Discussion, (8) Conclusions, (9) References, (10) Figure captions, (11) Tables.

Pharmaceutical products have to be given with their international generic names. Protected registered trade names should appear with the symbol® (e.g. Aspirin®).

Use the standard international way of writing units (unit converter www.vin.com/calculators/default.htm). On the title page include the title of the manuscript, its running head (condensed title, not exceeding 60 characters with spaces), the name and surname of each author, each author's affiliation, the correspondent footnote (name and surname, postal code, telefax number, e-mail address) and footnote(s) indicating the present location of authors no longer at the institution at which the work was performed. The authors' affiliations must be indicated after each name in superscript numbers.

Abstracts

Provide abstracts in Croatian and English, each limited to 250 words and structured by the following run-in heads "Background", "Methods", "Results", "Conclusions". For authors from abroad the abstract will be translated by the Editorial Office.

Keywords

Provide up to five keywords in Croatian and English (a keyword may also be a noun phrase consisting of three or more words). Keywords should allow an appropriate classification of the paper in regard to the methodology and field of application. Choose them according to Index Medicus, do not merely duplicate words from the title.

References

References may be made to published work and papers in press. Reference citations are not permitted in the abstract of a paper. Authors are responsible for the accuracy of all literature citations. Work in progress, in preparation, unpublished work and personal communications should be cited as footnotes to the text. References should be listed numerically in the text in the order of appearance as parenthesized consecutive numbers, e.g. [1, 2]. Where there are more than two references, the citation should appear as a range of numbers separated by a dash, e.g. [1–3]. Abbreviations of journals should conform to those used in Medline.

The following formats must be used:

Journal article with maximum six authors: all of the authors are listed. Journal article with more than six authors: only the first three authors are listed followed by "et al.".

Journal article in press (manuscript has been accepted for publication): Journal name (in press).

Abstracts: authors, title, journal, volume, page, year, abstract number, e.g. Proc Am Soc Clin Oncol 2007;25:215s (abstr 4571).

References should be quoted according to the Vancouver Format (Ref.), e.g. Journal article Smith JJ. The world of science. Am J Sci 1999;36: 234–5.

Book: Blenkinsopp A, Paxton P. Symptoms in the pharmacy: a guide to the management of common illness. 3rd ed. Oxford: Blackwell Science; 1998.

Book chapter: Wyllie AH, Kerr JFR, Currie AR. Cell death: the significance of apoptosis. In: Bourne GH, Danielli JF, Jeon KW, editors. International review of cytology. London: Academic; 1980. pp. 251–306.

Article by DOI: Slifka MK, Whitton JL (2000) Clinical implications of dysregulated cytokine production. J Mol Med (in press). DOI: 10.1007/s103530000086.

Online document: Doe J (1999) Title of subordinate document. In: The dictionary of substances and their effects. Royal Society of Chemistry. Available via DIALOG. http://www.rsc.org/dose/title of subordinate document. Cited 15 Jan 1999.

Always use the standard abbreviation of a journal's name according to the ISSN List of Title Word Abbreviations, see www.issn.org.

Text

1. Please write your text in Microsoft Word. Use a Times New Roman plain font 12 for text.
2. Use the automatic page numbering function to number the pages.
3. Do not use field functions.
4. For indents use tab stops or other commands, not the space bar.
5. Use the table functions of your word processing program, not spread sheets, to make tables. For equations use the Microsoft Word equation editor or Math Type.
6. Place all figure captions or tables at the end of the manuscript.
7. Submit all figures as separate files and do not integrate them in the text.

Figures and Tables

The preferred figure formats are EPS for vector graphics exported from a drawing program and TIFF for halftone illustrations. EPS files should contain a preview in TIFF of the figure. The file name (one file for each figure) should include the figure number. Figure captions should be included in the text and not in the figure file. Scan resolution: scanned line drawings should be digitized with a minimum resolution of 800 dpi relative to the final figure size. For digital half tones, 300 dpi is usually sufficient.

Color illustrations: Store color illustrations as CMYK in TIFF format.

Vector graphics: All fonts used in the vector graphics must be embedded. The minimum line width should be 0.2 mm (i.e. 0.567 pt) relative to the final size.

The number and final size of illustrations and tables must be kept to the minimum required for clarification of the text. Do not give the same data both in tables and graphs. Do not use pie charts. Do not integrate figures or tables into the text. Number figures and tables consecutively in separate series. In addition to the number, each figure must have a legend and each table must have a title. All figures and tables must be cited in the text. Plan figures to fit into the column width of 81 mm. The maximum space available on one page is 169 by 240 mm. Scale the length, width, point size of type and symbols, and line weights of a figure proportionally. At the final size of a figure, no capital letter or symbol should be smaller than 1.5 mm and no line weight should be less than 0.25 mm. Group the legends for figures on a separate sheet. Give the title and all footnotes of a table directly above and below the table.

Full Paper and Review

Full papers and reviews should be written as concisely as possible without impairing the clear and precise presentation of the subject matter. Both form and content of the paper should be carefully checked to exclude the need for corrections.

Short Communication

Short communications should be prepared as described above except for the following: the average length of short communications should not exceed 1500 words and a maximum of two figures or tables is accepted. The abstract should not exceed 80 words.

Case Report

Case reports should have educational value and/or highlight the need for a change in a certain clinical practice. They must provide an original description of a previously unreported entity or report a new presentation of a known disease or a new perspective of a case which poses a diagnostic and therapeutic challenge. Case reports should include a comprehensive review of similar cases and emphasize the differences between present case and the previous ones. Case reports should be accompanied by clinical images. Authors should seek from the patients a written and signed consent to publish the information.

Letter to the Editor

Letters to the editor are welcome, they should contain a maximum of 750 words, and may include a table or figure and references.

Corrections

All necessary corrections will be sent by e-mail to the corresponding author. The costs of corrections and changes, except the ones made in typesetting, which exceed 10% of typesetting costs, and were made after page numeration, will be charged to the author.

CONTENTS

Acta Chir Croat 2019; 16: 7-10

PREVALENCE OF OVERWEIGHT AND OBESITY IN PEDIATRIC SURGICAL POPULATION

Anko Antabak[1], Dino Papeš[1], Josipa Zaller[2], Luka Penezic[3], Kresimir Bulic[1], Tomislav Luetic[1]

ABSTRACT

Background: Obesity has become one of the most significant public health problems in the world in recent decades. It is associated with an increased incidence of postoperative complications, and there is a cause-and-effect relationship between obesity and increased prevalence of injury in children. Although there are studies in the world that indicate an increased incidence of obesity among surgically treated children, no such research has been conducted in Croatia so far.

Materials and methods: This study aimed to determine the nutritional status of children treated at the Department of Pediatric Surgery at two samples in the time lag of eight years (2010 and 2018). In 2010, 1205 children were treated in hospital, 790 children fulfilled the inclusion criteria. In 2018, 1316 children were treated in hospital, and 790 children met the inclusion criteria. The collected data were compared with the existing health statistics.

Results: The proportion of obese children was 29.7% in patients treated in 2010 and 29.7% in patients treated in 2018. In 2010 the highest prevalence of obese children was in pre-school age (23.8%) and overweight in school-age (16.7%). In 2018 the highest incidence of obesity in school children was 38.9%, in preschool-age 31.6%, in puberty 28.3%, and the lowest in adolescents 23.3%.

Conclusion: The prevalence of obesity in children undergoing hospitalization for surgical illness is higher than in the general population. There was no trend of increasing the incidence of obesity over a 10-year time lag.

Keywords:

obesity, pediatric surgery, pre-school, puberty, adolescents

INTRODUCTION

Over the last decade, obesity has become more obvious public health problem both in the world and in our country [1]. According to the International Classification of Diseases (ICD-10 edition), obesity is considered a disease (code E65 and E66), and for some time there has been a continuous increase in obesity in all age groups, regardless of the level of development of the social environment [2,3]. Obesity in children is determined by standardized gender and age-specific normograms. Children whose body mass index (BMI) is equal to or greater than the 85 percentile for their age and gender are considered obese. The lighter form of obesity we call an overweight, which is defined by BMI ≥85, <95. percentiles. A severe form of obesity we call obesity and is defined by a BMI ≥ 95 percentile.

In the Republic of Croatia (RH), nutrition status data are not part of the routine health statistics of children or adults. Data on the prevalence of obesity in a healthy population of the Republic of Croatia is known from the Croatian Health and Statistics Yearbook for 2009 (Croatian Institute for Public Health). The European Office of the World Health Organization has been implementing the child nutrition monitoring Initiative since 2006, and the Republic of Croatia has entered into a program for continuous monitoring of nutritional status of children, in a uniform methodology, from 2015/2016, therefore these results can be compared with the other 35 countries that are part of this program [4].

The negative consequences of obesity in children are manifested in childhood, and growing up, fat children are at high risk of developing cardiovascular, endocrine, gastrointestinal, renal, respiratory, and psychosocial disorders.

Surgical patients undergo preoperative anesthetic examination, which records the parameters of perioperative risk, and obesity is one of the factors that carries the risk of developing perioperative complications. The main activities of pediatric surgery include the treatment of acute and chronic diseases, injuries and malformations, through an elective and emergency program, including preoperative, operative and postoperative aspects. Children are mostly operated under general anesthesia, and in obese children there are special features in pharmacokinetics and dynamics of anesthetic. As there is no standardized protocols, the effects of anesthetic in children, due to the fat content in body weight, can be altered and lead to prolonged

[1]*Department of Surgery, University Hospital Centre Zagreb, Croatia*
[2]*School of Medicne, Univerity of Zagreb, Croatia*
[3]*Department of Urology, University Hospital Centre Zagreb, Croatia*
Corresponding author: *Prof. Anko Antabak, MD, Department of Surgery, University Hospital Centre Zagreb, Kišpatićeva 12, 10000 Zagreb, Croatia,*
e-mail: *aantabak@kbc-zagreb.hr*
DOI: *10.5281/zenodo.3517757*

awakenings and other perioperative complications. To what extent obesity affects the postoperative results of surgical treatment is not completely clear and is the subject of research [5]. In addition, some studies have confirmed the cause-and-effect relationship of obesity and the prevalence of injuries in children, as the incidence of obesity and the incidence of injuries increase [6,7]. Obesity in children and its prevalance among surgically treated children, and its impact on treatment outcomes, is a completely new problem, of which there are few studies [8]. It is known that in the USA, among surgically treated children there is the prevalence of increased body weight [9] and we have no knowledge about the prevalance of obese children among surgical patients, nor what trend is.

The aim of this study is to determine the nutritional status of children treated at the Department of Pediatric Surgery (malnutrition, normal body weight, overweight and obesity) in two samples in the time leg of eight years (2010 and 2018). The collected data will be compared with the existing health statistics of the Republic of Croatia.

MATHERIALS AND METHODS

This is a retrospective study of collected data from medical records of children treated at the Department of Pediatric Surgery, Clinical Hospital Centre Zagreb in the period 1.1.-31.12.2010., and 1.1.-31.12.2018. The inclusion criterion is from the age of two to eighteen years of age. Exclusion criteria are secondary causes of obesity and diseases that directly interfere with the growth of children. Children who were hospitalized multiple times for the same diagnosis in the same year were included in the analysis only at the first hospitalization, and when they were hospitalized multiple times with a different diagnosis in the same year, each hospitalization was analysed as a new case. Data were collected on age, gender, height, weight, final diagnosis, surgery (elective or emergency) The examined children were divided into four standardized groups: preschool (2-6 years), school (7-11 years), puberty (12-16 years) and adolescent group (17-19 years). Body mass index was calculated for all subjects, and based on the standard percentile curves of the body mass index by age and sex of the CDC, were classified into four groups: malnourished (below the fifth percentile), normal body mass (between the fifth and eighty-fifth percentiles), increased body weight (from 85 to 95 percentile), obesity (95 percentile and up). In 2010, 1205 children were treated in hospital, 790 children fulfilled the inclusion criteria. In 2018, 1316 children were treated in hospital and 790 children fulfilled the inclusion criteria. The Hospital Information System (HIS) computer database was used to collect the data, and Microsoft Office Excel was used for the table data processing.

RESULTS

The first survey period 2010. The age range was 2-18 years and the average was 9 years. A detailed overview by gender and age group is shown in Table 1.

In the study group of children, 59.5% of children were normal body weight, malnourished 10.8%, and obese 27.7%. In terms of gender, girls are slightly more, but the difference is not statistically significant ($\chi2 = 1,060$). A detailed review of the nutritional status by gender can be seen in Table 2.

The highest prevalence of obese children is in pre-school age (23.8%) and overweight in school age (16.7%).

There are 32.6% of obese preschool children, 36.7% of school children, 28.3% at puberty and 23.9% of adolescents. By analysing the distribution of BMI by age group, the $\chi2$ test is positive for male children ($\chi2 = 45.95$, $p < 0.001$), and it is reliable for male children ($\chi2 = 35.05$, $p < 0.001$), and for female children, because of the small sample, the test was not reliable. There were 609 operatively treated and 181 non-operated. The difference in the prevalence of obesity in the operated and non-operated was not statistically significant ($\chi2 = 1.93$, $p = 0.58$).

The second research period is 2018. The age range was 2-18 years and the average was 9.1 years. A detailed overview by gender and age group is shown in Table 3.

A detailed overview of the nutritional status by gender is presented in Table 4.

The highest prevalence of obesity in school children is 38.9%, in preschool age 31.6%, in puberty 28.3%, and the lowest in adolescents 23.3%. By analysing the distribution of BMI by age group, the $\chi2$ test is positive for male children ($\chi2 = 43.1$, $p < 0.001$), and it is reliable for male children ($\chi2 = 33.0$, $p < 0.001$) and for the female due to the small sample, the test was not reliable.

The biggest prevalence of obese children in the preschool age is (23.9%) and overweight in school age (14.9%). There were 622 surgically treated and non-operated 179. The difference in the prevalence of obesity in the operated and non-operated is not statistically significant ($\chi2 = 1.89$, $p = 0.56$).

DISCUSSION

In 2010, the prevalence of obese children treated in paediatric surgery was 30%. Obese girls are 31.2%, of whom 17.7% are overweight and 13.5% are obese. Obese boys are 29%, of whom 17% are overweight and 12% are obese.

At the eight-year time lag (2018), a second group of surgically treated children was analysed. The apportionment by age and sex is very similar, but also to representation by age groups.

It is interesting that the 2010 and 2018 years the percentage of obese children has no growth trend. For both groups, the apportionment by age is similar, that is, there is no significant difference in the prevalence of

obese children treated at the Department of Paediatric Surgery, KBC Zagreb in 2010 and 2018.

In the general child population, according to the Croatian Statistical Yearbook for 2009, there were 21.5% obese female children, 12.8% overweight and 8.7% obese.3 there were obese male children 24.8%, of which 13.6% were overweight and 11.2% were obese.

By comparison with our results, it is evident that the prevalence of obese children is higher in children treated in paediatric surgery. Similar results are reported by a group of authors (Naifu and associates) who, in an analysis involving 6017 surgically treated children aged 2-18 years in the United States, found that the prevalence of obese children was 31.6% of which, 14.4% increased body weight and 17.2% were obese [9].

According to a study on the nutritional status of patients aged 3-20 years of life at the Clinical Hospital Centre Split, Clinic for Paediatric Diseases, conducted in 2004, on 632 children, there were 7.9% of malnourished children, 73.7% of normal body weight, 74% overweight and 10.9% obese [10]. This study analysed a group of children that is different from our group in terms of demographics, but a smaller incidence of obese children in this study is certainly noticeable. The causes of this difference are not the subject of this study. That same year, a nutrition analysis study was conducted in ten counties (1997-2002), at 4924 students, aged 7-15.

They reported an increased body weight in 11, 2% of boys, and 9.8% of girls, and obese were 5.7% of boys and 5.4% of girls [11]. This means that they recorded 16.9% of obese male and 15, 2% of female children. In the period 2005-2009, 13.6% of boys and 12.8% of girls were overweight, and 11.2% of boys and 8.7% of girls were obese (CIPH 2010). According to research by CIPH, in 2018 as part of the EU survey "European Initiative for Observing the Obesity of EU Children"), in 5664 children, 73.2% of boys were normal weight and 81.7% of girls, while less than 1% of children were malnourished [4].

Overweight boys are 21.3% and obese are 17.2% , and overweight girls are 20.3% and 10.7% are obese. This study was conducted in three HR regions, and the least of the overweight and obese children are in the city of Zagreb.

Therefore, according to the latest survey in the population of school children in the Republic of Croatia, 38.5% of boys and 31.5% of girls are obese. However, from this and Antolić-Degač and associates study from 2004 of a healthy child population in the Republic of Croatia, it seems that the trend of increasing incidence of obese children in the Republic of Croatia is clearly expressed. In the analysis of hospital-treated children, in the paediatric surgery department in the analysis of groups of school children in the 2010 survey, there were 36.7% obese children and in 2018 little more, 38.9%. This is a statistically insignificant increase, i.e. there is no trend of a significant increase in the prevalence of obese surgically treated children. However, when comparing the prevalence of obese children in the general population and surgically treated children, the prevalence of surgically treated children remains higher, although the methodology for research and data collection is not identical. Therefore, this comparison should be taken with a limit. Overall, we can conclude that the proportion of obese children is higher in surgically treated children, but the reasons for this were not analysed in this paper.

CONCLUSION

The prevelance of obesity in children undergoing hospitalization for surgical illness is higher than in the general population. There was no trend of increasing the prevelance of obesity over a 10-year time lag.

CONFLICT OF INTEREST:

The authors declare that there is no conflict of interest.

REFERENCES:

1. *Musić Milanović S, Bukal D. Epidemiologija debljine-javnozdravstveni problem.Medicus 2019;27:7-13.*
2. *Devaux m, Sassi F. Social inequalities in obesity and owereight in 110 ECD countries. Eur J Pub Healt 2013;23:464-9.*
3. *Hrvatski zdravstveno-statistički ljetopis. HZJZ. Ur. Baklaić Ž, Zagreb, 2010. https://hzjz.hr/wp-content/uploads/2013/11/Ljetopis_2009.pdf*
4. *Europska incijativa praćenja debljine u djece. Ur. Capak K. HZJZ, Zagreb, 2018.*
5. *Nafiu OO, Reynolds PI, Bamgbade OA, Tremper KK, Welch K, Kasa-Vubu JZ. Childhood body mass index and perioperative complications. Paediatr Anaesth 2007;17(5):426-30.*
6. *Cordelie E, Saman S, Avery B at al. Obesity in pediatric trauma. J Pediatr Surg. 2017;52:628-32.*
7. *Annette A, Jeffry K, Krikor D at al. Associations between childhood obesity and upper and lower extremity injuries. Inj Prev. 2013;19:191-7.*
8. *Brian P. Blackwood, Colin D et al. Overweight and Obese Pediatric Patients Have an Increased Risk of Developing a Surgical Site Infection. Surg Infect (Larchmt). 2017;18(4):491-497.*
9. *Nafiu OO, Ndao-Brumlay KS, Bamgbade OA, Morris M, Kasa-Vubu JZ. Prevalence of overweight and obesity in a U.S. pediatric surgical population.J Natl Med Assoc. 2007;99(1):46-8, 50-1.*
10. *Dropulić N, Meštrović J, Krželj V, Šonjić M, Krcatović D. Uhranjenost boelsnikahospitaliziranih na klinici za dječje boelsti KBC Split. Pediatr Croat. 2006;50:173-77.*
11. *Antolić-Degač K, Raić-Rak A, Mesaroš-kanjski E, Petrović Z, Capak K. Stanje uhranjenosti i prehrambene naviske školske djece u RH. Pediatr Croat.2004;48:9-15.*

Table 1. Age and gender apportionment of children, 2010

		Male	Female	Total
PARTICIPANTS	n	524	266	790
	%	66,3	33,7	100
AGE GROUPS				
Preschool	n	191	83	274
	%	36,5	32,3	31,5
School	n	173	53	226
	%	33	19,9	28,6
Pubertal	n	149	103	252
	%	28,4	38,7	31,9
Adolescent	n	11	24	35
	%	2,4	9	4,4

Table 2. Children nutrition status, 2010

		Male	Female	Total
Malnourished	n	60	25	85
	%	11,5	9,4	10,8
Normal body weight	n	312	158	470
	%	59,5	59,4	59,5
Overweight	n	63	36	99
	%	12	13,5	12,5
Obese	n	89	47	136
	%	17	17,7	17,2
TOTAL	n	524	266	790
	%	100	100	100

Table 3. Age and gender apportionment of children, 2018

		Male	Female	Total
PARTICIPANTS	n	530	260	790
	%	66,6	33,3	100
AGE GROUPS				
Preschool	n	192	83	274
	%	36,6	32,1	31,5
School	n	176	51	227
	%	34	19,2	28,6
Pubertaln		151	100	251
	%	28,6	38,5	31,9
Adolescent	n	14	23	37
	%	2,5	8,9	3,9

Table 4. Nutritional status of children by gender, 2018

		Male	Female	Total
Malnourished	n	65	20	85
	%	9,5	8,4	8,8
Normal body weight	n	322	158	470
	%	60,5	59,4	59,5
Overweight	n	60	36	96
	%	12	13,5	12,5
Obese	n	92	47	136
	%	18	17,7	17,2
TOTAL	n	530	260	790
	%	100	100	100

Acta Chir Croat 2019; 16: 11-15

SURGICAL TREATMENT OF PERIHILAR CHOLANGIOCARCINOMA: 10-YEAR EXPERIENCE AT A SINGLE INSTITUTION

Ivan Romić, Igor Petrović, Ante Zvonimir Golem, Emil Kinda, Tihomir Kekez, Goran Augustin, Hrvoje Silovski, Tomislav Bruketa, Trpimir Morić, Željko Jelinčić

ABSTRACT

Background: Our study evaluates surgical outcomes of patients treated for perihilar cholangiocarcinoma in a single institution and demonstrates postoperative (90 days) morbidity and mortality rates and potential prognostic factors associated with complications.

Methods: Medical records of all patients with a diagnosis of perihilar cholangiocarcinoma (pCC) between 2007 and 2017 who underwent a surgical procedure at the University hospital centre Zagreb, were retrospectively evaluated. Statistical analysis to determine predictors of postoperative mortality was performed using the Chi-square test and Fisher exact probability test where appropriate.

Results: Out of 52 surgically treated patients, 43 underwent radical and 9 palliative procedures. Hilar resection and hilar resection along with right hepatectomy were the most commonly performed procedures in 34 radically treated patients. Overall morbidity and mortality rates were 46% and 5.7%, respectively. Significantly higher morbidity rate was observed in a group of patient with untreated preoperative jaundice and in those aged 70 and over.

Conclusion: Current guidelines favor extension of radicality in treatment of pCC by performing left or right hepatectomy in addition to hilar resection. This may increase R0 resection rates and prolong disease free survival. Our experience shows similar mortality/morbidity rates as reported in other centers and confirms that in selected patients, concomitant hepatectomy for perihilar pCC is a safe and feasible surgical strategy.

Keywords:

Perihilar cholangiocarcinoma, surgical outcome, survival, Klatskin tumor

Introduction

Perihilar cholangicarcinoma (pHCC) is the second most common primary cancer of the liver with a generally poor prognosis and it represents a difficult challenge for hepatobilliary surgeons not just because of aggressive nature of the cancer, but also because of variable anatomical relations of hillar structures. Historically, the surgery for this cancer was characterized by high mortality rates but also surgeons were confronted with a low resectability rate (up to to 50%) and high recurrence rates (50% to 70%) [1,2].

However, over the last decade, studies suggest some progress in treatment and slight improvement in outcome that have mainly derived from a better understanding of the tumor spread and improved technique of liver parenchymal transection [3].

In addition, it can be also explained by wider indications and adopting more radical surgical procedures that include standard or extended hepatectomies along with resection of extrahepatic biliary ducts and portal vein/hepatic artery if necessary.

Material and Methods

Our paper is a review of 10 years' experience with 574 resections for perihilar cholangio- carcinoma in a tertiary institution. Medical records of all patients with a diagnosis of perihilar cholangiocarcinoma (pCC) between 2007 and 2017 who underwent surgical procedure at our surgical department were retrospectively evaluated.

We demonstrate demographic and baseline patient characteristics, intraoperative factors and outcome in terms of morbidity mortality and 1 year disease free survival. Results are expressed as the medians, range and proportions. Statistical analysis to determine predictors of postoperative mortality was performed using the Chi-square test and Fisher exact probability test where appropriate.

Results

Baseline characteristics of patients are shown in Table 1. Out of 52 patients, 31 were men and the median age was 69 years. In the majority of cases (28) preoperative endoscopic stenting was placed to manage obstructive jaundice. Resection procedures comprised 83% of cases (34 R0, 7 R1 and 2 R2 resections) and 9 included exploration laparotomy and palliative procedure only due to distant metastasis or advanced local disease (Figure 1.)

Department of Surgery, University Hospital Centre Zagreb, Croatia
Corresponding author: *Ivan Romić, MD, Department of Surgery, University Hospital Centre Zagreb, Kišpatićeva 12, 10000 Zagreb, Croatia,*
e-mail: *i.romic@gmail.com*
DOI: *10.5281/zenodo.3517763*

Bile duct resection alone (hilar resection) was performed in 17 and bile duct resection coupled with right hepatectomy in 12 patient. Other types of hepatectomies, left and extended right, were less commonly done, in 3 and 2 cases, respectively. Portal vein resection and reconstruction was required in 4 cases to achieve adequate radicality. Other procedures included resections of primary tumor and metastasectomy (4 cases), pancreaticoduodenectomy (2 cases), partial gastrectomy and transverse colon resection. In resectable cases, a bilio-digestive anastomosis was constructed by means of hepatico-jejunostomy in 37, and choledocho-jejunostomy in 6 cases. Intraoperative characteristics and outcomes are demonstrated in Table 2. Median operative time was 420 minutes and average blood loss 800ml. Hospital stay ranged from 8 to 51 days with a median of 19 days.

Overall morbidity rate was 46% and posthepatectomy liver failure according to the 50:50 definition was the most common complication followed by intraabdominal sepsis and bile leakage. No intraoperative death was seen and three patients died of complications in a postoperative period (within 90 days from the day of surgery).

Liver failure associated with infectious complications was cause of death in all cases.

We conducted comparative univariate study which identified prognostic factors for severe (grade III or more) postoperative morbidity: age over 70 years, and preoperative jaundice that both showed significant association with outcome. Additional hepatectomy resulted in higher, although not statistically significant, morbidity rate while other variables were not associated with morbidity (Table 3).

Discussion

Despite certain advances in chemotherapy, pCC remains a disease which can be cured only by radical surgical procedure. In the last 2 decades, there was a progress in treatment of pCC which can be attributed to significant decrease in surgical morbidity/mortality and expanded indications for radical procedure [4].

Our results presented here are comparable to those reported in literature and we adopted most recent recommendations in this field of surgery[5]. Thereby, we performed surgery in cases which were considered unresectable 10 years ago. This includes vascular infiltration and cases where combined resections of pancreas, stomach, liver or transverse colon are required.

Most authors agree that the goal of surgical treatment of pCC should be an R0 resection, therefore, radical intervention for type 3 and 4 tumors should include resection of the bile duct with lymphadenectomy of the hepatic hilum and right/left hepatectomy including the caudate lobe. However, treatment of type 1 or 2 tumors is more controversial and most authors suggest hilar resection alone, while the minority of them favor to add right hepatectomy to ensure adequate radicality[6].

In our research, over the time period studied, there was an increasing trend of performing a hilar resection coupled with hepatectomy especially when last 3 years are compared to the period between 2007 and 2009. Such a trend was reported in other research papers as well, and may be explained by the increasing number of evidence that hepatectomy improves R0 resection rates and yields better oncological outcomes compared to hilar resection alone. In addition, our results as well as relevant literature show that hepatectomy does not have significant negative effect on morbidity and mortality rates[7].

We want to highlight that rate of unresectable disease discovered at laparotomy was low (8%). This confirms evolution of more accurate preoperative investigation such as PET scan, EUS and ERCP. Furthermore, in all unresectable cases, some type of palliative procedure was done.

Liver transplantation for pCC was not performed in our institution and this is still under scientific and professional evaluation, but some excellent results have been reported [8]. Preoperative stent insertion is controversial since some authors argue that it may lead to unacceptable risk of cholangitis. However, we adopt the stance to manage biliary obstruction prior to surgery if technical and organizational capabilities allow it.

Conclusions

In conclusion, surgical treatment for pCC has been evolving and improved surgical technique led to expanded surgical indications, satisfactory morbidity/mortality rates and promising oncologic outcome. The most important goal is to achieve R0 resection and preserve sufficient future liver volume. Considering relatively low incidence of pCC, future studies on larger number of patients are required to establish clear guidelines in management of this malignancy.

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.

REFERENCES:

1. *Zhang W, Yan LN. Perihilar cholangiocarcinoma: Current therapy. World J Gastrointest Pathophysiol. 2014;5(3):344-54.*
2. *Jarnagin WR, Fong Y, DeMatteo RP, Gonen M, Burke EC, Bodniewicz BS J, Youssef BA M, Klimstra D, Blumgart LH. Staging, resectability, and outcome in 225 patients with perihilar cholangiocarcinoma. Ann Surg. 2001;234:507–517; discussion 517-519.*
3. *Ramos E. Principles of surgical resection in perihilar cholangiocarcinoma. World J Gastrointest Oncol. 2013;5:139–146.*
4. *Kosuge T, Yamamoto J, Shimada K, Yamasaki S, Makuuchi M. Improved surgical results for perihilar cholangiocarcinoma with procedures including major hepatic resection. Ann Surg. 1999;230:663–671.*
5. *Hu HJ, Mao H, Shrestha A, et al. Prognostic factors and long-term outcomes of perihilar cholangiocarcinoma: A single-institution experience in China. World J Gastroenterol. 2016;22(8):2601-10.*
6. *Kow AW, Wook CD, Song SC, Kim WS, Kim MJ, Park HJ, Heo JS, Choi SH. Role of caudate lobectomy in type III A and III B perihilar cholangiocarcinoma: a 15-year experience in a tertiary institution. World J Surg. 2012;36:1112–1121.*
7. *Lillemoe KD, Cameron JL. Surgery for perihilar cholangiocarcinoma: the Johns Hopkins approach. J Hepatobiliary Pancreat Surg. 2000;7:115–121.*
8. *Mantel HT, Westerkamp AC, Adam R, et al. Strict Selection Alone of Patients Undergoing Liver Transplantation for Perihilar Cholangiocarcinoma Is Associated with Improved Survival. PLoS One. 2016;11(6):e0156127. Published 2016 Jun 8. doi:10.1371/journal.pone.0156127*

Patient, disease and procedural characteristics	
Patient Characteristics	All patients (n=52)
Sex (M/F)	31/21
Age (median, range), years	69 (31-82)
BMI, kg/m2 (median, SD)	32.6 (±3.3)
Child-Pugh (A/B)	44/8
ASA score, median	2
Cirrhosis (yes/no)	5/47
Preoperative biliary stent (yes/no)	28/24
Disease characteristics	All patients (n=48)
Preoperative diagnosis(yes/no)*	41/11
Type of tumor **	
- Type I	5 (9.6%)
- Type II	10 (19.2%)
- Type III	23 (44%)
- Type IV	14 (26.4%)
Curative/Palliative	43/9 (83%/17%)
Type of resection	
R0	34
R1	7
R2	2
Palliative	9
**Preoperative pathohistological confirmation*	
*** According to Bismuth classification*	

Table 1. Baseline demographic and disease data

Operative and outcome results		
	All patients (N=52)	
Type of procedure		
- Hilar resection	17	
- Hilar resection+ right hepatectomy	12	
- Hilar resection+ left hepatectomy	3	
- Hilar resection+ right hepatectomy	2	
- Palliative bilio-digestive bypass	9	
- Combined portal vein resections	4	
- Other	8	
Intraoperative factors	**All patients (N=52)**	
Operative time (median, min)	420/±112	
Estimated blood loss (ml)	800±180	
ICU stay (median, range) days	6 (3-42)	
Hospital stay (median, range) days	19 (8-51)	
Outcomes		
90 days mortality	3(5.7%)	
Postoperative morbidity*	24(46%)	
1 year disease free survival	38 (74%)	
Causes of postoperative morbidity	N=24	
- Liver failure	11	
- Septic complications	7	
- Hemorrhage	2	
- Bile leakage	2	
- Other	2	
Trends in additional hepatectomy**		p value
-2007-2012	4/23	0.08
-2012-2017	13/25	
**According to Clavien-Dindo classification grade >III*		
*** Prevalence of hepatectomy in two consecutive*		
5-year periods for resectable cases		

Table 2. Intraoperative factors and outcome

Predictors of severe morbidity			
Variable	No.patients	Morbidity (%)	*P value*
Age,years			*0.006*
<70	27	8 (29.6)	
>70	25	16 (64)	
Preoperative jaundice			*0.02*
yes	28	17 (60)	
no	24	7 (29)	
Concomitant hepatectomy			*0.72*
yes	17	11 (64.7)	
no	17	9 (52.9)	
Extent of liver resection			*0.9*
<50%	11	7 (64)	
>50%	6	4 (66)	
Blood loss (ml)			*0.82*
<800	29	13 (56)	
>800	23	11 (47)	

Table 3. Univariate analysis of predictors of severe postoperative morbidity

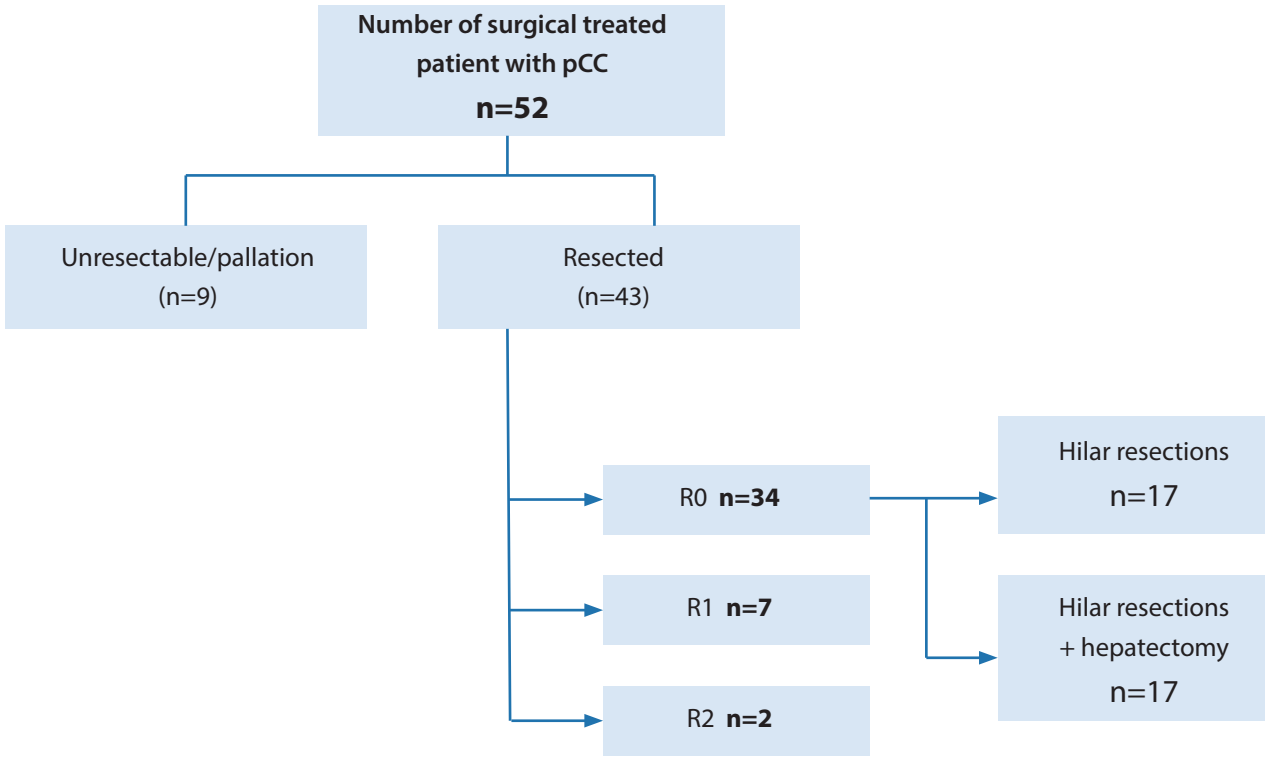


Figure 1. Flowchart of the study

Acta Chir Croat 2019; 16: 17-19

MALIGNANT TRANSFORMATION OF GERM CELL TUMOR WITH TERATOMATOUS COMPONENT INTO ADVANCED RETROPERITONEAL SARCOMA - CASE REPORT AND LITERATURE REVIEW

Igor Petrović[1], Ivan Romić[1], Goran Pavlek[1], Ana Ettinger[2], Mirna Bajt[3], Tonko Čolić[3]

ABSTRACT

Background: A late-relapse germ cell tumor (GCT) might contain malignant non-germ cell tumor cells, known as „somatic-type malignancy" (SM). Development of secondary SM is extremely rare and occurs in only 1% of patients with GCT.

Case study: We present the case of a 42-year-old patient who developed retroperitoneal tumor with duodenal and right colon involvement 6 years after he underwent left orchidectomy for stage IIC mixed GCT (95% seminoma, 5% teratoma). Since the tumor markers for germline tumor were normal, another type of tumor was highly suspected, most likely a malignant alteration of the residual teratoma. Tumor was completely removed and pathological report suggested undifferentiated sarcoma.

Conclusion: R0 resection and postoperative patient follow-up by the protocol for at least 6 years after orhidectomy is the key to successful treatment of GCT. Malignant tumor transformations are rare, but still possible so clinicians should be aware of the importance of frequent and adequate patient monitoring.

Keywords:
sarcoma, retroperitoneal, germ cell tumor

INTRODUCTION

Testicle is an organ with double function consisting of embryonic and supporting cells. The process of spermatogenesis emerges from the germ cells, while the supporting (Leydig) cells represent an endocrine part of the organ which releases male sex hormone - testosterone. While tumors can originate from both types of cells, tumors of germinal epithelium are far more common [1].

Germ cell tumors represent 95% of all testicular tumors, and are among the most common tumors of young men aged 15 to 34 years. Their incidence has increased in the last 30 years, especially in industrialized countries. Multidisciplinary approach, advance in diagnostic methods, surgical technique and chemotherapy have led to increased survival rate of more than 90% and 20 years ago, that value was mortality rate of this malignancy [2-4].

Seminomas have the best prognosis, while prognosis of non-seminomatous tumors depends on the presence and type of its different components-embryonic carcinoma, teratoma, yolk-sack tumors, choriocarcinoma, and can also be mixed GCT [5,6].

Despite appropriate treatment, 10 to 30% of patients may experience relapse within 2 years after orhidectomy. A very late relapse (over 5 years following orhidectomy) happens even more rarely, in about only 1% of cases, but such tumors often contain malignant cells of non-germinal type (somatic-type malignancy, SM). It is possible that malignant transformation arises either from the pre-existing teratomatous component of the tumor, or from the totipotent germ cell. The most common type of SM associated with GCT is the sarcoma in more than 50% of cases, with rhabdomyosarcoma being its most common subtype. It is followed by neuroectodermal tumors, adenocarcinomas and undifferentiated sarcomas [7-9].

Sarcomas are a heterogeneous group of rare solid tumors of mesenchymal origin with specific clinical and pathological features. They are usually divided into two major categories - soft tissue sarcomas (fat, muscle, nerve and nerve envelopes, blood vessels and other connective tissues) and sarcomas of the bone. Anatomical localization of the tumor process is a very important variable that affects the choice of treatment method and outcome of the disease [7,9].

Accordingly, the NCCN soft tissue sarcoma (STS) guidelines are classified into the following subcategories: STS of extremities, surface / placed on the trunk, head or neck; retroperitoneal or intraabdominal STS; GIST; desmoid tumors (aggressive fibromatosis); rhabdomyosarcomas. STSs most commonly metastasize into the lungs, while abdominal STSs are mainly conveyed in the liver and peritoneum [10].

Due to the rare occurrence and complexity of this condition, an optimal treatment strategy has not yet

[1]*Department of Surgery, University Hospital Centre Zagreb, Croatia*
[2]*Department of Surgery, General Hospital Varaždin, Croatia*
[3]*School of Medicine, University of Zagreb, Croatia*
Corresponding author: *Ivan Romić, MD, Department of Surgery, University Hospital Centre Zagreb, Kišpatićeva 12, 10000 Zagreb, Croatia,*
e-mail: *i.romic@gmail.com*
DOI: *10.5281/zenodo.3517765*

been established. For this reason it is important to involve multidisciplinary teams of experts in treating STS and follow evidence-based recommendations.

CASE STUDY

A 42-year-old patient has been hospitalized at the Abdominal Surgery Department because of epigastric pain, anorexia, nausea and vomit soon after meal intake. In addition, he had insomnia and night sweating. He had a history of stage IIC mixed testicular carcinoma (95% seminoma, 5% teratoma) which was treated in 2012. He underwent left-sided orhidectomy and had 4 cycles of adjuvant chemotherapy by BEP protocol (bleomycin / etoposide / cis-platinum), followed by retroperitoneal lymph node dissection. Due to iatrogenic urethral injury, left nephrectomy was performed in the same procedure. Thereafter, the patient did not show up for regular urological and oncological follow-ups.

An abdominal CT revealed a large retroperitoneal tumor with compression and partial obstruction of the duodenum. Since tumor markers for germinative tumor were normal, another type of tumor was highly suspected, most likely a malignant alteration of residual teratoma.

Surgery was indicated and included complete extirpation of the retroperitoneal tumor, partial duodenal and jejunal resection with terminolateral duodenojejunal anastomosis, left hemicolectomy and apendectomy. Histopathological suggested undifferentiated sarcoma.

Postoperative course was complicated with infected intraabdominal collection *(E.fecalis, C. albicans and C. Dubliniensis),* increased inflammatory parameters and elevated liver enzymes (AF 436; GGT 690; AA 161). Antibiotic and supportive therapy with percutaneous drainage of collections was done and patient's condition improved and was discharged on postoperative day 24.

DISCUSSION

Malignant transformation of germ cell tumors into somatic malignancy is rare clinical scenario and sarcomas are the most commonly reported type of malignancy observed in GCTs. Hypothesis regarding the origin of malignant transformation suggests that the sarcoma may arise from "malignant transformation" of teratomatous mesenchyme [11,13].

Prior to selecting the treatment method for STS, it is necessary to do a biopsy and set a pathohistological diagnosis. The correct diagnosis is very important in primary testicular tumors as well as in the metastases since it will allow appropriate therapy. The most frequently selected method of treatment is the surgical resection of the tumor and all affected structures, and it is of utmost importance that the resection margins are free of malignant tissue. This is the only potentially curative option for patients with retroperitoneal / intraabdominal STS [11,12].

Patients with stage I disease have good prognosis, but in metastatic disease it is generally poor. Radical surgery is the only curative modality, although there have been a couple of findings on achieving promising results with Doxorubicin-based chemotherapy for transformed tumour component [13].

A large study on 500 subjects showed median survival of 103 months for complete resection compared to 18 months for incomplete resection. Radiotherapy can be used as a preoperative procedure in resectable tumors, or as a primary treatment for non-curable ones, but is not appropriate substitute for radical surgical resection [14].

A deeper understanding of the biology of this phenomenon is essential for clinicians who are involved in such malignancies in order to improve treatment outcomes.

CONCLUSION

R0 resection and postoperative patient follow-up by the protocol for at least 6 years after orhidectomy is the key to successful treatment of GCT. Malignant tumor transformations are rare, but still possible so clinicians should be aware of the importance of frequent and adequate patient monitoring.

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.

REFERENCES:

1. *Kim I, Young RH, Scully RE. Leydig cell tumors of the testis. A clinicopathological analysis of 40 cases and review of the literature. Am J Surg Pathol. 1985;9(3):177-92.*
2. *Fukawa T, Kanayama HO. Current knowledge of risk factors for testicular germ cell tumors. Int J Urol. 2018;25(4):337-344.*
3. *Durand X, Fléchon A, Murez T, et al. CCAFU french national guidelines 2016-2018 on testicular germ cell tumors. Prog Urol. 2016;(Suppl 1):S147–65.*
4. *Groll RJ, Warde P, Jewett MA. A comprehensive systematic review of testicular germ cell tumor surveillance. Crit Rev Oncol Hematol. 2007;64:182–97.*
5. *Cools M, Looijenga L. Update on the Pathophysiology and Risk Factors for the Development of Malignant Testicular Germ Cell Tumors in Complete Androgen Insensitivity Syndrome. Sex Dev. 2017;11(4):175-181.*
6. *R.J.Motzer, A. Amsterdam, V. Prieto et al., Teratoma with malignant transformation: various malignant histologies emerging in men with germ cell tumors, Journal of Urology, 1998; 159(1): 133-8.*
7. *Ahmed T, Bosl GJ, Hajdu SI. Teratoma with malignant transformation in germ cell tumors in men. Cancer. 1985;56(4):860-3.*
8. *Mikuz G, Colecchia M. Teratoma with somatic-type malignant components of the testis. A review and an update. Virchows Arch. 2012;461(1):27-32.*
9. *Ulbright TM. Germ cell tumors of the gonads: a selective review emphasizing problems in differential diagnosis, newly appreciated, and controversial issues. Mod Pathol. 2005;18 Suppl 2:S61-79.*

10. *Von mehren M, Randall RL, Benjamin RS, et al. Soft Tissue Sarcoma, Version 2.2016, NCCN Clinical Practice Guidelines in Oncology. J Natl Compr Canc Netw. 2016;14(6):758-86.*
11. *Ulbright TM, Loehrer PJ, Roth LM et al. The development of non-germ cell malignancies within germ cell tumors. A clinicopathologic study of 11 cases. Cancer, 1984; 54:1824–1833.*
12. *Guo CC, Punar M, Contreras AL, et al. Testicular germ cell tumors with sarcomatous components: an analysis of 33 cases. Am J Surg Pathol. 2009;33(8):1173–1178.*
13. *Malagón HD, Valdez AM, Moran CA, et al. Germ cell tumors with sarcomatous components: a clinicopathologic and immunohistochemical study of 46 cases. Am J Surg Pathol. 2007;31:1356–1362.*
14. *Michael H, Lucia J, Foster RS, et al. The pathology of late recurrence of testicular germ cell tumors. Am J Surg Pathol. 2000;24:257–273.*

FIGURES AND CAPTIONS

Figure 1. Abdominal CT scan showing large retroperitoneal tumour mass which caused duodenal obstruction

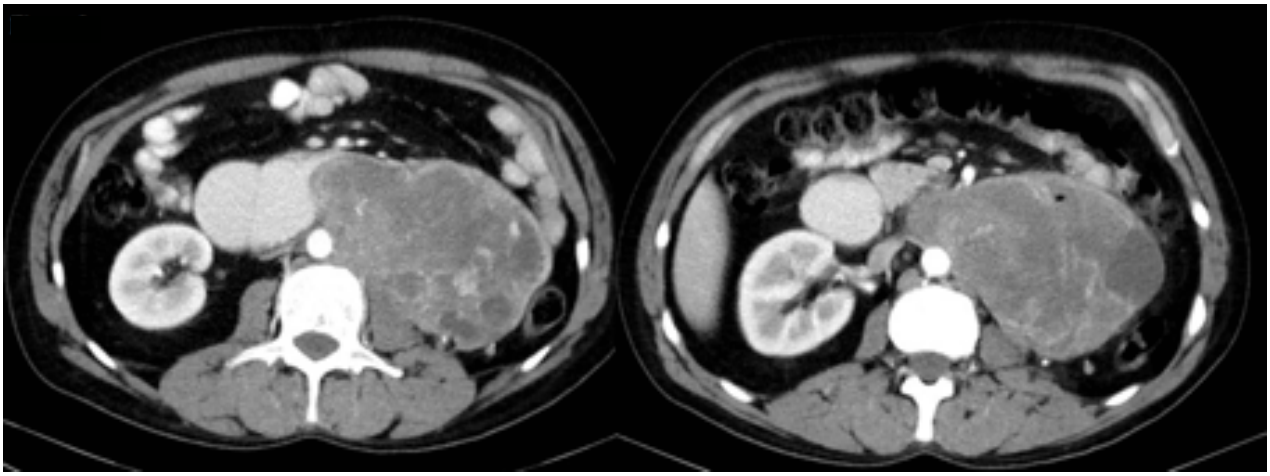


Figure 2. Surgical specimen after "en block" resection of tumor, fourth segment of duodenum and proximal part of jejunum

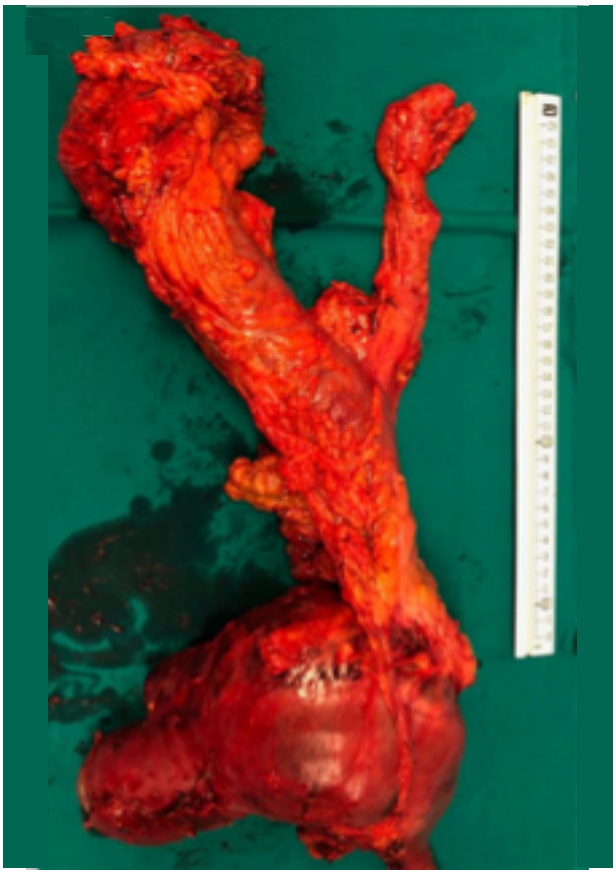
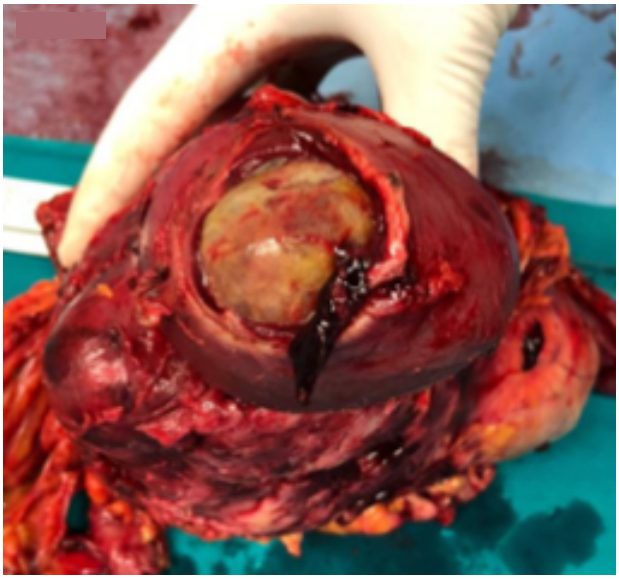


Figure 3. Tumor as seen after the fibrous capsule was partially removed



Acta Chir Croat 2019; 16: 21-23

STUMP APPENDICITIS AFTER LAPAROSCOPIC APPENDECTOMY: A RARE CLINICAL ENTITY

Asmir Jonuzi, Nusret Popović, Zlatan Zvizdić, Emir Milišić

ABSTRACT

Background: Stump appendicitis is an acute inflammation of the residual part of the appendix and a rare complication of incomplete appendectomy. This is a rare delayed complication after appendectomy with the reported incidence of 1 in 50,000 cases. Clinically it can be presented as acute abdomen and presents a diagnostic dilemma. Prompt recognition is important to lead to an early treatment, thus avoiding serious complications.

Casestudy: We presenta 14-year-old girl with diagnosis of stump appendicitis, who underwent surgical treatment (open appendectomy) after having laparoscopic appendectomy a month before. Radiologically (UZV and CT scan) was diagnosed an inflammatory mass with abscess dimension 41 x 21 mm in the right iliac fossa. During operation a 1,5 cm-diameter appendiceal stump was noted in the anatomical region of the appendix. The appendiceal stump was resected and inverted into the cecal wall. Histopathology examination showed acute inflammation and patchy necrosis of the appendiceal stump. The post-operative course was uneventful. Patient was discharged on third post-operative day.

Conclusion: Stump appendicitis is a rare but serious complication of appendectomy. The prevalence and incidence of stump appendicitis has been increasing in the recent years. Clinical presentation of stump appendicitis mimics symptoms and signs of acute appendicitis or acute abdomen and with a previous appendectomy. So it must be considered in the differential diagnosis of acute abdomen despite the patient's open or especially laparoscopic appendectomy history.

Keywords:
open appendectomy, stump appendicitis, acute abdomen.

INTRODUCTION

Stump appendicitis is the inflammation of the residual appendiceal tissue after an appendectomy. It is a rare complication with a frequency that is underreported as well as underestimated. Therefore, physicians as well as surgeons need to be aware of this clinical entity and not to assume that previous appendectomy precludes recurrent/stump appendicitis. Failure to recognize this possibility may lead to delay in treatment and may result in complications such as perforation, abscess formation, and sepsis. Aim of article was to present case of stump appendicitis in 14-year-old girl, 1 month following laparoscopic appendectomy.

CASE STUDY

A 14-year-old girl was admitted with a complaint of right lower quadrant pain which lasted for 24 hours with vomiting and fever. Patient had undergone laparoscopic appendectomy 1 month back and had a recovery without noted problems. Positive clinical findings included a right lower quadrant tenderness and leukocytosis i.e. 14x109/L with 78% neutrophils and CRP 125,7 mg/L. Electrolytes, urinalysis, transaminases and amylase were all in the normal range. Vital signs were normal. Plain abdominal X-ray was normal. Abdominal ultrasound and abdominal computerized tomographic scan-CT (Figure 1.) revealed inflammatory, intraperitoneal collection at the right iliac fossa (radiologically was diagnosed as an inflammatory mass with abscess dimension 41 x 21 mm). IV fluid with parenteral antibiotics was included in therapy. The presumptive preoperative diagnosis was stump appendicitis. During operation a 1,5 cm-diameter appendiceal stump was noted in the anatomical region of the appendix (Figure 2.). The appendiceal stump was resected and inverted into the cecal wall with 2-0 vicryl suture. Histopathology examination showed acute inflammation and patchy necrosis of the appendiceal stump. The post-operative course went without problems. Patient was discharged on third post-operative day.

DISCUSSION

Stump appendicitis is the re-inflammation of the residual appendiceal tissue after an appendectomy. It represents a rare delayed complication of appendectomy which is unknown to most clinicians [1,2,3]. Appendectomy is one of the most commonly performed surgical emergencies. Claudius Amyand in 1735, performed the

Clinic of Pediatric surgery, Clinical Center University of Sarajevo, Bosnia and Herzegovina
Corresponding author: *Asmir Jonuzi, MD, MSc, Clinic of Pediatric surgery, Clinical Center University of Sarajevo, Patriotske lige 81, 71000 Sarajevo, Bosnia and Herzegovina,*
e-mail: *jonuziasmir@hotmail.com*
DOI: *10.5281/zenodo.3517770*

first appendectomy and Reginald Fitz in 1886, described the clinical features and pathological abnormalities of appendicitis. In 1945, Rose was the first to describe stump appendicitis in two patients who had undergone appendectomy for acute appendicitis in past [1].

The appendix arises from the postero-medial wall of cecum about 3 cm below the ileocecal valve. Its variable position and subserous length, combined with acute inflammation, may result in misidentification of the appendicio-cecal junction. Dissecting the recurrent branch of the appendiceal artery and following the teniae coli on the cecum helps in identifying the true appendicular base. Generally, an appendix stump shorter than 5 mm reduces the risk of stump appendicitis [2,3]. Moreover, the incidence of this complication seems to increase until the introduction of laparoscopic approach, probably due to absence of tactile feedback [4,5]. The time of onset ranges from 2 weeks to decades after appendectomy [6].

Incomplete appendectomy leaving a stump longer than 5 mm, severe local inflammation preventing adequate identification of the appendiceal base, retrocecal or subserosal appendix, and, last but not least, the insufficient experience of the surgeon may influence the occurrence of this condition [7-9]. Moreover some sources have suggested that the growing use of laparoscopic appendectomy may increase the frequency of stump appendicitis. This may be the result of leaving a longer stump, secondary to a smaller field of vision, lack of three-dimensional perspective, and the absence of tactile feedback [10,11]. The incidence of stump appendicitis can be minimized simply by adequately visualizing the base of the appendix and creating a stump less than 3 mm in length. Therefore, if performed properly, there is no reason why laparoscopic appendectomy should lead to a higher incidence of stump appendicitis.

Clinical features of stump appendicitis do not differ from that of acute appendicitis, even though the history of a previous appendectomy can be misleading causing a delay in diagnosis. Ultrasonography can be useful in identifying inflammatory changes. The abdominal CT is the gold standard for the diagnosis and should be performed in patients with right lower quadrant symptoms after appendectomy. It allows the recognition of retained fecolith or postoperative abscess as well as cases of stump appendicitis [1]. Stump appendicitis has a higher risk of complications with perforation being reported in nearly 70% of the cases. It is therefore imperative that patients with stump appendicitis undergo complete appendectomy as soon as possible. Stump appendicitis should be considered in any patient with a previous history of appendectomy and history of acute appendicitis. Treatment by re-surgery and complete removal of the appendix will resolve the problem.

Stump appendicitis is one of the rare delayed complications after appendectomy with the reported incidence of 1 in 50,000 cases [12]. Prompt recognition is important to lead to an early treatment, thus avoiding serious complications like wound infection, intra abdominal abscess, and intestinal perforation with peritonitis, bleeding, and adhesions with sub-acute intestinal obstructions [13].

Stump appendicitis can represent a diagnostic dilemma if the treating physician is unfamiliar with this uncommon clinical entity. Clinically, patients present with sign and symptoms mimicking appendicitis or acute abdomen along with a previous history of appendectomy as seen in our case. The presence of an appendectomy scar does not rule out the possibility of stump appendicitis [14]. The incidence and prevalence of stump appendicitis has been increasing in the recent years. It has been reported following both open and laparoscopic appendectomy [4,15]. It has been suggested that no appendicular stump longer than 3mm should be left behind [16].

The common conditions leading to stump appendicitis have been broadly classified under the anatomical and surgically related factors. One common denominator is the inappropriate indentification of the appendicular base i.e appendicular-cecal junction. The anatomically related factors may be a retrocecal or subserous appendix or a duplicated appendix, a rare developmental abnormality seen in about 0.004% in appendectomy patients [17]. The surgical factors predisposing for stump appendicitis may be inadequate indentification of the appendicular base because of severe local inflammation, leaving long stump due to fear of cecal injury or difficult dissection and local ulcerations due to fecoliths [18]. The stump appendicitis has been reported following open appendectomy with stump ligation, open appendectomy with stump inversion, and laparoscopic appendectomy where appendiceal stump is either closed with an endoloop or by stapling. Both the surgical techniques i.e. inversion of stump or simple ligation of stump cannot prevent the possibility of stump appendicitis [19].

Radiological evaluation by ultrasound and computed tomography (CT Scan) helps in the preoperative diagnosis of stump appendicitis [6,20]. CT scan of the abdomen is more specific than ultrasound for the accurate pre-operative diagnosis of stump appendicitis because it excludes other etiologies of acute abdomen. CT findings may be similar to those seen in acute appendicitis. They include pericecal inflammatory changes, abscess formation, fluid in the right paracolic gutter, cecal wall thickening, and an ileocecal mass. In the era of laparoscopy a diagnostic laparoscopy may prove to be the next diagnostic and therapeutic option in case of ambiguity [21].

Completion appendectomy either by open or by laparoscopic intervention is the treatment of choice for stump appendicitis [22]. Very rarely, extensive surgery such as ileocolic resection may be necessary if there is significant inflammation around the ileocecal region. It is imperative to adequately visualize the appendicular base and the ileocecal region to ensure that a stump not more than 5 mm remains after appendix removed [13,17].

CONCLUSION

Stump appendicitis is a rare but serious complication of appendectomy. The prevalence and incidence of stump appendicitis has been increasing in the recent years. Clinical presentation of stump appendicitis mimics symptoms and signs of acute appendicitis or acute abdomen and with a previous appendectomy. So it must be considered in the differential diagnosis of acute abdomen despite the patient's open or especially laparoscopic appendectomy history. The diagnosis is often missed or delayed if the clinician is unaware of this rare clinical entity. Clinical awareness and a high level of suspicion would prevent unnecessary delay in initiating treatment thus avoiding serious complications.

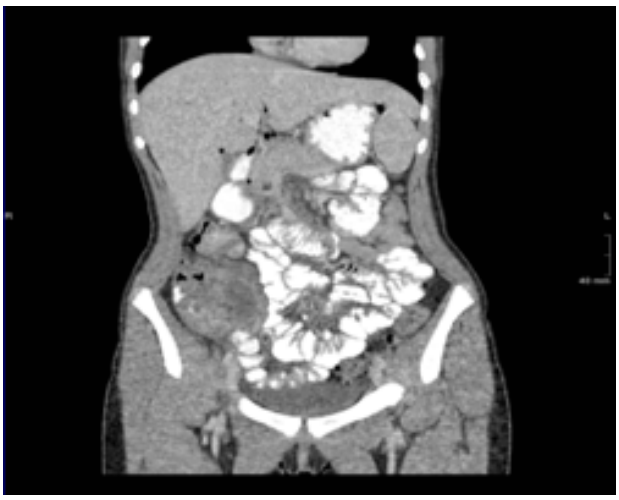
The authors declare that there is no conflict of interest.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms

REFERENCES

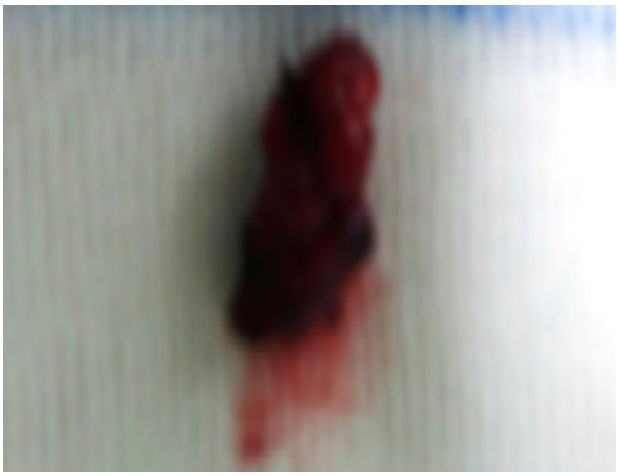
1. *Gupta R, Gernshiemer J, Golden J, Narra N, Haydock T: Abdominal pain secondary to stump appendicitis in a child. J Emerg Med 2000;18:431-433.*
2. *Waseem M, Devas G: A child with appendicitis after appendectomy. J Emerg Med 2008; 34:59-61.*
3. *Erzurum VZ, Kasirajan K, Hashmi M: Stump appendicitis: a case report. J Laparoendosc Adv Surg Tech A 1997; 7:389-91.*
4. *Uludag M, Isgor A, Basak M: Stump appendicitis is a rare delayed complication of appendectomy: A case report. World J Gastroenterol 2006; 12:5401-5403.*
5. *Levine CD, Aizenstein O, Wachsberg RH: Pitfalls in the CT diagnosis of appendicitis. Br J Radiol 2004; 77:792-9.*
6. *Baldisserotto M, Cavazzola S, Cavazzola LT, Lopes MH, Mottin CC: Acute edematous stump appendicitis diagnosed preoperatively on sonography. AJR 2000:503-504.*
7. *Aschkenasy MT, Rybicki FJ. Acute appendicitis of the appendiceal stump. J Emerg Med. 2005; 28:41-3.*
8. *Simon SB, Jacqueline JC, Celia MD. Stump appendicitis af-ter open and laparoscopic appendectomies. Am Surg. 2012; 78:143-4.*
9. *Kanona H, Al Samaraee A, Nice C, Bhattacharya V. Stump appendicitis: a review. Int J Surg. 2012; 10:425-8.*
10. *Gasmi M, Fitouri F, Sahli S, Jemai R, Hamzaoui M. A stump appendicitis in a child: a case report. Ital J Pediatr. 2009; 35:35*
11. *Patel RP, Kan JH. Stump appendicitis. Pediatr Radiol. 2009; 39:306.*
12. *Liang MK, Lo HG, Marks JL. Stump appendicitis: A comprehensive review of literature. Am Surg. 2006;72:162–6.*
13. *Durgun AV, Baca B, Ersoy Y, Kapan M. Stump appendicitis and generalized peritonitis due to incomplete appendicectomy. Tech Coloproctol. 2003;7:102- 4.*
14. *Truty MJ, Stulak JM, Utter PA, Solberg JJ, Degnim AC. Appendicitis after appendectomy. Arch Surg. 2008;143:413–5.*
15. *Greenberg JJ, Esposito TJ. Appendicitis after laparoscopic appendectomy: A warning. J Laparoendosc Surg. 1996;6:185–7.*
16. *Wallbridge PH. Double appendix. Br J Surg. 1962;50:346–7.*
17. *Clark J, Theodorou N. Appendicitis after appendicectomy. J R Soc Med. 2004;97:543–4.*
18. *Mangi AA, Berger DL. Stump appendicitis. Am Surg. 2000;66:739–41.*
19. *Roberts KE, Starker LF, Duffy AJ, Bell RL, Bokhari J. Stump appendicitis: A surgeon's dilemma. JSLS. 2011;15:373–8.*
20. *Shin LK, Halpern D, Weston SR, Meiner EM, Katz DS. Prospective CT diagnosis of stump appendicitis. AJR Am J Roentgenol. 2005;184(Suppl 3):S62–4.*
21. *Watkins BP, Kothari SN, Landercasper J. Stump appendicitis: Case report and review. Surg Laparosc Endosc Percutan Tech. 2004;14:167–71.*
22. *O'Leary DP, Myers E, Coyle J, Wilson I. Case report of recurrent acute appendicitis in a residual tip. Cases J. 2010;3:14.*

Figure 1.



CT scan of abdomen showing abscess in right iliac fossa dimension 41 x 21 mm

Figure 2.



Residual appendix after resection

Acta Chir Croat 2019; 16: 25-27

LAPAROSCOPIC RESECTION OF COMPLICATED MECKEL`S DIVERTICULUM: REPORT OF TWO CASES

Ivo Soldo, Marko Sever, Martin Grbavac, Iva Simović, Saša Palček, Goran Pažur, Anamaria Soldo, Branko Bakula, Lucija Stojčić

ABSTRACT

Background: Meckel`s diverticulum is a true congenital diverticulum of a small intestine. It is remnant of the omphaloenteric duct and positioned at about 50-100 cm proximal to ileocecal valve on an antimesenteric border of ileum. Meckel`s diverticulum can be found in approximately 2% of population and it is asymptomatic in most people. Most common complication is intestinal obstruction (intussusception) (36.5%), inflammation of diverticulum with or without perforation (12.7% and 7.3%) and hemorrhage from ulceration due to an ectopic gastric mucosa (11.8%).

Case study: We report two cases of complicated Meckel`s diverticulum (inflammation and hemorrhages) which were treated laparoscopically.

Conclusion: We find that laparoscopic resection of Meckel`s diverticulum with endostapler is as safe method as open resection with all already known benefits of laparoscopic surgery.

Keywords:
Meckel`s diverticulum, laparoscopy

INTRODUCTION

Meckel`s diverticulum is a true congenital diverticulum of a small intestine. It is remnant of the omphaloenteric duct and positioned at about 50-100 cm proximal to ileocecal valve on an antimesenteric border of ileum. It was named by Johann F. Meckel who had first described it in 1809 [1]. Meckel`s diverticulum can be found in approximately 2% of population and it is asymptomatic in most people [2-4]. About 4% of patients with Meckel`s diverticulum will develop complications. Most common complication is intestinal obstruction (intussusception) (36.5%), inflammation of diverticulum with or without perforation (12.7% and 7.3%) and hemorrhage from ulceration due to an ectopic gastric mucosa (11.8%). In rare cases neoplasm (3.2%) or fistula (1.7%) can occur in Meckel`s diverticulum [5].

Very few patients with asymptomatic Meckel`s diverticulum are aware of having it. In these patients it is most often incidentally found during laparotomy for some other reason. Patients who develop complications like ileus, perforation or inflammation have clear clinical picture of acute abdomen and therefore diagnosis is made through emergency operation. Patients with occult bleeding from Meckel`s diverticulum are hard to diagnose. These patients most often present with chronic anemia of unknown cause. In some cases upper GI barium series can reveal ileal diverticulum [6]. Despite that, nuclear imaging with Technetium-99m is probably the best tool for diagnosis ectopic gastric mucosa that can cause peptic ulcer of a small intestine with consequently bleeding [7]. Technetium behaves in a manner that is analogous to halide anions (eg, chloride, iodide). The mucoid surface cells of gastric mucosa, whether located normally or ectopically, actively accumulate and secrete pertechnetate into the intestine. In rare cases of heavier bleeding with hematochezia selective mesenteric arteriography is indicated. It has high accuracy with bleeding at rate 2-3ml/min and in these cases superselective embolization should be considered, if available, so that surgery can be performed under stable conditions [8].

With increasing surgeons` experience more and more complicated Meckel`s diverticula are being treated laparoscopically.

CASE STUDY

Case of a first patient was 37 years old male, who was admitted to our emergency department due to epigastric pain which migrated several hours later to right lower quadrant of abdomen. Laboratory tests demonstrated leukocytosis (11.8×1000/mm3) and an elevated C- reactive protein (47 mg/L). All other laboratory tests were within normal limits. Native abdominal X- ray was nonspecific. Due to a suspected acute appendicitis no further preoperative imaging diagnostic tests were made and emergency laparoscopic exploration was performed.

Patient was positioned on a table in standard laparoscopy position. Surgeon and assistant were standing on the patient`s left side. Pneumoperitoneum was established using veress needle to the pressure of 15 mm Hg. Trocars were placed in our standard fashion for laparoscopic appendectomy with optical trocar at umbilicus and two

Department of Surgery, Sveti Duh University Hospital, Zagreb, Croatia.
Corresponding author: *Branko Bakula, Department of Surgery, Sveti Duh University Hospital, Zagreb, Croatia,*
e-mail: *bakulabranko@gmail.com*
DOI: *10.5281/zenodo.3517779*

working trocars, 10 mm trocar in left lower quadrant and 5 mm trocar suprapubicly. (Figure 1)

During operation normal appendix was found. Further exploration revealed at about 70 cm from ileocecal valve phlegmonous Meckel`s diverticulum (Figure 2). Diverticulum was excised with linear endostapler (Endo GIA™ 60 mm) at its base and was brought out in an endobag (Figures 3, 4 and 5). Surgery and postoperative course went with no complications. Patient was discharged from the hospital on the 3rd postoperative day in good general condition.

Second patient was 19 years old female, who had been conservatively treated for sideropenic anemia during three years before radionuclide scintigraphy was made and set suspicion on bleeding Meckel`s diverticulum. Preoperative erythrocytes level was 3,45 (normal values 4,0 – 5,0 x 10 6 /mL) and hemoglobin level was 102 g/L (normal values 121 – 151 g/L). Laparoscopic exploration was scheduled.

Position of patient on the table and trocar placement was according to our standard laparoscopic appendectomy procedure, same as with first patient.

During operation long Meckel`s diverticulum was found with diameter of approximately 2 cm (Figure 6). It was resected with linear endostapler (*Endo* GIA™ 60 mm) (Figure 7). Surgery and postoperative course went with no complications. Patient was discharged from the hospital on the 4th postoperative day in good general condition. Pathohistological diagnosis confirmed ectopic gastric mucosa in the Meckel`s diverticulum with peptic ulcer and blood clot at the base of the excised diverticulum. Postoperatively normalization of hemoglobin and iron levels was found.

DISCUSSION:

In only less than 10 % of patients with complicated Meckel`s diverticulum the preoperative diagnosis is correct [9-11]. The condition is often hard to distinguish from other causes of acute abdomen as for example appendicitis, colonic diverticulitis, Crohn`s disease or obstructive ileus caused by other condition than Meckel`s diverticulum [12]. In most cases the definitive diagnosis is made intraoperatively.

On the other hand, in a study of 776 patients by Kusumoto *et al.,* 88% of patients presenting as bleeding had a correct preoperative diagnosis versus 11% with symptoms other than bleeding [13]. Bleeding Meckel`s diverticulum does not present with clinical picture of acute abdomen and in most cases the bleeding is not so severe to obstruct adequate diagnosis process that in most cases will require scintigraphy.

Surgical resection is a standard of treatment of a complicated Meckel`s diverticulum. The resection can be achieved by the simple diverticulectomy or by the resection of a segment of a bowel containing the diverticulum. In cases where intestinal ischemia, perforation or severe inflammation is present segmental bowel resection is recommended. Similar, when ectopic tissue at the diverticular-intestinal junction or palpable mass is found segmental resection should be done. In other cases simple diverticulectomy is considered as a safe procedure.

Whether a asymptomatic, incidentally found Meckel`s diverticulum should be resected or not is still a matter of a debate. Intraoperatively it is usually impossible to determine whether the diverticulum is at increased risk of developing complications. Even though most surgeons do not recommend prophylactic diverticulectomy some authors found that male patients older than 40 and with diverticulum longer than 2 cm are at higher risk of developing complications and therefore in such cases they recommend diverticulectomy of an incidentally found Meckel`s diverticulum [14,15].

CONCLUSIONS

We find that laparoscopic resection of Meckel`s diverticulum with endostapler is as safe method as open resection with all already known benefits of laparoscopic surgery. We also think that laparoscopic surgery has special advantage when dealing with Meckel`s diverticulum because very often one encounters diagnostic dilemma which then can be solved with much less invasive technique of laparoscopy. In our opinion during any laparotomy done for acute abdomen, if Meckel's diverticulum is found, diverticulectomy or intestinal with anastomosis should be performed to avoid secondary complications arising from it.

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.

REFERENCES:

1. *Meckel JF. Uber die divertikel am darmkanal. Arch Die Physio 1809;9:421-53.*
2. *Stone PA, Hofeldt MJ, Campbell JE, et al. Meckel diverticulum: ten-year experience in adults. South Med J 2004;97:1038-41.*
3. *Turgeon DK, Barnett JL. Meckel diverticulum. Am J Gastroenterol 1990;85:777-81.*
4. *Yahchouchy EK, Marano AF, Etienne JCF, et al. Meckel's diverticulum. J Am Coll Surg 2001;192:658-62.*
5. *Yamaguchi M, Takeuchi S, Awazu S. Meckel diverticulum. Investigation of 600 patients in the Japanese literature. Am J Surg 1978;136:247-9*
6. *Pantongrag-Brown L, Levine MS, Buetow PC, et al. Meckel"s enteroliths: clinical, radiologic, and pathologic findings. AJR Am J Roentgenol. 1996 Dec. 167(6):1447-50.*
7. *Swaniker F, Soldes O, Hirschl RB. The utility of technetium 99m pertechnetate scintigraphy in the evaluation of patients with Meckel"s diverticulum. J Pediatr Surg. 1999 May. 34(5):760-4*
8. *Okazaki M, Higashihara H, Yamasaki S, et al. Arterial embolization to control life-threatening hemorrhage from a Meckel"s diverticulum. AJR Am J Roentgenol. 1990 Jun. 154(6):1257-8.*

9. *Bani-Hani K.E., Shatnawi N.J. Meckel's diverticulum: comparison of incidental and symptomatic cases. World J. Surg. 2004;28:917–920.*
10. *Ludtke F.E., Mende V., Kohler H., Lepsien G. Incidence and frequency or complications and management of Meckel's diverticulum. Surg. Gynecol. Obstet. 1989;169:537–542.*
11. *Yahchouchy E.K., Marano A.F., Etienne J.C., Fingerhut A.L. Meckel's diverticulum. J. Am. Coll. Surg. 2001;192:658–662.*
12. *Mendelson K.G., Bailey B.M., Balint T.D., Pofahl W.E. Meckel's diverticulum: review and surgical management. Curr. Surg. 2001;58:455–457*
13. *Kusumoto H., Yoshida M., Takahashi I., Anai H., Maehara Y., Sugimachi K. Complications and diagnosis of Meckel's diverticulum in 776 patients. Am. J. Surg. 1992;164:382–383.*
14. *Mackey WC, Dineen P. A fifty year experience with Meckel's diverticulum. Surg Gynecol Obstet. 1983 Jan; 156(1):56-64.*
15. *Stone PA, Hofeldt MJ, Campbell JE, Vedula G, DeLuca JA, Flaherty SK. Meckel diverticulum: ten-year experience in adults. South Med J. 2004 Nov; 97(11):1038-41.*

FIGURES:

Figure 1:

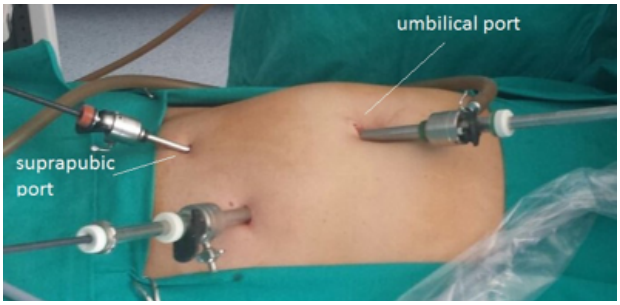


Figure 2:

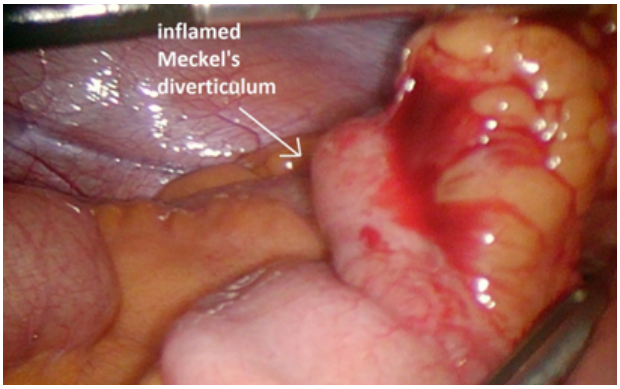


Figure 3:

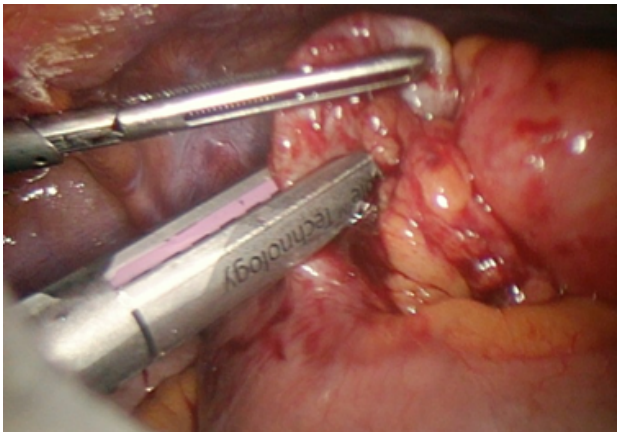


Figure 4:

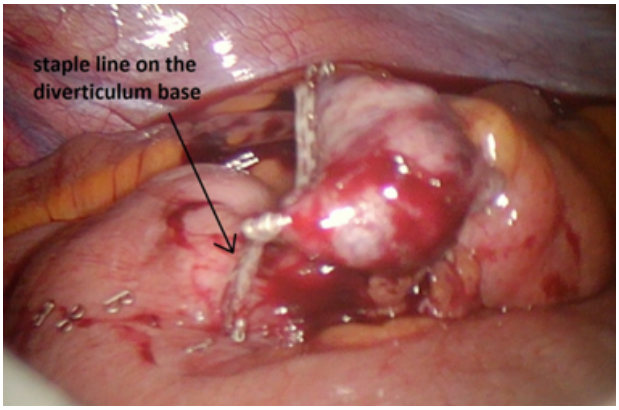


Figure 5:

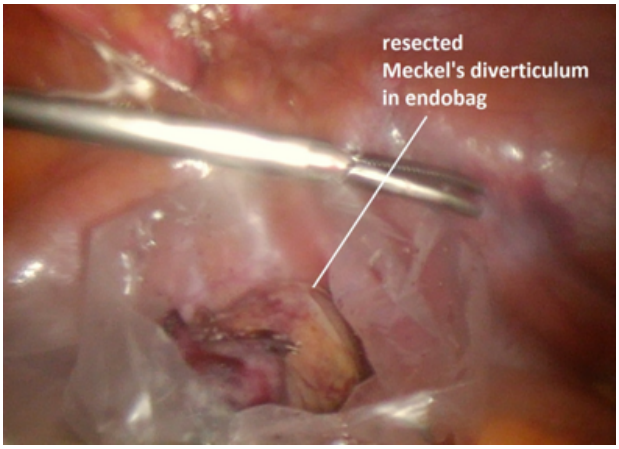


Figure 6:

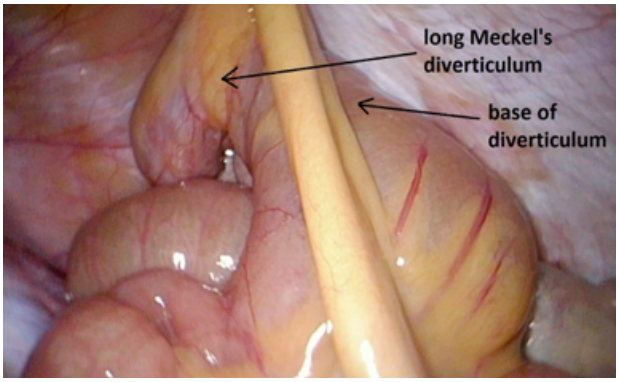
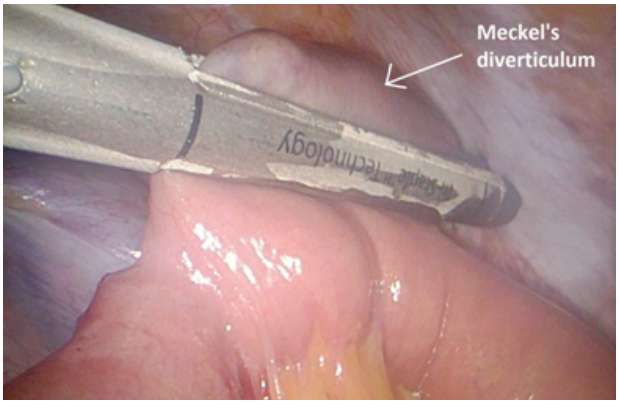


Figure 7:



Acta Chir Croat 2019; 16: 29-33

RELAPSE OF THE PSEUDOMYXOMA PERITONEI AFTER A CYTOREDUCTIVE SURGERY WITH PERITONECTOMY AND HIPEC, 10-YEAR FOLLOW-UP - CASE REPORT WITH A LITERATURE REVIEW

Dino Bobovec[1], Lana Jurlin[2], Mate Majerović[1]

ABSTRACT

Background: Pseudomyxoma peritonei is a rare clinical condition characterized by mucin-secreting epithelial cells which lead to formation of jelly-like structures within the peritoneal cavity and the accumulation of mucinous ascites. Most commonly it arises from the intra-abdominal spread of appendiceal mucinous tumors. Few therapeutic options exist, but a combination of cytoreductive surgery and hyperthermic intraperitoneal chemotherapy, as described by Sugarbaker, stands for the treatment of choice in many tertiary centers nowadays.

Case study: We present a 62-year-old female patient who was initially presented as acute appendicitis. Later pathohistological diagnosis of Pseudomyxoma peritonei just confirmed intraoperative suspicions. Definitive diagnosis was followed with a complete cytoreductive surgery and hyperthermic intraperitoneal chemotherapy. After a 10 year relapse-free follow up, she presented with colon adenocarcinoma and a recurrence of jelly-like incapsulated structures within the abdominal cavity.

Conclusion: In patients diagnosed with pseudomyxoma peritonei, according to current findings, best results are achieved using complete cytoreductive surgery and hyperthermic intraperitoneal chemotherapy. However, recurrences still do occur and there is no real consensus regarding their optimal treatment.

Keywords:
Pseudomyxoma peritonei, cytoreductive surgical procedures, hyperthermic intraperitoneal chemotherapy

Introduction

Pseudomyxoma peritonei is a rare clinical condition with unknown cause and estimated overall incidence of 1-2 per million [1]. The primary tumor usually originates from the appendix, but can also appear in ovarium, urinary bladder or any other part of the gastrointestinal system [2]. Female patients are more likely affected. Synchronous tumors of appendix and ovaries were described, but recent immunohistochemical studies suggest that „synchronous" ovarian tumor is actually a metastatic process of primary appendiceal tumor [3]. Pseudomyxoma peritonei begins as an appendiceal adenoma which starts to produce mucus intraluminally and consequently leads to obstruction and rupture of the appendix. Therefore, tumor cells are spread intraperitoneally, where they continue with their mucus production. Pseudomyxomal tumorous cells tend to characteristically accumulate around the liver, omentum and in the lower pelvic region, unlike some other gastrointestinal tumor metastases which usually incorporate close to their perforation site. As the tumor progresses, metastatic cells spread to entire abdomen, but there is always absence of tumor masses on the free portion of the small intestine, which is described as a pathognomonic sign [4, 5]. An intraabdominal accumulation of jelly-like masses causes unspecific symptoms like abdominal or pelvic pain, distension of abdomen, and infertility. After a long period of accumulation, eventually, mechanical obstruction of the gastrointestinal tract occurs. Because of that, early diagnosis is difficult to make and it is often misdiagnosed. Differential diagnosis in female patients is even harder as it includes ovarian tumors, which can also produce mucus and cause distension of the abdomen with similar symptoms. Therapy of pseudomyxoma peritonei depends on the tumor size and a stage of the disease. In the majority of patients, long term prognosis is poor and depends upon the histological type of tumor with 50% of 5-year survival [6]. When possible, cytoreductive surgery combined with intraperitoneal chemotherapy shows best long-term results [7]. Other therapy options include „debulking" surgery, classic intravenous chemotherapy or expectative follow-up. We report a case of a female patient diagnosed with pseudomyxoma peritonei who was treated by complete cytoreductive surgery and peritonectomy accompanied by hyperthermic intraperitoneal chemotherapy (HIPEC). The patient had uneventful recovery in a 10-year relapse-free follow up but then presented with colon adenocarcinoma and a recurrence of jelly-like incapsulated structures within the abdominal cavity.

[1]*Department of Surgery, University Hospital Centre Zagreb, Croatia*
[2]*Department of Otorhinolaryngology, General Hospital Varaždin, Croatia*
Corresponding author: *Dino Bobovec, MD, Department of Surgery, University Hospital Centre Zagreb, Kišpatićeva 12, 10000 Zagreb, Croatia,*
e-mail: *dbobovec@gmail.com*
DOI: *10.5281/zenodo.3517773*

Case study

A 62-year old female patient presented to the emergency department with a seven-day history of constant periumbilical pain, localized to the right lower quadrant. 8 months prior to arrival, she experienced the same symptoms which regressed spontaneously. Her medical history revealed hypertension and uterine myoma. By the time of examination, she did not feel nausea, anorexia, neither had diarrhea. Physical examination revealed an obese female patient with rebound tenderness over the right lower abdominal quadrant. Vital signs were within normal limits (RR 140/90mmHg, BPM 78/min). Laboratory findings showed an elevated white blood count (11,21x109/L) and CRP (80mg/L). The ultrasound study showed a collection of dense fluid at the ileocecal region, which was described as an abscess formation. The plain abdominal radiograph showed no signs of pneumoperitoneum. Pararectal laparotomy revealed the intraabdominal cavity fulfilled with gelatinous masses and numerous cystic lesions, which covered peritoneum and liver. Appendix was thickened >3 cm in the distal part but showed no signs of inflammation or rupture. Appendectomy was done with pathohistological diagnosis of carcinoid tumor of the appendix and peritoneal carcinomatosis. For the next 34 months she was treated conservatively with an obvious deterioration of her clinical condition and progression of the radiological findings (Figure 1.). Her main complaint was distension of the abdomen accompanied with anorexia and constipation (Figure 2.).

Laparotomy was done, intraabdominal jelly-like masses (Figure 3.) were evacuated and complete peritonectomy together with cytoreductive surgery followed by hyperthermic intraperitoneal chemotherapy (mitomycin/cisplatin) was undertaken. The pathohistological diagnosis was pseudomyxoma peritonei.

Postoperative period passed uneventfully and the patient was in regular surgical and oncologic follow up for the next 10 years. During that period she was asymptomatic and felt occasional nausea and bloating. Because of sideropenic anemia, a diagnostic colonoscopy was undertaken, which revealed a stricture of the ileocolic anastomosis. Laparotomy showed infiltrating process at the location of the previously formed anastomosis. Tumorous process was resected and the new anastomosis was formed. During the procedure, a pseudocystic formation was noticed in the retrogastric area which was left untouched and the patient was radiologically reevaluated. CT of abdomen and pelvis revealed an unusual, well defined, lobulated cyst which was located in omental bursa and was in close contact with the pancreas (Figure 4.).

Additional PET/CT was done which showed a multicystic expansive formation under the left lobe of the liver which incorporates antral gastric region and shows no metabolic activity, probably due to the majority of jelly-like content. Relaparotomy was made and showed an egg-like white encapsulated process in the bursa omentalis which was in close connection with the surrounding structures, but does not infiltrate them (Figure 5. A). The tumor was easily removed and opened afterward. It was formed of mucinous and jelly-like content (Figure 5. B). The pathohistological diagnosis was mucin and lipoma.

Discussion

In the past, patients diagnosed with pseudomyxoma peritonei were treated by surgical reduction of the tumor and repeated drainages of mucinous ascites, with poor results and common relapses, which resulted in low quality of life. [8]. In 1980. Spratt et al. introduced hyperthermic intraperitoneal chemotherapy as a new therapy option [9]. A few years later, Sugarbaker proposed treatment of pseudomyxoma peritonei with the combined procedure, which included cytoreductive surgery accompanied by hyperthermic intraperitoneal chemotherapy [10]. Despite a lack of multicentric prospective randomized studies, literature published during the last two decades confirms the efficiency of the combined procedure, which became the treatment of choice in many tertiary centers worldwide nowadays [11-16]. It is usually used for appendiceal low-grade tumors with peritoneal dissemination [17], with overall 10-year survival of 63% [18]. Miner et al. questioned the efficiency of complete cytoreductive surgery combined with HIPEC because of its radical approach and variety of perioperative complications. The study included a group of patients treated by series of less aggressive cytoreductions (average 2.2 surgeries per patient) and with selective usage of intraperitoneal chemotherapy with a median survival of 12.8 years for patients with low-grade tumor and only 4-year survival for patients with high-grade tumors. The overall 10-year survival was 23% [8]. On the other hand, Sugarbaker et al. analyzed a similar group of patients, who were treated by primary aggressive cytoreduction (1.3 surgery per patient) followed by intraperitoneal chemotherapy and showed 20-year survival of 70% [19]. The success of combined therapy lies in primary aggressive cytoreduction which tends to totally eradicate tumorous mass in just one radical surgical procedure which is followed by intraperitoneal chemotherapy directed towards the residual microscopic tumor. This kind of approach allows early and total removal of the tumor, which is not possible in a series of less aggressive palliative procedures, because of intraabdominal adhesions formation with every new laparotomy. Also, there is no evidence that perioperative morbidity and mortality are related to intraperitoneal chemotherapy. Possible complications are in fact related to duration and extensiveness of radical surgical procedure. Despite combined therapy, tumor relapses still occur and present a diagnostic and

therapeutic problem. Delhorme et al. analyzed a tumor recurrence in a group of patients treated by primary cytoreductive surgery and HIPEC. During an 85 months follow-up 26% of patients had a tumor relapse, with a median of 25 relapse free months. In the majority of cases (76%), primary relapse was localized intraperitoneally, as it is described in the reported case [20]. Even though the treatment of intraperitoneal recurrences is still debatable, redo surgery for isolated intraperitoneal recurrence is proposed by most of the authors [21-25]. There is an absence of consensus regarding redo HIPEC, considering its failure to prevent relapse of the disease. With no doubt, large randomized studies are necessary to define which groups of patients would have a benefit and should be treated by cytoreductive surgery and HIPEC. Described patient fits in a group of successfully treated patients with the combined method, despite intraabdominal status worsening and disease relapse after a 10-year follow-up.

Conclusion

There is a lack of evidence based guidelines which would define the best treatment options for patients diagnosed with pseudomyxoma peritonei. According to current findings, complete cytoreductive surgery and hyperthermic intraperitoneal chemotherapy is a treatment of choice nowadays. A fact that pseudomyxoma peritonei is a rare clinical condition shouldn't lessen a need for further investigations which will lead to clear guidelines based on strong evidence.

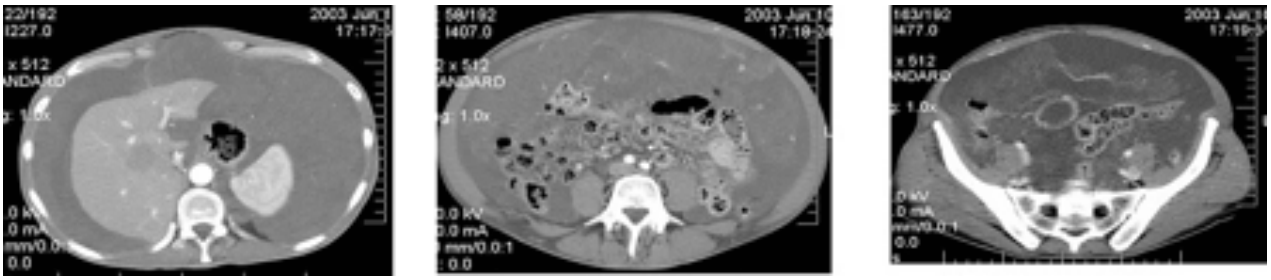
The authors declare that there is no conflict of interest.

REFERENCES

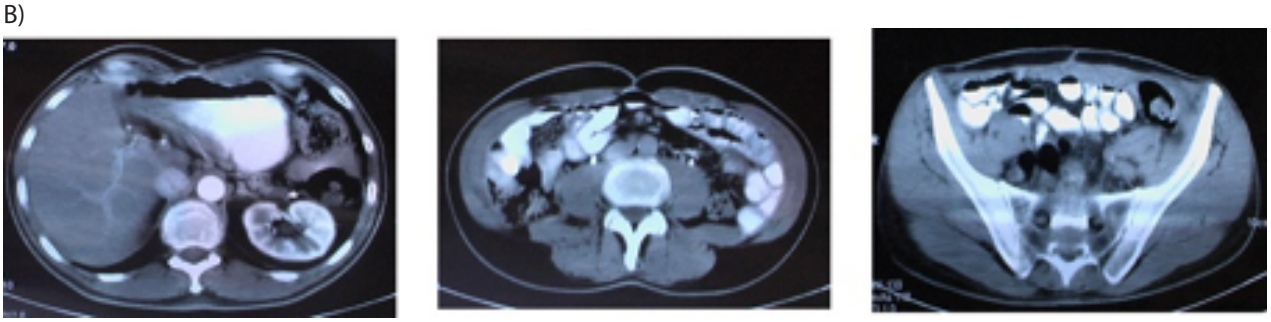
1. *Smeenk RM, van Velthuysen ML, Verwaal VJ, Zoetmulder FA. Appendiceal neoplasms and pseudomyxoma peritonei: a population based study. Eur J Surg Oncol 2008;34:196–201.*
2. *Mukherjee A, Parvaiz A, Cecil TD, Moran BJ. Pseudomyxoma peritonei usually originates from the appendix: a review of the evidence. Eur J Gynaecol Oncol 2004;25:411–414.*
3. *Ferreira CR, Carvalho JP, Soares FA, Siqueira SA, Carvalho FM. Mucinous ovarian tumors associated with pseudomyxoma peritonei of adenomucinosis type: immunohistochemical evidence that they are secondary tumors. Int J Gynecol Cancer 2008;18:59–65.*
4. *Moran BJ, Cecil TD. The etiology, clinical presentation, and management of pseudomyxoma peritonei. Surg Oncol Clin N Am 2003;12:585–603.*
5. *Sugarbaker PH. Pseudomyxoma peritonei. A cancer whose biology is characterized by a redistribution phenomenon. Ann Surg 1994;219:109–111.*
6. *Hinson FL, Ambrose NS. Pseudomyxoma peritonei. Br J Surg 1998;85:1332–1339.*
7. *Stewart JH 4th, Shen P, Levine EA. Intraperitoneal hyperthermic chemotherapy for peritoneal surface malignancy: current status and future directions. Ann Surg Oncol 2005;12:765–777.*
8. *Miner T.J., Shia J., Jaques D.P., Klimstra D.S., Brennan M.F., Coit D.G. Long-term survival following treatment of pseudomyxoma peritonei: an analysis of surgical therapy. Ann Surg 2005;241:300–308.*
9. *Spratt J.S., Adcock R.A., Muskovin M., Sherrill W., McKeown J. Clinical delivery system for intraperitoneal hyperthermic chemotherapy. Cancer Res 1980;40:256–260.*
10. *Sugarbaker PH, Kern K, Lack E. Malignant Pseudomyxoma peritonei of colonic origin. Natural history and presentation of a curative approach to treatment. Dis Colon Rectum 1987;30:1772–1779.*
11. *Moran B, Baratti D, Yan T, Kusamura S, Deraco M. Consensus statement on the loco-regional treatment of appendiceal mucinous neoplasms with peritoneal dissemination (pseudomyxoma peritonei). J Surg Oncol 2008;98:277–282.*
12. *Elias D, Honoré C, Ciuchendéa, R. et al. Peritoneal pseudomyxoma: results of a systematic policy of complete cytoreductive surgery and hyperthermic intraperitoneal chemotherapy. Br J Surg 2008;95:1164–1171.*
13. *Sugarbaker PH. Cytoreductive surgery and peri-operative intraperitoneal chemotherapy as a curative approach to pseudomyxoma peritonei syndrome. Eur J Surg Oncol J Eur Soc Surg Oncol Br Assoc Surg Oncol 2001;27:239–243.*
14. *Güner Z, Schmidt U, Dahlke MH, Schlitt HJ, Klempnauer J, Piso P. Cytoreductive surgery and intraperitoneal chemotherapy for pseudomyxoma peritonei. Int J Colorectal Dis 2005;20:155–160.*
15. *Loungnarath R, Causeret S, Bossard N et al. Cytoreductive surgery with intraperitoneal chemohyperthermia for the treatment of pseudomyxoma peritonei: a prospective study. Dis Colon Rectum 2005;48:1372–1379.*
16. *Youssef H, Newman C, Chandrakumaran K, Mohamed F, Cecil TD, Moran BJ. Operative findings, early complications, and long-term survival in 456 patients with pseudomyxoma peritonei syndrome of appendiceal origin. Dis Colon Rectum 2011;54:293–299.*
17. *Ahmed S, Stewart JH, Shen P, et al. Outcomes with cytoreductive surgery and HIPEC for peritoneal metastasis. J Surg Oncol 2014;110:575-584.*
18. *Chua TC, Moran BJ, Sugarbaker PH, et al. Early- and long-term outcome data of patients with pseudomyxoma peritonei from appendiceal origin treated by a strategy of cytoreductive surgery and hyperthermic intraperitoneal chemotherapy. J Clin Oncol 2012;30:2449-2456.*
19. *Sugarbaker PH, Ronnett BM, Archer A, et al. Pseudomyxoma peritonei syndrome. Adv Surg 1996;30:233–280.*
20. *Delhorme JB, Honoré C, Benhaim L, Dumont F, Dartigues P, Dromain C, et al. Long-term survival after aggressive treatment of relapsed serosal or distant pseudomyxoma peritonei. Eur J Surg Oncol 2017;43:159-167.*
21. *Golse N, Bakrin N, Passot G, et al. Iterative procedures combining cytoreductive surgery with hyperthermic intraperitoneal chemotherapy for peritoneal recurrence: postoperative and long-term results. J Surg Oncol 2012;106:197–203.*
22. *Smeenk RM, Verwaal VJ, Antonini N, Zoetmulder FAN. Progression of pseudomyxoma peritonei after combined modality treatment: management and outcome. Ann Surg Oncol 2007;14:493–499.*
23. *Klaver YLB, Chua TC, Verwaal VJ, de Hingh IHJT, Morris DL. Secondary cytoreductive surgery and peri-operative intraperitoneal chemotherapy for peritoneal recurrence of colorectal and appendiceal peritoneal carcinomatosis following prior primary cytoreduction. J Surg Oncol 2013;107:585–590.*
24. *Yan TD, Bijelic L, Sugarbaker PH. Critical analysis of treatment failure after complete cytoreductive surgery and perioperative intraperitoneal chemotherapy for peritoneal dissemination from appendiceal mucinous neoplasms. Ann Surg Oncol 2007;14:2289–2299.*
25. *Bijelic L, Yan TD, Sugarbaker PH. Treatment failure following complete cytoreductive surgery and perioperative intraperitoneal chemotherapy for peritoneal dissemination from colorectal or appendiceal mucinous neoplasms. J Surg Oncol 2008;98:295–299.*

Figure 1.

A)

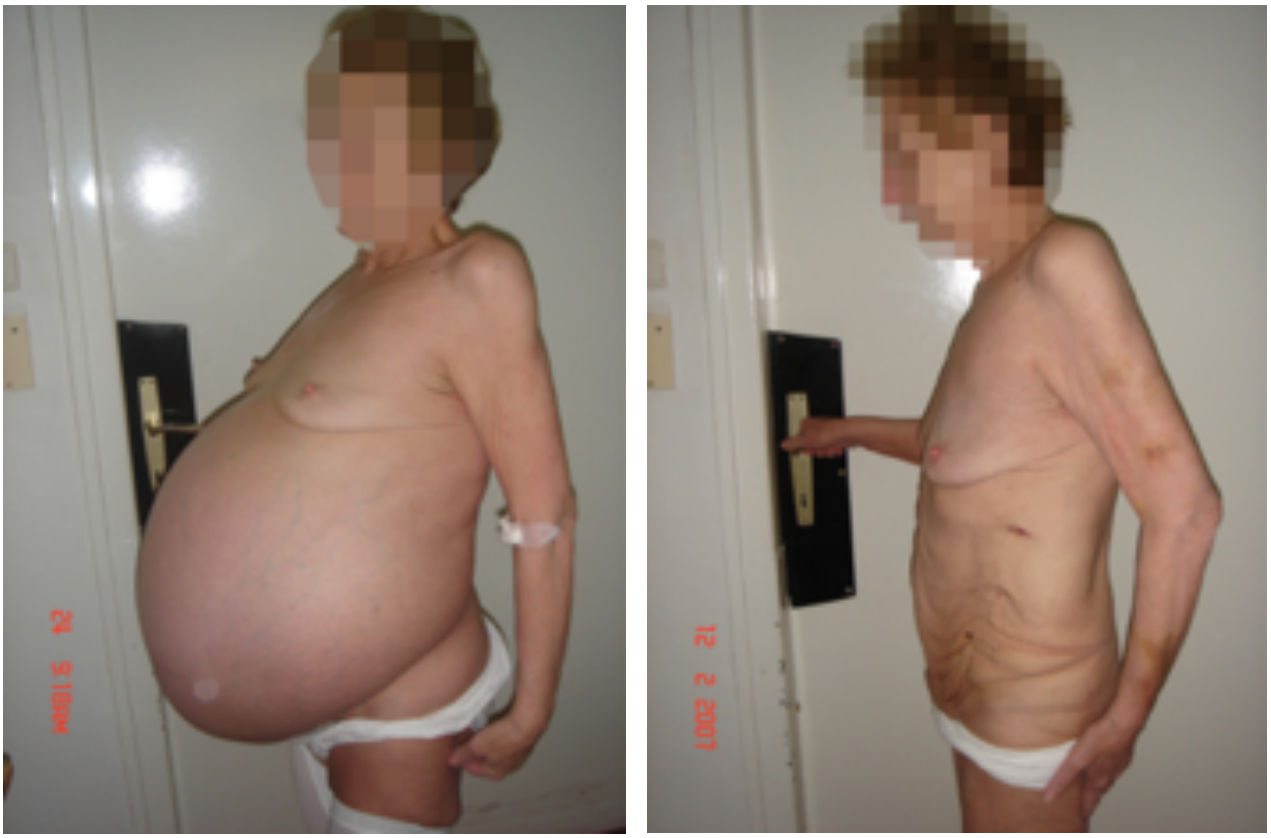


B)



A) Preoperative MSCT of abdomen and pelvis showing diffuse collections of gelatinous material and mucus around liver, spleen, and intestines. B) Postoperative MCST of abdomen and pelvis after a complete cytoreductive surgery with HIPEC.

Figure 2.

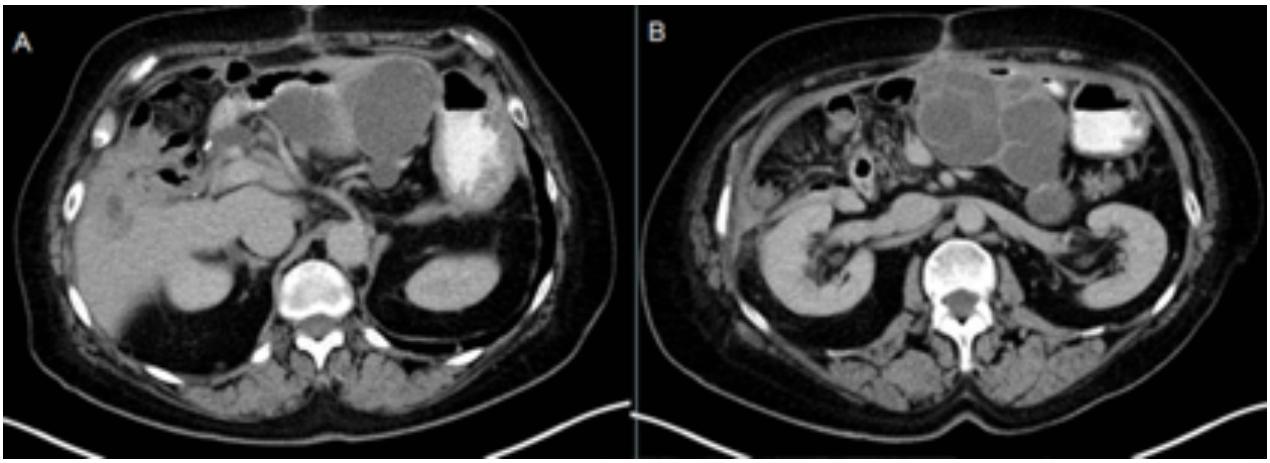


On the left preoperative picture of the patient with massive distension of the abdomen. On the right postoperative picture of the patient with uneventful recovery.

Figure 3.

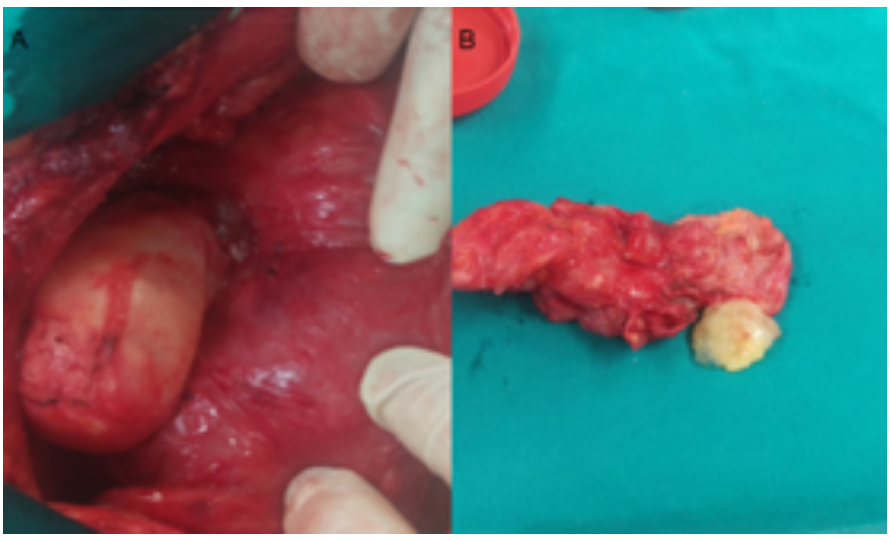
Intraoperative picture showing evacuated gelatinous material and mucus.

Figure 4.



Abdominal CT scan of well defined, lobulated cyst in omental bursa which is in close contact with the pancreas.

Figure 5.



A) Intraoperative photo of the cyst located in retrogastric area protruding through the hepatic recess.
B) Photo of the surgical specimen after extirpation and opening which revealed mucinous material.

Acta Chir Croat 2019; 16: 35-37

PATIENT WITH MALIGNANT VULVAR NEOPLASM: CASE REPORT

Mariam Samara[1], Ema Somen[1], Davor Mijatović[2]

ABSTRACT

Background: Vulvar carcinoma most often occurs on the outer surface area of the female genitalia. The vulva is the area of skin that surrounds the urethra and vagina, including the clitoris and labia. It is quite rare cancer of the female reproductive system and accounts for 3-4% of all genitourinary tract neoplasms. Though it can occur at any age, vulvar cancer is most common in older adults. When present in young women it is mostly associated with human papillomavirus (HPV)–related dysplasia. The most common histology is squamous cell carcinoma of the vulva.

Case study: This report presents a patient with verified planocellular vulvar carcinoma who underwent radical hysterectomy with adnexectomy and lymphadenectomy due to cervical neoplasm (squamous cell carcinoma). After chemo-radio therapy, the patient underwent radical vulvectomy and two months later, resection of the distal urethra and vulvar and vaginal reconstruction with gracilis muscle. Full-thickness skin graft was used to reconstruct the distal part of the urethra.

Conclusion: Combination of flaps and full-thickness skin graft can be used in reconstruction of vulva and urethra.

Keywords:
Malignant vulvar neoplasm, planocellular carcinoma, reconstructive techniques, vulvectomy

INTRODUCTION

Vulvar cancer is rare cancer of the female reproductive system and accounts for only 3-4% of all genitourinary tract neoplasms. The peak of its incidence is in the 7th and 8th decade of life. In most cases (90%) it is planocellular carcinoma, second most common is basocellular carcinoma (4%), while the rest (6%), are cases of rare carcinoma types, such as melanoma, Paget's disease, Bartholin gland carcinoma [1-2].

There are at least two types of planocellular vulvar carcinoma. The first type is usually a result of VIN (vulvar intraepithelial neoplasia) progression, most commonly occurs in younger women and is associated to HPV infection, promiscuity, and condyloma. Vulvar intraepithelial neoplasia is premalignant dysplasia of vulvar squamous epithelium which differs from carcinoma because the epithelial basement membrane is intact. Depending on a height of epithelium it is divided into three stages. In the first stage, VIN 1, only one-third of the thickness of the surface layer of the vulva is affected; in the second stage, VIN 2, two-thirds are affected, while in the third stage, VIN 3 (carcinoma in situ) the full thickness of the surface layer of the vulva is affected [3-5].

The second type most commonly occurs in obese, diabetic, and women with hypertension. Besides already mentioned, risk factors are also considered to be smoking, lichen sclerosus, squamous hyperplasia, chronic granulomatous diseases, and squamous vaginal or cervical carcinoma. The labia majora and clitoris are the most common sites. Vulvar cancer usually presents with palpable, slow-growing mass, and patients visit their doctors because of itching, pain, bleeding, vaginal discharge, or burning sensation during urination [1-3].

Vulvar cancer can spread locally (ex.urethra, urinary bladder, vagina, anus or rectum), hematogenous, in inguinal lymph nodes or from inguinal lymph nodes to pelvic and paraaortic lymph nodes [5,6].

Treatment consists of radical wide excision and removal of inguinal lymph nodes (in patients with early stage disease) while chemoradiation is an alternative to radical vulvectomy with en bloc inguino-femoral lymphadenectomy for advanced disease [5,6].

CASE STUDY

A 64-year-old patient was admitted to the Department of Plastic, Reconstructive, and Aesthetic Surgery at the Clinical Hospital Center Zagreb for the excision of the malignant vulvar neoplasm.

In April 2011, the patient underwent radical hysterectomy with adnexectomy and lymphadenectomy in Department of Gynaecology and Obstetrics at Clinical Hospital Center Zagreb due to cervical neoplasm (squamous cell carcinoma). Radiotherapy (administered for 29 consecutive days), brachytherapy (50 hours), and concomitant chemotherapy (cisplatin 40 mg/m2, once a week, 4 consecutive weeks) followed.

In May 2012, the patient underwent radical vulvectomy after pathohistological analysis confirmed planocellular carcinoma with negative margins. Two months later, resection of the distal urethra and vulvar and vaginal

[1]*School of Medicine, University of Zagreb, Croatia*
[2]*Department of Surgery, University Hospital Centre Zagreb, Croatia*
Corresponding author: *Mariam Samara, School of Medicine, University of Zagreb, Croatia,*
e-mail: *mariam.samara@hotmail.com*
DOI: *10.5281/zenodo.3517775*

reconstruction with gracilis muscle were performed. Full-thickness skin graft (Wolf-Krausse) was taken from the left thigh and was used to reconstruct the distal part of the urethra. This is somewhat more risky procedure than using split-thickness graft because of the much higher percentage of graft failure and consequent graft contraction. The area from which the graft was taken heals faster and less painful than in case of split-thickness graft. Very appreciated characteristic of full-thickness graft is also a lower incidence of secondary contraction, which enables great reconstructive results. In this case, the full-thickness graft was taken from the left thigh, and distal urethral part was reconstructed. During the surgical procedure, the patient was under general anesthesia, a fusiform skin graft was taken from thigh using inguinal crease as long axis. Perineal skin defect was covered using Thiersch split-thickness skin graft. In May 2018 the patient had discovered lesion and experienced difficulty with urination. She was referred to the department of gynecology and obstetrics, University Hospital Centre Zagreb. Gynecological examination revealed cystic formation, located left paraurethral, movable size 4 cm. The biopsy was performed. Cytological as well as microbiological analysis was done. Preoperative abdominal ultrasound revealed hypoechoic zone with posterior accumulation caudal from urinary bladder towards rectum, without Doppler signal (used for vascularity assessment), possible cystic mass/fluid retention in postoperative scar tissue. Removal of the superficial structures revealed tumorous cystic formation (figure 1).

According to the results, differential diagnoses included malignant neoplasms of the vulva. Surgical removal, excision (without lymphadenectomy), was performed (figure 2). Pathological and cytological analysis, were made. A smear test was positive for malignant cells.

Postoperative pathology reported lots of abnormal squamous cells, less specialized with degenerative changes and necrotic cellular debris. According to the clinical cytologist the lesion was planocellular carcinoma. Pathohistological examination of tumor-associated material showed abnormal tissue size 7x1.5x1.3 cm, constructed of a poor cellular connective tissue with cystic formation, partially thickened with atypical moderately differentiated epithelium. Described areas were also found in the stromal connective tissue. Skeletal muscle tissue was described marginally as well.

Postoperative course was uneventful. The patient was discharged from the hospital in good condition, on the 7th postoperative day.

DISCUSSION

Female urethral reconstruction is complex, and one must carefully evaluate patients. There are some differences from male urethral reconstruction because of the different urethral length. The female urethra is shorter than the male urethra, leading to a higher risk of incontinence. Reconstructive techniques can be categorized as anastomotic, flap-based or graft-based. The surgical approach for these techniques of repair can be described by its relative position to the urethra; dorsal (that is 12 o'clock), ventral (that is 6 o'clock) and circumferential. Ventral (supravaginal) and dorsal accesses have been the most frequent ones, although the small number of patients have undergone ″tubularized urethroplasty″[5-7].

The advantage of using a ventral approach is the minimal mobilization of the urethra, however, this access lead to the higher risk of developing a urethrovaginal fistula.

Potential benefits of the dorsal approach would include the avoidance of a vaginal incision and its associated post-operative complications, including issues with urethrovaginal fistula and wound complications. The dorsal access provides some more positive facts such as good mechanical support and well-vascularized base for receiving the transplant. However, surgeons confront some disadvantages using a dorsal approach as well. The injury of the sphincter mechanism causing incontinence is possible, as well as sexual dysfunction caused by a neurosensory disorder [5-7].

Female urethral reconstruction can be performed with lingual flap and buccal mucosal graft urethroplasty, but the highest success rates have been reported to treat a heterogeneous group of urethral disorders, such as urethral stricture and urethral loss. Urethral replacement can be accomplished by in situ tubularized full thickness buccal mucosal graft based on the clitoris creating the neourethral tube. Neourethra is then covered with the gluteus tissue, or the periurethral tissue can be used as well. The disadvantage of the buccal mucosal graft is a low, but significant risk of the salivary glands injuries [6-9].

The disadvantage of the full-thickness graft is a higher risk of the transplant rejection. Also, the healing process is slower due to full-thickness skin graft.

Several more reconstructive techniques have been described. These have included vaginal or labial flaps. Flaps are the most common and the earliest published reconstructive technique utilized for female urethral stricture. Flaps are particularly advantageous because of their mobility and good vascularity, meaning they can be raised with relative ease. In this case, these techniques were not possible due to radical vulvectomy [7-9].

CONCLUSIONS

The prognosis for vulvar carcinoma is fairly poor due in part to their rare nature and a relative lack of evidence-based management strategies, but the most important is early diagnosis. Early diagnosis can improve prognosis in vulvar carcinoma or other aggressive tumors and, in the case of recurrences, may lead to changes in therapy. Early diagnosis followed by prompt surgical operation remains the best treatment for patients at present.

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.

REFERENCES

1. *Buchanan T, Mutch D. Squamous cell carcinoma of the vulva: a review of present management and future considerations. Expert Rev Anticancer Ther. 2019;19(1):43-5.*
2. *Te Grootenhuis NC, Pouwer AW, de Bock GH et al. Prognostic factors for local recurrence of squamous cell carcinoma of the vulva: A systematic review. Gynecol Oncol. 2018;148(3):622-631.*
3. *Faber MT, Sand FL, Albieri V, Norrild B, Kjaer SK, Verdoodt F. Prevalence and type distribution of human papillomavirus in squamous cell carcinoma and intraepithelial neoplasia of the vulva. nt J Cancer. 2017;141(6):1161-1169.*
4. *Del Pino M, Rodriguez-Carunchio L, Ordi J. Pathways of vulvar intraepithelial neoplasia and squamous cell carcinoma. Histopathology. 2013;62(1):161-75.*
5. *Dellinger TH, Hakim AA, Lee SJ, Wakabayashi MT, Morgan RJ, Han ES. Surgical Management of Vulvar Cancer. J Natl Compr Cancer Netw JNCCN. 2017;15(1):121–8.*
6. *Mijatović D, Smud S, Dujmović A. Our experience in vulvar reconstruction after radical and hemivulvectomy. Lijec Vjesn. 2012;134(5–6):192–4.*
7. *Hoag N, Chee J. Surgical management of female urethral strictures. Transl Androl Urol. 2017;6(Suppl 2):S76–80.*
8. *Faiena I, Koprowski C, Tunuguntla H. Female Urethral Reconstruction. J Urol. 2016;195(3):557-67.*
9. *Osman NI, Chapple CR. Contemporary surgical management of female urethral stricture disease. Curr Opin Urol. 2015;25(4):341–5*

FIGURE CAPTIONS

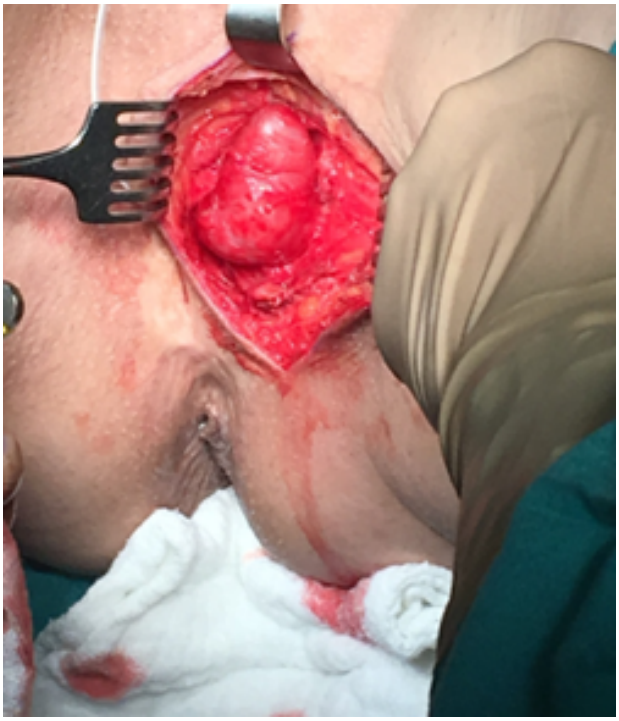


Figure 1: Cystic tumorous mass found during procedure

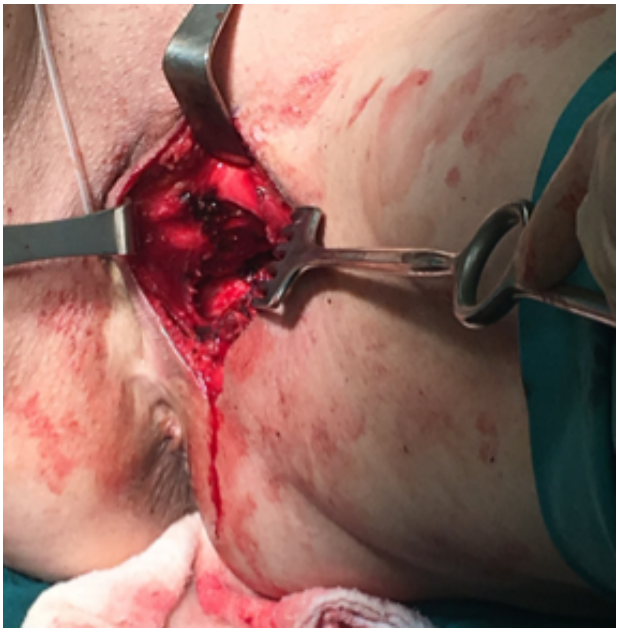


Figure 2: postoperative status (after cystic mass removal)

AUTHOR'S INDEX