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Complicated Hydatid Cysts of the Lung: Is Capitonnage Better than Uncapitonnage Method? Comparison Between Capitonnage and Uncapitonnage Technique in the Complicated Hydatid Cysts of the Lung

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Abstract: After evacuation of the cyst, the residual cavity, always has some bronchial openings, must be managed appropriately. About residual cavity in complicated and noncomplicated hydatid cysts and capitonnage there is controversy. The aim of this study is to comparison advantages of capitonnage and non-capitonnage in the complicated hydatid cystof the lung because there is not enough research in this issue. This study is a retrospective descriptive cross-sectional study that was done on patient with impending to perforation and complicated pulmonary hydatid cyst. Demographic information consist of: age, sex, home place, career contact with livestock and clinical data consist of: Hospital stay, complications and mortality was noted. All data analyzed with SPSS. In our study was surveyed 75 patient in tow group with Capitonnage (CTG) (36) and Uncapitonnage (UCTG) (39). Mean age and standard deviation in (CTG) was (30±17.1) years and patient (UCTG) was (37.3±18.5) years. There were not significantly different between number of gender in 2 groups ($p = 0.08$). Hospital stay in our study in (UCTG) was 6.49±1.05 and in (CTG) 3+0.75±0.65 ($p<0.0001$). Post-operative broncho pleural fistula in (CTG) was 8.3% and in (UCTG) was 23.1 ($p = 0.48$) and post-operative air leak in (UCTG) was 25.6% and in (CTG) was 8.3% ($p = 0.48$). Mean average day for removal chest-tube in (UCTG) was 4.6 ± 0.99 day and in (CTG) was 2.64±0.59 day ($p<0.0001$) and also cumulating the fluid in cavity in (UCTG) was 3.79±0.61 and in (CTG) was 1.61± 0.49 month ($p<0.0001$). Post-operative air leak in (UCTG) was 25.6% and in (CTG) was 8.3% ($p = 0.48$). Mortality was not seen in two groups. Our study shows that capitonnage have less post-operative complications than uncapitonnage and is preferred method in surgery of complicated pulmonary hydatid cyst.

Key words: Pulmonaryhydatid cyst, capitonnage, complicated, non-complicated hydatid cyst, removel chest-tube

INTRODUCTION

Hydatidosis remains endemic to many parts of the world, most notably the Mediterranean region, Australia, New Zealand, the Middle East and South America. In the endemic areas, it is still a major public health problem (Kuzucu *et al*., 2004; Aghajanzade *et al*., 2008; Bagheri *et al*., 2011). Most of the patients are young and the majority are <40 years of age (Kuzucu *et al*., 2004; Aghajanzade *et al*., 2008; Bagheri *et al*., 2011). All hydatid cysts carry the risk of rupture (Bagheri *et al*., 2011). Pulmonary hydatid cysts usually remain asymptomatic until the time of rupture or compress the lung and complicated (Kuzucu *et al*., 2004; Aghajanzade *et al*., 2008; Bagheri *et al*., 2011; Nickolaos *et al*., 2009) and the clinical presentation in these patients is directly related to intact or ruptured cyst status. Rupture may occur during anthelmintic therapy or percutaneous aspiration and can lead to severe complications such as massive hemoptysis and tension pneumothorax (Sonmez *et al*., 2001). Surgery remains the treatment of choice for hydatid cysts of the lung. However, pulmonary hydatidosis primarily affects children of young adults who may be infected again and usually they have great lung capacity for expansion (Kuzucu *et al*., 2004; Sonmez *et al*., 2001). Thus, it is important to always use the most conservative surgical methods possible. More radical surgical procedures may be needed in complicated cases (Kuzucu *et al*., 2004; Aghajanzadeh *et al*., 2008; Bagheri *et al*., 2011; Sonmez *et al*., 2001). In cases with a large pericystic cavity, the preferred method is closure of the bronchial openings, partial pericystectomy, elimination of the residual cavity by capitonnage and closure of its edges with continuous sutures (Sonmez *et al*., 2001). After

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evacuation of the cyst, the residual cavity, always has some bronchial openings, must be managed appropriately (Bagheri *et al*., 2011; Sonmez *et al*., 2001). Some researchers had advised only suturing of the bronchial opening (Bagheri *et al*., 2011; Yalcinkaya *et al*., 1999). This method, can not sufficiently prevent air leak. Others advocated a method named capitonnage in this practice, folding of the pericystic zone by sutures (Sonmez *et al*., 2001; Erdogan *et al*., 2005; Sokouti *et al*., 2011; Turna *et al*., 2002; Eisa *et al*., 2013). Each of these methods in the hands of their proponents, yielded good results. About capitonnage in the complicated pulmonary hydatid cyst there is not accurate study in literatures. Therefore, we present our experiencein this retrospectively study in patients with pulmonary hydatidosis and to compared the, hospital stay residual cavity, Thoracic drainage tubes time, air leakage, bronchopleural fistula, complication and mortality between capitonnage and uncapitonnage method in complicated pulmonary hydatid cyst.

MATERIALS AND METHODS

The hospital records of 221 patients who under went surgery for pulmonary hydatid cysts in Razi and Arya hospital in Guilan University of Medical Sciences, Rasht, Iran between 2009 and 2014 retrospectively were reviewed. Of these patients, 75 were complicated (intra bronchial (Fig. 1a, b), intra pleural rupture (Fig. 2a-c), infection (Fig. 3a-c), pneumonia (Fig. 4a-c) and air entrance between pericyst and laminated membrane (Fig. 5a-c) and operated on by with capitonnage (36) and uncapitonnage (39). Choice of method was made preoperatively. The 2 groups were compared with respect to postoperative complications, mortality, duration of hospitalization, post operative drainage time, postoperative air leak (>4 days), time of radiologic improvement and recurrencerate. The criteria for radiologic improvement were loss of the shadow of the operated cyst and resorption ofthe postoperative pericystic inflammation and pleuralreaction. Radiologic evaluation was performed incollaboration with the radiology department. All patients who underwent lobectomy, wedge resection and segmentectomy

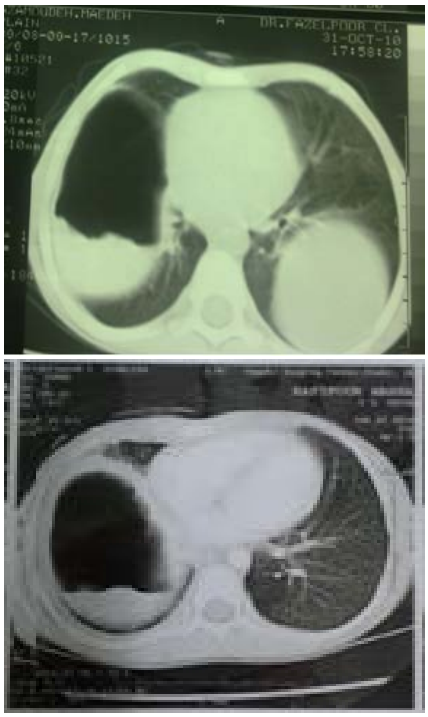

Fig. 1: Intra bronchialrupture (water lily sign)

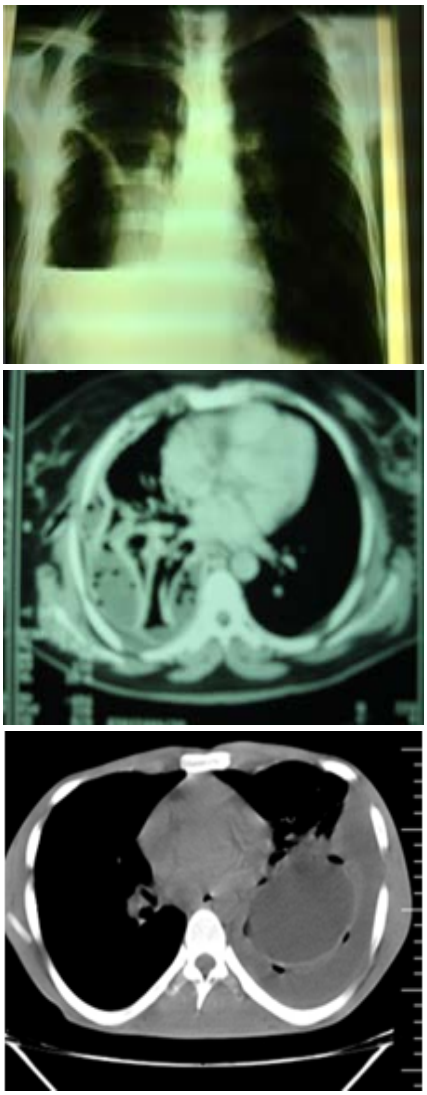

Fig. 2: Hydatidcyst with intrapleural ruptures (pleural effusion, hydropneumothrax)

Fig. 3: Abscess formation

excluded of this study. Each group underwent the same standard preoperative preparation and postoperative management. The preoperative diagnosis was based primarily on chest radiographs and CT-scan. Abdominal ultrasound was used to assess liver involvement. However, a definite diagnosis was established by postoperative pathological assessment. Due to their lack of specificity, immunologic and serologic tests were not used routinely.

A posterolateral thoracotomy through the 5th or 6th intercostal space was accomplished with the patient inthe lateral decubitus position in 51 patients. Fourteen patients underwent anterolateral thoracotomy through the 5th or 6th intercostal space. Another nine patients with bilateral involvement underwent one staged

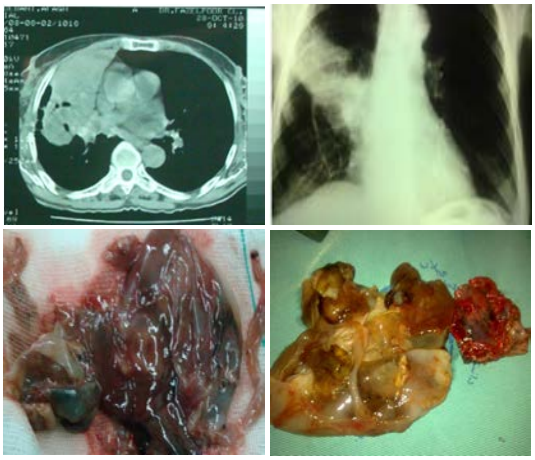

Fig. 4: Recurrent pneumonia

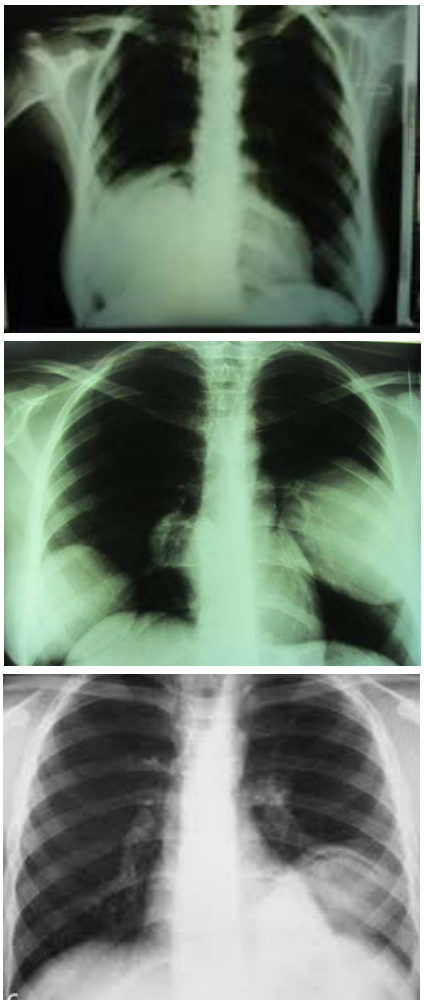

Fig. 5: Intrabronchial rupture, air between pericyst and laminated membrane

Fig. 6: Capitonnage method in remnant cavity after evacuation of laminated membrane and pericyst

Antero-lateral thoracotomy, four of this patients with concomitant liver hydatid cyst underwent one stage anterolateral thoracotomy and pherenotomy and evacuation of cyst when the complicated hydatid cyst was identified, the surgical wound and adjacent lung tissuewere covered with gauze with 7% hypertonic saline. Cystotomy and removal of the laminated membrane were performed. Inpatients treated without capitonnage, only the bronchial openings were closed with 2/0 vicryl obliteration of residual cavity wasnot performed. In the capitonnage group, large bronchial openings were closed with 2/0 vicryl suture and then residual cavity was obliterated with imbricating sutures from within (Fig. 6). The uncapitonnage and capitonnage groups were compared using the unpaired t-test for two independent groups. Values of $p<0.05$ were accepted as significant.

Table 1: Age, gender and preoperative complications

	(CTG)		(UCTG)		Total		
Variables	No	%	No	%	No	%	p-values
Age	30.00±17.08		37.28±18.52		33.79±18.1		0.080
Gender							
Female	17	47.2	17	43.6	34	45.3	0.752
Male	19	52.8	22	56.4	41	54.7	
Total	36	100	39	100	75	100	
Preoperative complication							
Intrabronchial rupture	13	36.1	16	41	29	38.7	0.692
Impending to* complications	12	33.3	14	35.9	26	34.7	
Hydropneumo thorax	5	13.9	4	10.3	9	12	
Pneumonia	1	2.8	0	01	1.3		
Pleural effusion	1	2.8	0	0	1	1.3	
Infection	1	2.8	2	5.1	3	4	
Parapneumonic effusion	2	5.6	0	0	2	2.7	
Pneumothorax	1	2.8	1	2.6	2	2.7	
Pyopneunomo	0	0	2	5.1	2	2.7	

*CXR and CT-scan of the chest reveled air between pericyst and laminated membrane (Fig. 1)

RESULTS

In our study 75 pulmonary hydatid cyst in two groups with Capitonnage (CTG) (36) and Uncapitonnage (UCTG) (39) was surveyed. The mean age 38 years (range, 12-75 years) mean age and standard deviation in (CTG) was (30±17.1) yearand patient (UCTG) was (37.3±18.5) years. In (CTG) 52.8% of patients was male and 47.2% females and in NCTG 56.4% of patients was male and 43.6% females, there were not significantly different between number of gender in 2 groups ($p = 0.08$). Approximately, 72% of the patients lived in rural areas. The follow-up times for all 75 patients ranged from 8 months to 4 years. In both groups, lower lobe involvement by the cyst was more frequent than upper lobe involvement and there was a total of 42 cysts in the 39 (UCTG) and 48 cysts in the 36 (CTG). The most prominent symptoms in both groups were cough and thoracic pain. The (91%) patients in the (UCTG) and (94%) of the (CTG) were symptomatic there were not significantly different between groups ($p = 0.692$). The most common preoperative complications in our patients was intrabronchial rupture, infection, hydropneumo thorax. Demographics of patients was shown in Table 1. The most common tools for diagnosis was Chest radiography and CT-scan of the chest (Fig. 1). The most Approaches to surgery was cystostomy and evacuation, bronchial opening closure, other approaches are shown in Table 2. Hospital stay in our study in (UCTG) was 6.49±1.05 day and in (CTG) was 3+0.75±0.65 day ($p<0.0001$). Mean average day for removal chest-tube in (CTG) was 4.64±0.99 day and in (UCTG) was 2.64±0.59

Table 2: Approaches to surgery

	Capitonnage		Uncapitonnage		Total		
Variables	No.	%	No.	%	No	%	p-values
Approaches to surgery							
Cystostomy+EBOC**	10	24	25.6	66.7	34	45.3	0.0001
Pericystectomy,	17	2	43.6	5.6	19	25.3	
EBOC+Cystostomys	12	8	30.8	22.2	20	26.7	
Decortication							
Cystostomy+EBOC	0	2	0.0	5.6	2	2.7	
EBOC+Decortication							
Total	39	36	100.0	100.0	75	100.0	

**Evacuation bronchial opening closure

Table 3: Hospital stay, removal chest-tubetime and cumulating the fluid in cavity

Variables	Capitonnage	NU*	Mean	SD	p-values*
Hospital stay (day)	Yes	36	3.75	0.65	0.0001
	No	39	6.49	1.05	
Removal chest-	Yes	36	2.64	0.59	0.0001
tube time (day)	No	36	4.64	0.99	
Remnant cavity	Yes	36	1.61	0.49	0.0001
(month)	No	39	3.79	0.61	

T: Test independenty; *NU: Number

Table 4: Postoperative air leak and postoperative bronchopleural fistula

	(CTG)		(NCTG)		Total		
Variables	NU*	%	NU	%	NU	%	p-values
Postoperative air leak							
Yes	3	8.3	10	25.6	13	17.3	0.0480
No	33	91.7	29	74.4	62	82.7	-
Postoperative bronchopleural fistula							
Yes	3	8.3	9	23.1	12	16.0	0.0820
No	33	91.7	30	76.9	63	84.0	-

T: Test independenty; *NU: Number

Table 5: Postoperative morbidity

Causes	CTG (n = 36)	UCTG (n = 39)
Wound infection	3	2
Atelectasis	2	2
Pneumonia	2	1
Empyema	2	1
Hemoptysis	1	2
Pleural effusion	2	1

day ($p<0.0001$) and also cumulating the fluid in cavity in (UCTG) was 3.79±0.61 and in (CTG) was 1.61±0.49 month. ($p<0.0001$) others data was shown in Table 3. In survey of postoperative bronchopleural fistula in (UCTG) was 8.3% and in (CTG) was 23.1 % ($p = 0.48$) and post-operative air leak in patients (UCTG) was 25.6% and in (CTG) group was 8.3% ($p = 0.48$) others data was shown in Table 4. Atelectasis developed in four patients and managed by intensive respiratory physiotherapy and nasotracheal aspiration and thoracic empyema developed in two patients managed by chest tube drainage. Others postoperative morbidity was shown in Table 5. Mortality were not seen in two groups. There was no recurrence of hydatid cyst in during follow up period in this series.

DISCUSSION

Hydatidosis are an endemic disease to many parts of the world as Mediterranean region, New Zealand, the Middle East and South America. In the endemic areas, it is still a major public health problem (Aghajanzade *et al*., 2008; Eisa *et al*., 2013). Most of the patients are young and the majority are <40 years of age (Kuzucu *et al*., 2004; Aghajanzade *et al*., 2008; Bagheri *et al*., 2011; Sonmez *et al*., 2001; Dogan *et al*., 1989). All Pulmonary Hydatid Cysts (PHC) carry the risk of rupture. (PHC) usually remain asymptomatic until the time of rupture or compress the lung and cyst become complicated (Kuzucu *et al*., 2004; Aghajanzade *et al*., 2008; Bagheri *et al*., 2011; Dogan *et al*., 1989). Most intact pulmonary hydatid cysts are asymptomatic and found incidentally and others patient presents with cough, dyspnea and chest pain, especially in huge or centrallocated PHC usually are symptomatic (Bagheri *et al*., 2011). The clinical pictures of complicated cysts is depends on the nature of the perforation (Aghajanzade *et al*., 2008; Bagheri *et al*., 2011; Sonmez *et al*., 2001; Turna *et al*., 2002). In most cases the cyst ruptures into a bronchus in such cases, fluid of cysts evacuate by cough and solid remnants of the collapsed parasitic membrane are left in the cavity as a source of recurrent infection and pneumonia (Kuzucu *et al*., 2004; Peleg *et al*., 1985; Ramos *et al*., 2001; Aarons, 1999). Three patient in our study presented with recurrent pneumonia (Fig. 4). Such patients present with cough and expectoration of hydatid fluid and remnants of parasitic membrane, hemoptysis, fever or expectoration purulent sputum of cystic contents can lead to acute respiratory failure, massive hemoptysis and anaphylactic shock (Bagheri *et al*., 2011; Ramos *et al*., 2001; Aarons, 1999). Three of our patients presented with recurrent pneumonia (Fig. 4).

Rupture of a PHC into the pleural cavity usually causes pneumothorax, pleural effusion or empyema and tension pneumothorax (Kurkcuoglu *et al*., 2002a; Aghajanzadeh *et al*., 2014; Aribas *et al*., 2002) tension pneumothorax occurred intwo of our patients. Simple pneumothorax in patients with pulmonary hydatidosis ranged from 2.4-6.2% and Empyema is reported to occur in 7.6% of patients (Aribas *et al*., 2002; Safioleas *et al*., 1999). In our series, pneumothorax occurred preoperatively in 2.6% of all patients, empyema in two patients, allergic and anaphylactic reaction was reported (Sonmez *et al*., 2001; Yalcinkaya *et al*., 1999; Eisa *et al*., 2013; Dogan *et al*., 1989; Peleg *et al*., 1985; Ramos *et al*., 2001). Two patients in our series had an allergic episode. Optimal treatment of PHCs are surgery but

some researchers recommended, treatment with oral mebendazole or albendazole (Kuzucu *et al*., 2004; Sonmez *et al*., 2001; Grande *et al*., 1993; Keramidas *et al*., 2004). Reports in the literature of treatment with mebendazole or albendazole have been documented as requiring urgent surgery due to massive hemoptysis and severe hypersensitivity reactions (Kuzucu *et al*., 2004; Sonmez *et al*., 2001; Kramidas, 2004; Kurkcuoglu *et al*., 2002b). This medical treatment should only be used for patients who are high surgical risks or for preventing recurrence (Sonmez *et al*., 2001; Grande *et al*., 1993; Kramidas, 2004; Kurkcuoglu *et al*., 2002a). The most appropriate procedures for PHCs are open surgery involving removal of the cyst membrane, closure of the bronchial openings with capitonnage or uncapitonnage (Kuzucu *et al*., 2004; Bagheri *et al*., 2011; Ramos *et al*., 2001; Aarons, 1999; Safioleas *et al*., 1999). About capitonnage and uncapitonnag there are some controversial, specially in complicated PHCs and there are not an accurate paper in literature about capitonnage. Therefore in this study we want to show advantages of capitonnage to uncapitonnage techniquein complicated PHCs. Because hospital stay, removal chest-tube time, cumulating the fluid in cavity, bronchopleural fistula and Postoperative air leak in capitonnage patients arelower than uncapitonnage technique. After removal of the cyst, the residual cavity which is devoid of epithelium and always has some bronchial openings, must be managed appropriately (Bagheri *et al*., 2011; Xanthakis *et al*., 1972; Ramos *et al*., 2001). However, Cystectomy and pulmonary resection have been advocated as the best ways of decreasing the most frequent postoperative complications as bronchopleural fistula and post-cystectomy residual cavity (Xanthakis *et al*., 1972; Peleg *et al*., 1985; Aribas *et al*., 2002; Safioleas *et al*., 1999). In this series, no residual cavity was encounteredin the uncapitonnage cases. In cystotomy with capitonnage, thereis no loss of lung tissue; the uncapitonnage method also avoids parenchymal loss (Eisa *et al*., 2013; Balci *et al*., 2002). Cases of large peripheral cysts in children, capitonnage could restrict expansion of the lung (Grande *et al*., 1993). With positive pressure during anesthesia, capitonnage sutures could lead to laceration of the pulmonary tissue, especially incomplicated cysts (Kuzucu *et al*., 2004). The imbricating sutures of capitonnage could damage vessels and bronchia, resulting in bleeding and atelectasis (Kuzucu *et al*., 2004; Eren *et al*., 2005). In our patient with capitonnage we have two patients with mild hemoptysis. Capitonnage has been reported to have no advantage in the long term and it could lead to distortion of the residual lobe or lung (Grande *et al*., 1993; Eren *et al*., 2005). Thus, it hasbeen performed in small cysts only (Erdogan *et al*., 2005; Grande *et al*., 1993). We used capitonnag in large cysts in our patients but we don't see any distortion or wrinkle of lung. In past our approach to residual cavity after cystotomy been only sutured bronchial openings and resection of fibrotic tissue of pericyst (Ramos *et al*., 2001). Although, Eren *et al*. (2005) and coworker encountered higher morbidity in uncapitonnagepatients, re-operation due to complications was neededonly among capitonnage cases. Higher morbidity maybe due to discharge or relaxation of sutures that settle onbronchial openings in patients with infected or perforate cyst (Eren *et al*., 2005) but we don't see such complications in our patients.

In a study Akin Kuzucu reported, after the bronchial openings were closed, the residual cavity was obliterated with separate purse-string sutures that were placed into the cavity from the deepest level to the surface (capitonnage). In the complicated cysts that exhibited a thickened or liquified pericyst layer and had damaged the adjacent parenchyma, the closure of bronchial fistulas was done more carefully using deeper sutures and placing them closer together as our patients (Kuzucu *et al*., 2004). In our patients after cystotomy and evacuation of laminated membrane, debrises, remnant cavity irrigate with normal saline carefully and bronchial opening closed, final capitonnage performed. In another study, Sonmez *et al*. (2001) reported which the hospital stays was not difference between capitonnage and uncapitonnage groups whereas it was significant statistically between chest tube removal days ($p<0.05$). Capitonnage shortens postoperative chest tube drainage period in PHCs, resulting in lower morbidity compared with the uncapitonnage cases (Sonmez *et al*., 2001) asourstudy. The uncapitonnage method may be a good alternative to the capitonnage procedure for PHCs. Better management of bronchial openings should improve the results of the uncapitonnage method (Eren *et al*., 2005).

A study reported which total hospitalization time of CTG and UCTG was same inuncomplicated PHC, ($p = 0.03$). But there was significant statistical difference between CTG and UCTG groups regarding the development of prolonged air leak and empyema. There was no significant difference regarding the rate of disease recurrence between CTG and UCTG. No mortality was reported in both groups. Safioleas *et al*. (1999) reported median hospital stay for uncomplicated cases was 12 days median stay was 21 days for complicated cases for PHCs. Complicated PHCs are associated with higher postoperative morbidity and mortality than uncomplicated cysts (Aribas *et al*., 2002; Dogan *et al*., 1989; Balci *et al*., 2002). In complicated cases, infection and inflammation of the adjacent lung parenchyma may affect wound healing and lead to postoperative complications such as prolonged air leakage, empyema and pneumonia. Eisa *et al*. (2013) reported their experiences and recommended that Cystotomy and capitonnage method is safe and easy method for surgical managementin PHCs. Recent years our policy

in complicated PHCs are evacuation, irrigationremnant cavity of cyst and capitonnage. The complications in CTG and UNCTG was not significant (empyema, atelectasis, wound infection, hemoptysis and pneumonia) but removal chest-tube time, cumulating the fluid in cavity, bronchopleural fistula and post-operative air leak significantly was lower than in CTG. There was no mortality, recurrences in either of our patient groups.

CONCLUSION

Surgery is the safest curative treatment for complicated as well as uncomplicated pulmonary hydatid cysts. Surgical intervention before rupture of the cysts is essential. Ruptured cysts are associated with increased morbidity, more extensive surgery and longer hospital stays. All pulmonary hydatid cysts should be surgically treated as soon as they are diagnosed in order to avoid complications. The capitonnage method has more advantages in surgery of complicated pulmonary hydatid cysts and Capitonnage shortens postoperative chest tube drainage period, hospital stay, cumulating the fluid in cavity, bronchopleural fistula and post-operative air leakand resulting in lower morbidity.

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