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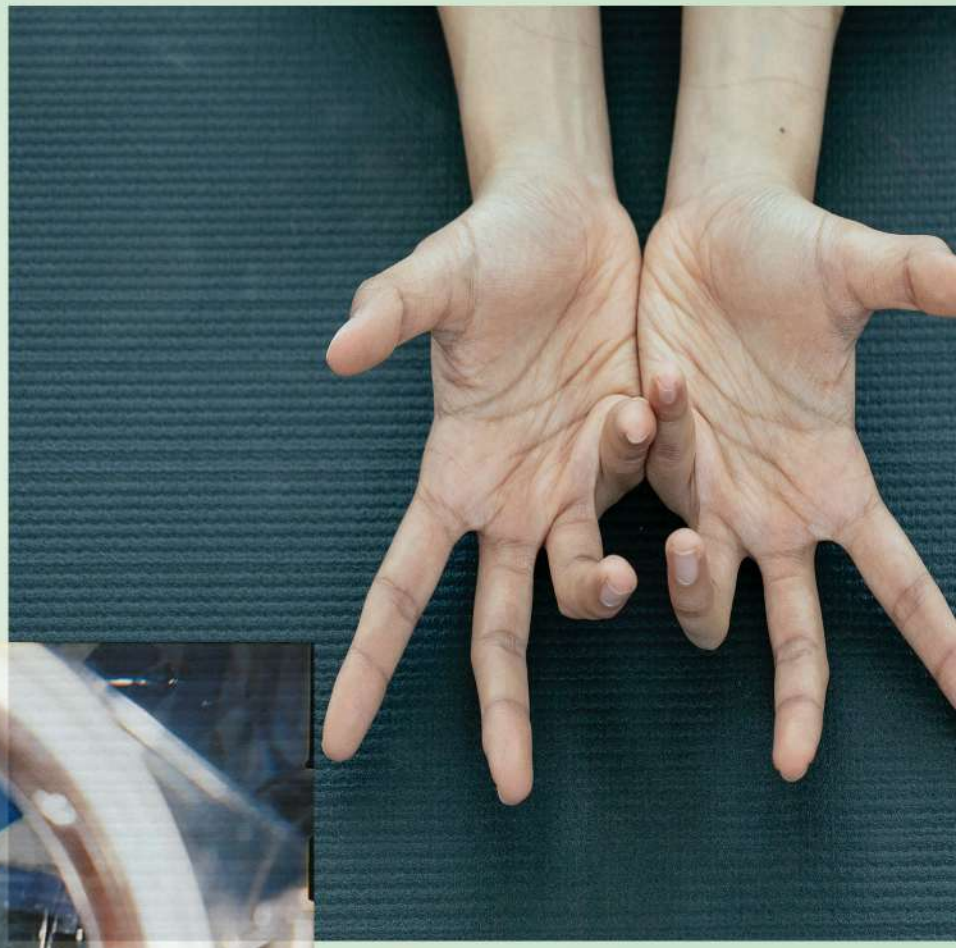

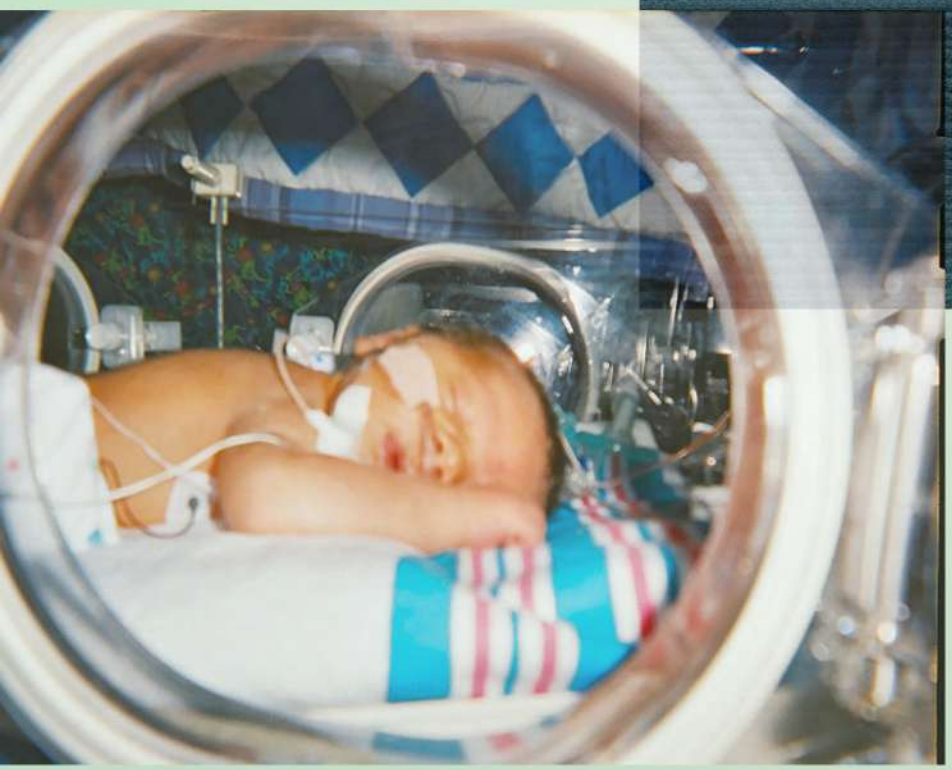

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Original Research

Seventy Cases of Partial Gastric Pull-Up According to the Schärli Technique for Esophageal Replacement in Pediatrics

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ABSTRACT

Introduction

Esophageal replacement (ER) is indicated in patients with long gap esophageal atresia (LGEA) or failure of the primary anastomosis. Also, severe caustic or peptic strictures, resistant to conservative treatment with medication and dilations, may require an ER. Numerous techniques with different organs and routes have been described, all with satisfactory results.

Objective

Our objective is to describe the experience obtained with partial gastric pull-up according to the Schärli principles (SGPA).

Materials and Methods

Medical records of patients who required a SGPA between October 1995 to June 2018 were reviewed. The analysis was observational, longitudinal, retro-prospective and descriptive. Epidemiological data, surgical aspects and postoperative complications of the patients were considered.

Results

Seventy ER were performed with SGPA. The indication was esophageal atresia (EA) in 58 cases (44 long gap and 14 failure of the anastomosis), 10 caustic strictures, one peptic stricture resistant to conservative treatment and the other due to a retained foreign body. The age of the ER was on average 2-years and 9-months. The route was: posterior mediastinal (35), retrosternal (29) and transpleural (6); without a thoracic approach in 59 patients. The duration of the procedure was 4.7-hours on average. There were 13 cases of intraoperative complications, 8 cases of pneumothorax, 5 bleeding injuries (3 in the spleen, 1 liver and 1 cervical), and one injury to the cervical trachea. Anastomotic dehiscence was observed in 37 patients (52%), which closed spontaneously after an average of 17.8-days in all except one patient. Thirty-one patients (44%) developed anastomotic stenosis, requiring redo anastomosis in 6 cases; 37% developed dumping and 23% gastroesophageal reflux disease. There were 3 deaths (4.2%): all in EA patients with associated malformations, following a morbid postoperative period with infectious complications in intensive care at 10-days, 7 and 8-months after ER. Follow-up was an average of 8-years. All 67 living patients are currently tolerating oral feeding.

Conclusion

In our experience with ER using SGPA, we observed a high incidence of complications, generating a longer post-operative evolution and with greater morbidity than that described in other techniques. Given these results, we changed our strategy to complete gastric transposition, in order to reduce morbidity and improve the evolution of this complex group of patients.

Keywords

Esophageal replacement; Partial gastric pull-up; Esophageal atresia; Caustic stricture.

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BACKGROUND

The resolution of complex esophageal atresia (EA) and esophageal strictures that are resistant to conservative treatment with dilations is challenging and remains controversial. Although, it is widely accepted in the literature that the esophagus itself is the best option for complex esophageal pathologies[1,2] there are certain circumstances in which loss of the organ is unavoidable and esophageal replacement (ER) is necessary. The ideal esophageal substitute should conform in function as far as possible to the original structure.[3] The patient should be able to swallow normally and experience no reflux symptoms.[4] An additional requisition in children is that the substitute should continue functioning for many decades without deterioration.[5] There is no perfect method for esophageal replacement due to the complexity of its reconstruction. Satisfactory results have been published with different organs and routes, all with both immediate and long-term complications; although the numbers reported are small, and long-term results are scarce.[3-6]

Most of the indications for ER are long gap esophageal atresia (LGEA) or severe anastomotic complications that end in the loss of the organ; and caustic (CS) or peptic strictures resistant to conservative treatment with medication and periodic dilations.[5,6]

In 1992 Schärli[7-9] described a technique that allows the preservation of the lower esophagus and the cardia, with mobilization and elongation of the stomach by means of an incision in the lesser gastric curvature and the ligation and section of the stomachic coronary artery, called partial gastric transfer with preservation of the cardia and distal esophagus.

The objective of this report is to analyze the evolution of a group of patients with complex esophageal pathology, in which we used an ER with partial gastric pull-up according to Schärli's principles (SGPA).

MATERIALS AND METHODS

Medical records of all patients who received a SGPA at Hospital Prof. Dr. Juan P. Garrahan between October 1995 and June 2018 were reviewed. The analysis was observational, longitudinal, retroprospective and descriptive. The following epidemiological data were analyzed: sex, underlying disease and reason for replacement, anatomical types of EA, associated malformations (only in the EA group), number of surgical procedures prior to ER, previous thoracotomies, and age at the time of ER. The surgical procedure was recorded: method used (conventional or video-assisted), intraoperative complications, use of thoracic approach, esophagectomy (in the CS group) and procedure duration (in hours). The following data were taken from the post-operative period: days of mechanical ventilation (MV), days of hospitalization in the intensive care unit (ICU), days of total hospitalization and the start of oral feeding.

Early post-operative complications such as anastomotic leakage, and late post-operative complications such as anastomotic stenosis, airway fistulas, gastroesophageal reflux disease (GERD), and dumping syndrome were investigated. Gastric necrosis, number of reoperations (related to the ER procedure), death, and follow-up in years were also considered. Enteral feeding status was verified at the time of this review. We define dehiscence of the anastomosis by observation of saliva through the cervical wound or leakage of contrast material during esophagogram. We consider stenosis of the anastomosis to be an area of reduced caliber in the esophagogram, requiring at least one esophageal dilation; whether or not it is associated with dysphagia. Dumping syndrome was diagnosed by the development of recurrent postprandial abdominal pain and distention, diarrhea, sweating, paleness and drowsiness, for a period of at least three months. This was always confirmed by abnormal oral glucose tolerance test. GERD is diagnosed by upper gastrointestinal endoscopy in post-operative controls, when observing signs of esophagitis. The digestive fistula to the airway is diagnosed with respiratory symptoms during oral feeding, with an esophagogram and confirmed with respiratory endoscopy.

Pre-Surgical Preparation

Before ER, the patient should be stimulated with "sham feeding" (oral feeding with exit of the material ingested through the cervical esophagostomy), from the moment the patient presents esophagostomy, to facilitate the initiation of oral feeding in the immediate post-operative period. In patients with a diagnosis of EA, a presurgical computed tomography angiography of the upper thorax and cervical region is performed, to investigate the presence of previously undiagnosed vascular rings, and in case they are, choose for the retrosternal pull-up route (RER) to perform the ER, avoiding the posterior mediastinal route (PMR). For positioning the patient, we use transverse enhancement in the thoracoabdominal junction and another cervical one, hyperextending the neck, to achieve a good exposure of the cervical esophagus and the esophageal hiatus, and align the mediastinum. A plastic tube can be left in place as a tutor during removal of the esophagostomy. The operative field must involve both the abdomen and the cervical region, as well as the thorax.

Abdominal Step

Through a median supraumbilical incision, the left lobe of the liver is mobilized and adhesions to the gastrostomy area (GTT) are released, removing it and neatly closing it in two planes. The cardia and hiatus are exposed to free the lower esophagus, which is dissected blindly and bluntly into the posterior mediastinum until it is brought into the abdomen in cases of EA and up to the maximum possible height in CS. The stomachic coronary artery (left gastric artery) is ligated after giving its second gastric branch, which allows it to gain 2-3 cm in length, the lesser curvature of the lesser omentum is released from the pylorus to the diaphragmatic hiatus, and the adhesions to the body and gastric fundus are freed with preservation of posterior gastric vessels and short vessels. In this way, the gastroesophageal junction can be mobilized outside the abdominal incision. According to the principles of Schärli,[7,8] a cutting linear mechanical suture is placed in the lesser curvature, perpendicular or oblique to the stomach[10] (achieving better gastric tubulization). Then, a thick probe is introduce in the created gastric tube, to ensure its adequate caliber. Mechanical suture is reinforced

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with resorbable seromuscular imbricating plane stitches.

Section of the lesser curvature allows to mobilize the esophagus and the cardia about 4-6 more centimeters (twice the length of the mechanical shot). If the distal esophagus loses vitality or is very small, it is resected and the anastomosis is performed over the cardia. In case of using the PMR, it is dissected from the esophageal hiatus towards the proximal, in a blunt and blind way until reaching the tracheal carina, in front of the vertebral column. If it is a CS, the esophagus will be freed on all sides in order to resect it. If the decision is made to perform RER, the esophageal hiatus is closed with separate non-absorbable sutures. A tunnel is carved immediately below the xiphoid, following the posterior aspect of the sternum, advancing towards the suprasternal hollow as much as possible. Kocher´s maneuver is performed to obtain maximum mobility of the pylorus to pull-up the stomach with the least possible tension. We did not perform pyloromyotomy or pyloroplasty or anterior partial fundoplication to restore the angle of the patient.

Cervical Step

The esophagostomy is disassembled and the proximal esophagus is freed 3-4 cm, taking care not to injure the cervical nerve bundle or the recurrent laryngeal nerve. The dissection of the esophagus should be close to its wall without perforating or devitalizing it. In the PMR, using blunt dissection, a path is made between the posterior aspect of the trachea (membranous) and in front of the prevertebral fascia, creating a tunnel with entrance to the posterior mediastinum (native esophagus route). The same maneuver is performed through the esophageal hiatus between the posterior cardiac aspect and the prevertebral plane simultaneously to unify the tunnel. In cases of CS, esophagectomy is performed. In RER, a space is created immediately behind the sternum following its posterior aspect in an abdominal direction. Special care is taken in not injuring the innominate vascular bundle. To avoid compression, the sternoclavicular muscle attachments can be released. Once the continuity of the tunnel is established, it must be widened so that it is occupied by the thickness of at least two fingers.

Reconstruction

A rubber catheter or Nelaton® catheter with vaseline jelly is placed through the tract, either using the PMR or RER, and the abdominal end of the catheter is attached with two points to the distal esophagus or cardia (if the distal esophagus was resected). They are identified with two sutures of different lengths, to ensure adequate gastric pull-up, avoiding torsion of the organ. From the cervicotomy, delicate traction of the probe is performed, guiding it from the abdomen, to ensure an adequate pull-up. The esophagoesophageal or esophagocardial anastomosis is performed in a full-wall plane with separate absorbable suture stitches in the neck. When finishing the posterior layer, a nasogastric tube is placed, to help finalize the anastomosis and avoid acute gastric distention in the immediate post-operative period. A rubber drainage is left in the cervical wound. In PMR, the ascended stomach is fixed to the diaphragmatic hiatus with non-absorbable suture to avoid herniations of abdominal organs towards the thorax and in RER it is anchored to the surrounding tissues. In those patients in whom a sufficient portion of the abdominal stomach remains after performing the ER, we perform a GTT, as described by Schärli, with placement of a gastrojejunal tube; and in those in which this is not possible due to the lack of an adequate gastric reservoir in the abdomen after pulling it up, we perform a feeding jejunostomy for the first post-operative days, until complete oral nutrition is established. If the use of the transpleural pull-up route (TPR) is necessary, it is performed through a classic thoracotomy. In patients with video-assisted approach, the same steps performed in conventional surgery are reproduced using five working paths; one umbilical 10 mm port for the optics, another of 10 mm in the right upper quadrant for mechanical suture introduction, and 3 ports of 5 mm: left, subxiphoid and left flank, similar to the laparoscopic Nissen fundoplication approach. Vascular sealant (Ligasure®) is used to treat the vessels. The mediastinal esophageal dissection is performed under direct vision (mediastinoscopy) until reaching the cervical area through the prevertebral plane. The gastric lesser curvature is sectioned with a cutting linear mechanical suture according to the technique. The stomach is guided towards the neck under laparoscopic control.

Post-operative Management

An oral esophagogram is performed, starting on the seventh post-operative day to detect early complications. If there is no leakage of the contrast material and there is good distal passage, the cervical drain and nasogastric tube are removed and oral feeding is started. Long-term esophagograms allow finding late complications.

RESULTS

Seventy SGPA were carried out in a 22-year period. Thirty-five patients were male. The indications were EA in 58 cases (82.8%), CS in 10 cases (14.2%), GERD in 1 case (1.5%) and the remaining was a case of retained foreign body (1.5%). Within the EA group, 44 were long gap ends (absence of the esophagus) and 14 were anastomotic failure (esophageal loss). In all cases of CS, the indication for ER was recurrent stenosis after conservative treatment with periodic dilations, as in the patient with peptic stenosis. The anatomical types in the EA group were: type I 29 (50%), II 2 (3.4%), III 24 (41.4%) and IV 3 (5.2%). Fifty percent of the EA´s (29/58) had associated malformations, most of them cardiovascular. Regarding the number of surgical procedures prior to ER, an average of 2.7 procedures was performed per patient (0-8). Forty-two percent of the patients had thoracotomies prior to ER (12 of them had 2 thoracotomies, and 2 patients had 3). The average age at the time of ER was 2-years and 9-months (2-months. 18-years). The method used for the SGPA was conventional in 66 cases and video-assisted in 4 (two of which were converted due to the presence of severe adhesions). In one case, thoracoscopy was used to perform the anastomosis. The routes used were: 35 PMR (50%), 29 RER (41.4%) and 5 TPR (8.6%).

Thirteen patients (18%) presented intraoperative complications: 8 pneumothorax (4 bilateral, 2 right and 2 left), which were

drained in the operating room; 5 bleeding injuries: 3 splenic, one hepatic when dismantling the GTT from surrounding adhesions, and one cervical vascular injury, when a tributary branch of the jugular vein was injured. All of these were resolved with hemostatic suture. A patient with a splenic lesion also presented a punctate perforation in the cervical trachea during esophagostomy release, which was sutured with two separate stitches of non-absorbable material. The thoracic approach was used in 11 cases (15.7%): 9 thoracotomies as the first surgical procedure (4 in CS to perform esophagectomy, 3 in EA to evaluate the possibility of anastomosis, and 2 in EA to close a tracheoesophageal fistula). One thoracotomy was necessary after the abdominal approach for esophageal release, and other thoracoscopy was approached for intrathoracic anastomosis of the ER. Esophagectomy was performed in 9 of the 10 cases of CS. The esophagus was left abandoned in only one patient due severe adhesions to the vascular elements of the mediastinum. The mean ER duration was 4.7-hours (3-10). All cases were post-operatively managed in the ICU. Hospitalization data is summarized in table N°1 (Table 1).

Table 1. *Hospitalization Data*

	Days	Range
ICU	17	3-241
MV	11	0-240
Total	42	10-241
Oral intake	22	7-65

ICU: intensive care unit
MV: mechanical ventilation

As early post-operative complications, anastomotic dehiscence was evidenced in 37 cases (52.8%). Thirty-six closed spontaneously in an average of 17.8-days (4-69). Only in one case surgical closure was needed. Closure was performed at 39-days, being one of the first cases in the series. Late complications reported 31 stenoses of the anastomosis (44%). Of the 31 strictures, 21 (68%) had previous history of dehiscence. All were treated with dilations, with an average of 4.2 dilations per patient (1-14). Six patients required resection of the stenosis and redo anastomosis due to failure of conservative management with dilations (one of these 6 required a second redo). Two ER by PMR with primary diagnosis of type III EA (whose primary anastomosis failed evolving with dehiscence and dehiscence plus tracheoesophageal refistula) presented fistula from the ER to the airway, which were treated surgically to separate the airway from the digestive tract. One of them required a pneumonectomy. Twenty-three percent (16/70) had signs of GERD in the post-operative endoscopic digestive controls that were successfully managed with increased doses of proton pump inhibitors and dietary measures. Thirty-seven percent of the cases (26/70) presented dumping syndrome, reversing their symptoms with a feeding of raw cornstarch and suspension of simple carbohydrates. One patient with long gap type III EA who underwent an ER by PMR presented severe digestive hemorrhage with hemodynamic decompensation, product of a fistula of the right carotid artery to the cervical anastomosis. The child had an undiagnosed right subclavian artery arising from the aortic arch as a last branch. Successful emergency surgical treatment was performed. There were no cases of gastric necrosis. Ten patients were reoperated (14%): 6 redo anastomosis due to stenosis; 2 fistula closure to the airway (one underwent right pneumonectomy, and in the same patient a hiatoplasty was also performed due to hiatal hernia later on); one esophagocutaneous fistula closure; and one emergency cervical arterioesophageal fistula closure. There were three deaths in our series (4.2%): the first patient was a type III EA III (with associated renal malformation) who presented a dehiscence after the anastomosis, was replaced by PMR and underwent a torpid post-operative period in the ICU after ER due to dehiscence of the anastomosis and stenosis, fistula to the airway that required surgery for its closure (with right pneumonectomy) and another surgery for hiatal hernia, who died 7-months after RE in the context of sepsis with multiple failure. The second one was a long gap type III EA (with associated cardiac and anorectal malformation) that was replaced by RER who died in the ICU 10-days after surgery due to mediastinitis with respiratory distress, active pulmonary bleeding, sepsis by acinetobacter Baumanii and a respiratory syncytial virus. The third one was a long gap type I EA associated with Down syndrome and congenital heart malformation that died 8-months after ER by to RER that evolved with stenosis, which required a series of dilations (without complications) and a tracheostomy because of prolonged intubation. He died of sepsis caused by central venous catheter related infection due to methicillin-resistant *Staphylococcus aureus* following a post-operative period with multiple infections to other resistant germs. This patient had severe pulmonary hypertension caused by a restrictive ostium secundum atrial septal defect and grade II pericardial effusion. Mean follow-up for the series was 8-years (18-months . 18-years). Four patients (5.9%) were lost of follow-up. The 67 living patients are currently tolerating the oral intake, with complementary nutritional support by gastrostomy in 16 cases.

DISCUSSION AND CONCLUSION

Most authors agree that the patient's native esophagus is better than any possible organ used for replacement.[1,2] While this might be theoretically true, the persistent efforts to preserve the native esophagus can be associated to innumerable complications such as multiple operations, mediastinitis, recurrent tracheoesophageal fistulas, long and stubborn strictures, prolonged hospitalizations, nutritional compromise, and pulmonary sequelae, among others, which in turn may lead to the preservation of a completely dysfunctional esophagus. Beginning in 2009, several authors[11,12] started to consider the possibility of performing an early ER, in order to reduce the morbidity associated to the prolonged efforts to preserve the esophagus at all costs. The ideal cases to which this idea could be applied were complex EA cases, but it could expand to patients with long or multiple caustic or peptic strictures resistant to conservative management.[9,12] The goal was to reduce the morbidity and mortality associated to the attempts to preserve the native esophagus by doing an early ER, ideally between 8 and 12-months of age.[5] The mean age at ER in our study was 2.75-years. As there is no ideal substitute for the esophagus, different techniques with their advantages and disadvantages have been published, both for the organ and the chosen route, such as colonic,[3] jejunal[4] interposition; total gastric pull-up,[5] or partial gastric tube,[6] including the Schärli technique, sectioning the lesser gastric

curvature.[7,8] All of them reproducible and with satisfactory results, both in the short and long-term.[10-12] In our department we chose the stomach as the first option for ER, since it is a more uncomplicated technique since it requires a single anastomosis, it reaches the cervical area without major difficulties and also presents excellent vascularization, a fact demonstrated in our series of 70 patients without any case of gastric necrosis. When choosing the stomach, a decision must then be made between a technique that uses the entire stomach[5] or only a partial form of it.[6,7]

Unlike what is described by Schärli, we did not perform pyloromyotomy or pyloroplasty (due to the high percentage of dumping), or anterior partial fundoplication to restore the his angle (since the fundoplication only works correctly when the anatomy of the gastroesophageal junction is preserved). Schärli also does not use PMR or TPR or video-assisted techniques for ER. That's the reason we say that our experience follows "the principles" of Schärli, since we do not reproduce exactly the technique described by him. We modified the orientation of the lesser curvature section with mechanical suture[10] (oblique shape), to better tubulize this gastric segment so that it is not so dilated in the thorax. Furthermore, it was not always possible to perform a GTT as described by the author, since in many cases we noticed the lack of an abdominal gastric reservoir after SGPA, so we performed a feeding jejunostomy in those cases. Also in some cases, we resect the lower esophagus because it is very rudimentary or is devascularized after dissection, performing the anastomosis directly to the cardia. We not only use this technique in patients with a diagnosis of long gap EA, but also extend the indication in those where the anastomosis failed, in caustic and peptic strictures cases, and in retained foreign body sequelae. Our complications, 52.8% dehiscence and 44% stenosis, were higher than other series consulted, regardless of the technique used, when compared with the use of the colon[3] (29 and 13.1%), jejunum[4] (0 and 27%), total gastric pull-up[5] (12 and 19.6%), and gastric tube[6] (9.5 and 9.5%). Two of these series even refer 2.9%[5] and 9.5%[6] of dumping, against 37% in our series. The large number of complications of the anastomosis can be explained by the fact that the gastric section interrupts the submucosal supply and contributes to the vascular problems of the anastomoses that trigger leakages and strictures. This could be minimized by performing a total gastric transposition with anastomosis to the gastric fundus.[5] Supporting our findings, Kudo and collaborators[13] studied the gastric parietal vascular flow with laser Doppler velocimetry and demonstrated in an adult population that the flow is significantly better in the gastric wall in the total gastric pull-up, than in the gastric tubes that suffer wall section. Although we learned that spontaneous resolution of dehiscence occurs in a very high percentage (97.3%) and the management of stenosis is successful in most cases through serial dilations (80.6%), the consequences of the high percentages of complications increase the number of days of hospitalization and delay the times for our patients to acquire complete oral feedings intake. The large amount of dumping could be explained by the modification of the gastric reservoir. It could be assimilated to the dumping observed in the post-operative period of other surgeries, where the gastric reservoir is also reduced, such as antireflux surgery, in cases of GERD,[14] and bariatric surgery, in obese patients.[15] Partial gastric pull-up cases have a smaller gastric reservoir since part of it is tubulized to form the neoesophagus. All cases of dumping had a good response to dietary management, however they prolonged weight and height gain in these patients, in addition of having bothersome symptoms when eating. Comparative studies between total and partial gastric replacement techniques show similar results, both in children[16] and in adults.[17] These last authors conclude that increasing gastric capacity and maintaining the submucosal vascular network provide better results using the entire stomach. The two fistulas to the airway occurred in patients with a history of type C EA in whom we used the PMR route. In both patients, a primary anastomosis was done initially, but was followed by dehiscence and refistula with the subsequent loss of the native esophagus. In these cases, the PMR is more complex, because it is performed blindly blunt on a territory with scars from previous surgeries and complications from anastomosis, and it is closely related to other important organs. This would explain that both cases occurred using the PMR. Fistula from the ER to the airway is a serious complication, which requires aggressive surgery (one of them required pneumonectomy and later died), and which should be prevented by overlapping the mechanical suture, although the same occurred in these cases, despite the fact that we always do the nesting. Other ways to avoid a fistula to the airway would be using a total stomach ER technique[5] (which does not contain a long suture in the mediastinum, a fact that could predispose to communication with the airway, and that only presents the suture of the closure of the GTT); or using RER, at least in cases with a diagnosis of EA with fistula. We have already demonstrated[18] that PMR without thoracotomy was safe for us without severe complications, despite the fact that most of our cases had several previous thoracic surgeries, and we prefer it because it is the shortest and straightest route. Aortoesophageal fistula is a rare cause of gastrointestinal bleeding, usually fatal. It can be acquired due to the presence of a suture in the aorta in contact with the esophagus, as in cardiovascular surgeries; or congenital in cases of vascular rings, associated with prolonged mechanical ventilation and nasogastric tube.[19,20] One patient in the series (EA III) with ER by PMR presented in the post-operative period a severe gastrointestinal bleeding with hemodynamic decompensation, due to a fistula of the right carotid artery to the cervical anastomosis.[21] The child had an undiagnosed right last branch subclavian artery (aberrant right subclavian artery). He had a successful emergency operation. From this case on, we perform a preoperative computed tomography angiography to rule out vascular rings in patients with EA and, if present, RER is used. There are many publications describing aortoesophageal fistulas, but we believe this is the first case described as a complication in the post-operative period of an ER. This complication should be quickly suspected in a post-operative period that presents with digestive bleeding of bright red blood, associated with prolonged tracheal and nasogastric intubation. Immediate exploration is recommended to avoid fatal outcome. The development of minimally invasive methods has made it possible to solve this group of complex patients in a satisfactory way. We use laparoscopy and mediastinoscopy to achieve dissection under vision like other authors[22-24] in four patients, two were converted to conventional surgery because they found severe esophageal adhesions when performing the PMR. Video-assisted surgery reproduces the steps of the conventional procedure, with the advantage of performing an esophageal dissection under direct vision (mediastinoscopy) using precise and

delicate maneuvers, unlike blunt and blind maneuvers. It is a feasible and reproducible alternative in trained centers, but it requires more numerous comparative series to evaluate the results with those of conventional surgery. We believe that the best candidates for this technique are those who do not have previous thoracic surgeries, such as EA I or CS that only received dilations. In our series we had 3 deaths (4.2%), an incidence similar to other series. All occurred as a result of severe systemic infectious complications in the ICU due to resistant germs triggering multiple organ failure and death. All had a history of multiple surgeries and congenital malformations. It is a high percentage of mortality for any type of surgery, but it must be taken into account that ER is a complex surgery in patients having comorbidities. In the literature is not clear which alternative can improve these results. Perhaps morbidity and mortality could be reduced by performing an earlier ER, avoiding unnecessary efforts to retain a diseased esophagus,[11,12] allowing to perform this procedure in a patient in a better clinical condition. We believe that this is the series with the highest number of cases using the Schärli technique as an ER strategy. We have found in the literature 4 reports using the Schärli procedure, two of them belong to Schärli himself and the other two to different authors. In all of them the authors report a small number of cases. Due to the fact that with our experience in SGPA, we had a high incidence of complications, generating longer post-operative hospital stay and greater morbidity than those described in other techniques; we decided to change it to complete gastric transposition,[5] in order to reduce morbidity and improve the evolution of this group of patients. However, we believe that the strategy of sectioning the lesser gastric curvature is useful in those patients in whom an extra elongation is necessary to bring the esophageal ends closer allowing to perform an anastomosis under less tension. We consider that it is important to identify those children who require an early ER, to prevent their clinical deterioration. Currently our technique of choice in ER is the total gastric pull-up by posterior mediastinal route.

INSTITUTIONAL REVIEW BOARD

This study does not need approval by the Institutional Review Board since it is a retrospective study.

CONFLICTS OF INTEREST

The authors declare that they have no conflicts of interest.

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Case Report

A Case Report of Congenital Oesophageal Atresia with Tracheo-Oesophageal Fistula and Review of the Literature

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ABSTRACT

Introduction
Congenital oesophageal atresia (COA) refers to a congenitally interrupted oesophagus. It is commonly referred to in the literature as oesophageal atresia (OA) with or without tracheo oesophageal fistula (TOF) but acquired TOF is a different entity.

Case Report
We present a case of OA with TOF which was not suspected antenatally despite the presence of polyhydramnios. The baby presented with respiratory distress and excessive oral secretions at the age of 3-hours following initiation of breastfeeding. Thoracoscopic repair was performed on the second day of life. The pre-, intra- and post-operative course was smooth with no major challenges. There was no associated anomalies in our case following thorough systemic evaluation including brain ultrasound, abdominal ultrasound, skeletal survey, ophthalmic assessment and echocardiography.

Conclusion
This case demonstrates the importance of maintaining a high index of suspicion for OA when faced with a combination of respiratory distress and persistent frothy oral secretions in a newborn. The antenatal and postnatal diagnostic approaches are discussed with highlights of associated anomalies and pre-operative assessment and management.

Keywords
Congenital oesophageal atresia (COA); Tracheo oesophageal fistula (TOF); Oesophageal atresia (OA); Vertebral defects, anal atresia, cardiac defects, renal anomalies and limb abnormalities (VACTERL); Choanal atresia, retardation of growth, genital abnormalities, and ear abnormalities (CHARGE).

INTRODUCTION

Congenital oesophageal atresia (COA) refers to a congenitally interrupted oesophagus. It is commonly referred to in the literature as oesophageal atresia (OA) with or without tracheo oesophageal fistula (TOF) but acquired TOF is a different entity. The congenital lack of the oesophageal connection with the stomach in OA prevents swallowing, and this in turn prevents normal feeding and may cause the baby to aspirate accumulated saliva or milk leading to aspiration pneumonia. The incidence of COA is 1 case in 3000-4500 births. Maternal polyhydramnios occurs in approximately 33% of mothers with foetuses with OA and distal TOF and in virtually 100% of mothers with foetuses with OA without TOF.

The characteristic presentation of babies with OA is with drooling and excessive oral secretions with or without overt respiratory distress. The baby will typically choke upon starting breast or bottle feeding. This may progress to significant respiratory distress. The diagnosis of OA should be highly suspected if a large bore gastric tube appearance on a plain chest radiograph shows the tube to coil back in the proximal oesophageal pouch. OA occurs in association with other significant congenital anomalies in 30-60% of babies. The associated congenital anomalies are the major source of morbidity and mortality associated with OA. Early identification and management of OA prevents respiratory compromise and improves the outcome.

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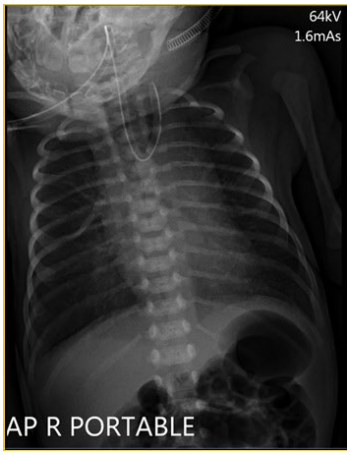
CASE PRESENTATION

A term male baby presented at the age of 3-hours of life with respiratory distress in the form of intermittent oxygen desaturations accompanied with tachypnoea and mild chest retractions. The lowest recorded oxygen saturation was 90% with no pre-post ductal oxygen saturations difference. Drooling and excessive thick oral secretions were noted. The secretions continued despite suction with the bulb syringe. The respiratory rate was 90-100 breath per minute. The baby was breastfed once prior to the onset of the respiratory distress. There was no reported choking. He passed meconium once. The 1st X-ray following admission showed the orogastric feeding tube to coil back inside the oesophagus at the level of the 4th thoracic vertebra (Figure 1). The diagnosis of OA was suspected. Furthermore, the presence of gastrointestinal aeration in the plain X-ray suggested the existence of a TOF. The respiratory distress progressed to mild respiratory acidosis which was managed by heated humidified high-flow nasal cannula. Intravenous antibiotics therapy was commenced following the septic screen. Total parenteral nutrition through peripheral intravenous central catheter (PICC) was initiated as the baby was kept nil by mouth.

Figure 1. *X-ray Chest and Abdomen Showing the Oro-gastric Feeding Coiling Back Inside the Oesophagus at the Level of the 4th Thoracic Vertebra*



The primigravida mother had a normal prenatal anomaly scan at 19-weeks of gestation. The foetal stomach bubble was seen during the anomaly scan with reported normal liquor volume. At 31-weeks of gestation the mother was diagnosed with gestational diabetes and polyhydramnios. A neonatal alert to the presence of polyhydramnios was not triggered to the neonatal team. There was no consanguinity and no family history of similar or other congenital anomalies.

The male baby, weighing 2361 grams, was born at 39+6-weeks of gestation *via* emergency caesarean due to foetal heart decelerations. He was born at Mediclinic Alnoor Hospital, Abu Dhabi, United Arab Emirates. He did not require active resuscitation at birth. The Apgar score was 9(1) and 9(5). The baby initial assessment was normal apart from the observed small for gestational age, low birth weight and mild ankyloglossia. There were no dysmorphic features pointing towards a recognisable malformation syndrome or pattern. The genitalia were normal with bilaterally descended testicles and the anus was patent.

Following the suspicion of tracheoesophageal fistula a large bore orogastric tube (size 10 F) was inserted to a predetermined length above the point of previous coiling inside the upper oesophageal pouch just above the level of the 4th thoracic vertebra. Low pressure continuous suction was applied to allow drainage and the head of bed was elevated to 450 to minimize the risk of aspiration. The paediatric surgeon was notified and the surgery was planned. It was decided not to use Riley double lumen suctioning tube as the surgery was planned for the following morning

Pre-operative evaluation was normal including cranial and renal ultrasounds. Thorough pre-operative assessment showed no signs of associated congenital malformations. The echocardiogram showed the aortic arch to be left-sided. There was non-significant moderate patent ductus arteriosus measuring 4 mm with left to right flow and a patent foramen ovale. Ophthalmic referral was arranged but there was no apparent iris coloboma. The skeletal survey did not show vertebral anomalies or radial dysplasia.

Thorascopic Repair

The baby was operated successfully on day 2 of life where the congenital defect was corrected with end-to-end anastomosis and separation of the fistula. This was done using thorascopic approach for the first time at our institute. The approach was right sided. There was no intra-operative complications and the baby did not require blood transfusion. Following the operation the baby required a short term mechanical ventilatory support. Analgesia was provided as required using the neonatal pain score. He remained nil by mouth for 48-hours following the operation. The baby was commenced on enteral Omeprazole in anticipation of the gastro-oesophageal reflux which is a universal finding in babies with OA. Enteral feeding using continuous milk pump was commenced with exclusive expressed breast milk and gradually increased till the total daily intake was achieved. On the 7th post-operative day, fluoroscopic guided contrast was done, followed by full reinsertion of the gastric tube under fluoroscopic guidance. The contrast study was satisfactory with no post-surgical leak or stenosis at the oesophageal anastomotic junction. Furthermore, the study showed normal caliber and mucosal pattern of the rest of the oesophagus. It is of note there was minimal reflux aspiration which slightly delineated the upper trachea yet it was not through any tracheo-oesophageal connections. No gastric hiatus hernia could be identified.

Bolus enteral feeding was commenced, on day 10 of life, following the contrast study. Recurrent desaturations and occasional bradycardias were noted, but they were all self-correcting. Both disappeared after commencement of infant Gaviscon. The bolus feeding was progressed steadily with no further challenges. Breast feeding was commenced on day 11 of life and the baby was discharged home on day 15 of life. The follow-up in the clinic after one and 2-months showed a well-growing baby with normal growth and development.

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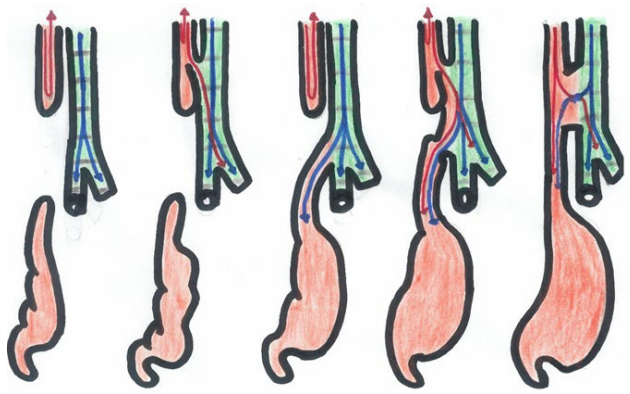
DISCUSSION

Congenital oesophageal atresia incorporates a variety of congenital anomalies with primary congenital interruption of the continuity of the oesophagus with or without a persistent communication with the trachea.[1] William Durston, in 1670, made the first english description of COA in a pair of conjoined twins.[2] In 1697, Thomas Gibson described OA and TOF with the most frequently encountered combination of trache-oesophageal anomalies, a proximal oesophageal atresia with a distal tracheoesophageal fistula.[3] His thorough description clearly described the symptoms and anatomy.

The majority of babies (86%) with OA has oesophageal atresia with distal tracheo-oesophageal fistula.[4] Pure OA (without fistula) is much less common occurring in approximately (6%) of total cases.[4] The characteristic feature of pure congenital oesophageal atresia is a gasless abdomen in the presence of oesophageal atresia. Gupta et al[5] in 2017 described an extremely rare case of an isolated membranous atresia causing near-complete obstruction of the esophagus. Isolated TEF is a very rare condition with scant epidemiological data.[6] Lewis Spitz reported cited an incidence of 4%.[1] The five subtypes of oesophageal atresia (Gross and Vogt Classification) are shown in Figure 2. OA with distal tracheoesophageal fistula is the most frequent type. Our case represents the most common type, type C with the frequency of 86% of all cases.[1]

Figure 2. *The Gross and Vogt Classification for Oesophageal Atresia*



The aetiology of OA is not clearly defined. It is very likely to be a multifactorial complex disease, with a combination of mainly genetic factors and added environmental uterine components.[7] There are no known human teratogens causing OA. However, recurrence of OA in families has been reported. Earlier studies have shown the estimated risk of recurrence in a sibling is 2% when an index case is affected. However, Choinitzki et al[8] results in 2013, in contrast to previous studies, suggested a very low recurrence risk for isolated OA/TOF and/or for malformations of the vertebral defects, anal atresia, cardiac defects, renal anomalies and limb abnormalities (VACTERL/VATER) association spectrum among first-degree relatives. The genetic component is further supported by the occasional association of COA with Trisomies 21, 13, and 18.[8] Tal Weissbach (Prenatal Diagnosis, July 2020) reported an increased incidence of oesophageal atresia with/without TOF (EA/TEF) among twins.[9] Nevertheless, genetic causes can be identified in less than 10% of the patients with EA.[10] Yet, it is estimated that 30-60% of the babies with COA with TOF has associated anomalies.[10] It is recognised, that associated anomalies as well as prematurity-related problems significantly affect the morbidity and mortality of COA babies.[11] The described anomalies include but not limited to tracheoesophageal fistula, urogenital system anomalies, VACTERL/VATER association. Coloboma, heart defects, choanal atresia, retardation of growth, genital abnormalities, and ear abnormalities (CHARGE) are also described.[11] Furthermore, neural tube defects, hydrocephalus, tethered cord and holoprosencephaly has been described in association with OA.[11] Other reported associated anomalies include duodenal atresia, ileal atresia, hypertrophic pyloric stenosis, omphalocele, intestinal malrotation, meckel diverticulum, unilateral pulmonary agenesis, diaphragmatic hernia, undescended testicles, ambiguous genitalia and hypospadias.[11] Porcaro F et al[11] reported airway anomalies such as tracheomalacia and tracheobronchial malformations in more than 40.5% of their cohort of 105 consecutive neonates with OA. This contribute to recurrent respiratory exacerbations due to impairment of the mucociliary function. The prognosis is generally good in the absence of these and other significant comorbidities or extreme prematurity. Concomitant laryngotracheal abnormalities are in particular associated with increased morbidity and higher mortality in babies with OA.[11]

The presence of right aortic arch is a rare finding in babies with OA with an incidence of 5%.[12] A recent multi-institutional retrospective study in the United states concluded surgical repair for neonates with right aortic arch is technically feasible *via* thoracotomy from either chest side.[12] However, the same study reported a higher incidence of anastomotic strictures with the right side approach.

The features suggestive of the diagnosis of OA on antenatal ultrasound are small or absent stomach with or without unexplained polyhydramnios.[11] These findings should not be taken as diagnostic as they have a very low positive predictive value. The assumption of the oesophageal atresia based on these findings may erroneously affect the clinical decisions with implications for timing or site of delivery.[11] Caroline Prady and her colleagues in 2019 published[13] a large systematic review and meta-analysis of the prenatal detection of OA. They concluded ultrasound alone is a poor diagnostic tool for identifying OA prenatally, due to the high rate of false positive diagnoses. They recommended magnetic resonance use plus amniotic fluid analysis following the ultrasound suspicion of OA. This recommendation was proposed many years ago by Langer et al.[14]

The post-natal clinical presentation of undiagnosed OA is variable but primarily reflects the effect of inability to swallow milk or the baby's own oral secretions. Frothy secretions which do not clear or recur after suction are typical findings in the majority of cases of OA. Some babies present with respiratory distress, choking and oxygen desaturations due to the reflux of the accumulated secretions from the hypopharynx into the trachea. The H-type fistulas symptoms are related to their caliber.[15] Gastric distension leading to persistent airway secretions in large fistulas presents

with respiratory distress, whereas small ones present with recurrent cyanotic episodes due to saliva and milk aspiration.[15]

Immediate Management of Suspected Cases

At birth, a 10-12 French gauge gastric tube should be passed through the mouth into the oesophagus for any baby born of a pregnancy complicated by polyhydramnios or if the antenatal scan findings are suggestive of oesophageal atresia. The same should be applied to babies presenting within 24-hours of birth with choking episodes, excessive oral secretions or respiratory distress. For preterm babies size 8 F gastric tube is adequate. Failure to pass the gastric tube beyond 9-10 cm from the lower alveolar ridge has been universally recognized as the classical sign of OA.[1] A plain X-ray of the chest and abdomen would then demonstrates the tip of the gastric tube to coil back within the superior mediastinum. The presence of gas in the stomach and intestine indicates the presence of a distal TOF.[1] However, it should be noted that radiological demonstration of a catheter reaching the stomach does not exclude the diagnosis of OA, as the gastric tube may take an alternative route (through the laryngeal inlet, trachea, tracheoesophageal fistula, and distal oesophagus to reach the stomach), which is a rare but well-known scenario.[16,17]

All babies with suspected OA should be managed within the neonatal intensive care unit as they should be kept nil by mouth with adequate intravenous fluids and nutrition. The baby should be nursed with the head raised around 450. Continuous or frequent intermittent low pressure suction should be applied to prevent the salivary secretions from accumulation as this may lead to aspiration pneumonia. A Replogle tube for continuous drainage of saliva from the upper oesophageal pouch should be considered in cases with very copious secretions or if the surgical repair is likely to be delayed beyond 48-72-hours. It should be positioned 0.5 cm above the distal end of the upper oesophageal pouch.

Broad-spectrum antibiotics (Penicillin or Ampicillin- and Gentamicin) are traditionally administered while preparations are made for surgery. It is advisable to avoid routine endotracheal intubation because of the risk of iatrogenic gastric perforation resulting from stomach distension through ventilation through the TOF. Accurate pre-operative radiological estimation of the gap between the upper and lower oesophageal pouches is essential in order to define the most appropriate surgical plan.[18]

Associated anomalies should be ruled out by thorough clinical examination and radiological assessment as they are mainly responsible for the medium- and long-term prognosis in these patients.[17] Echocardiography is highly advisable prior to the surgical correction. The surgical approach may be modified in the presence of a right-sided aortic arch.

The Surgery and Post-Operative Care

The recent years has seen major advances in the surgical approach for OA and TOF with survival now exceeding 90%.[18] The use of thoracoscopic surgery has minimised the long-term musculoskeletal morbidity associated with open surgery. However, the improved survival in recent decades is most attributable to better neonatal anaesthesia and peri-operative care. Despite this, long-gap OA still poses a number of challenges, and oesophageal replacement still may be required in some cases.[19]

Post-operative ventilation, if required, should be weaned as soon as possible. Adequate analgesia should be provided using neonatal pain score. Total parenteral nutrition is usually required as advancement of feeding may take few days. The trans-anastomotic chest tube may be removed within 48-72-hours if no leak is identified on the X-ray and the output of the chest drain is minimal. It has been customary to start the baby or continue antireflux medications as gastro oesophageal reflux is universal following the anastomosis.

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Nursing Pearls

• Antenatal diagnosis of polyhydramnios should be communicated to the neonatal team.
• Wide pore gastric tube should be inserted in all cases of antenatal suspicion of congenital oesophageal atresia. This includes cases of polyhydramnios.
• The combination of respiratory distress with recurrent excessive oral secretions or choking is highly suggestive of oesophageal atresia.
• Continuous or frequent intermittent suction (every 10-15-minutes) of the upper oesophageal pouch should be applied in all suspected or confirmed cases.
• A Replogle tube should be considered for continuous suction if the surgery is likely to be delayed more than 48-72-hours after presentation of if the secretions are very copious.

CONCLUSION

The diagnosis of COA should be suspected in the antenatal period if there is unexplained polyhydramnios and or if the stomach bubble is small or absent. Excessive frothy oral secretions, in the newly born baby, with or without accompanying respiratory distress are the typical post-natal presenting symptoms. The main modality of treatment is surgical correction which consists of end-to-end anastomosis of the proximal and distal oesophageal pouches and separation of any coexisting TOF and anastomosis of the oesophageal segment postnatally. Thorough pre-operative assessment is required as associated anomalies occurs in 30-60%. The long-term complications are common in surviving cases.

CONSENT

Signed consent is available.

CONFLICTS OF INTEREST

The authors declare that they have no conflicts of interest.

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Original Research

Knowledge, Attitudes and Practices of Parents Regarding Convulsion in Children Under Five Years in Muea Community, Cameroon

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ABSTRACT

Background
Convulsion is an event that can emotionally traumatise most parents. Inadequate knowledge regarding convulsion can cause parental anxiety.

Aim
This research sought to investigate the knowledge, attitudes, concerns and practices of parents regarding convulsion in children under five-years.

Methods
The study employed a community based cross-sectional survey design. Purposive, convenient and snowball samplings were used to select the study site and enroll participants to the study. The study was conducted in Muea Community, Buea Health District in Fako Division, South West Region of Cameroon. The study participants were made up of parents of children under 5-years of age and who had witnessed convulsion in a child. Respondents who met the inclusion criteria and gave their consent to participate in the study were selected. Data was collected using a semi-structured questionnaire made up of both open and closed-ended questions. Data was collected on the knowledge, attitudes, concerns and practices of parents regarding convulsions in children. Data collected was entered using a pre-designed EpiData version 3.1 and data from open-ended questions were analysed using systematic process of thematic analysis.

Results
A total of 100 respondents participated in the study. The study revealed that more than half of the respondents 53.7% had good knowledge on convulsion, 61.9% of the respondents had positive attitudes towards convulsion and 51.4% of parents had good practices regarding convulsion. This study also revealed that knowledge of convulsion had an association (p=0.05) with gender and marital status but was not dependent (p>0.05) on age and level of school attained.

Conclusion
The study concluded that even though more than half of the respondents were knowledgeable on convulsion, there is still need for proper parental education as inappropriate attitudes and practices like putting the child's head in the toilet pit, which can lead to complications are still being practiced.

Keywords
Knowledge; Convulsion; Attitudes; Concerns practices; Children under five; Parents.

INTRODUCTION

Febrile convulsion (FC) also known as febrile seizure (FS) or simply convulsion is the most frequently occurring type of convulsion which causes most of hospital admissions in children under five years of age.[1] Convulsions are usually due to high body temperature and affects 4-10% of children under 5-years of age.[2] To parents it is an extremely frightening, shocking and life-threatening scenario which is traumatizing emotionally and anxiety-provoking.[3] These feelings could be due to the fact that parents have

poor knowledge on convulsion,[4] which could be the reason for the implementation of inadequate first aid measures such as harmful traditional practices.[5] Some parents may lack the knowledge or adequate preparedness to offer first aid to a child who is having a seizure.[5] Parental anxiety and misconceptions about FC contribute to a significant decrease in the quality of life of children and their love ones after a FC.[6] Febrile convulsion usually occurs in children aged three months to five-years, commonly associated with fever but without evidence of intracranial infection or defined cause for the seizure. In developing countries the prevalence varied between 1.33% and 11.61%, whereas the reported prevalence rates from developed countries was between 2% and 5%.[7] This variation in prevalence may be due to differences in case definitions, ascertainment methods, geographical variation and cultural factors. In Cameroon a study conducted in Yaounde revealed that the proportion of children admitted with FC was 6.1% with the mean age being 24.6-months.[8] In Volta Regional hospital Ghana, Statistics show that FC in children under five years accounted about forty to 55% of admissions in 2013.[9]

Generally, convulsions occur at home and as such, parents are the first involved in their management. Hence, knowledge on FC is important for parents, particularly knowledge regarding when it will occur, manifestations, how to manage the seizure, and how seizure can be prevented.[10] In addition, correct and adequate knowledge of the association between fever and FC, and its usual good prognosis are important in lessening parental anxiety and apprehension associated with FC. Of significant importance is the knowledge on First Aid measures to be implemented when FC occurs at home. Comprehending parental knowledge, attitudes and practices regarding FC is vital in planning and delivering health education to parents during infant welfare clinics (IWC) or child well visits.[5]

It is important to note that parental knowledge on proper home management of fever and FC can prevent about 65% of pediatrics emergencies occurring in health facilities due to FC.[11] Studies have revealed that parents in less developed countries lack knowledge on convulsion thus, are frightened when faced with the dramatic manifestations of convulsion and hence, perform inappropriate first aid measures.[12] Meanwhile convulsion would have resolved spontaneously with minimal morbidity and mortality as obtained in technologically advanced countries. It is worth noting that various emergency home therapies employed in developing countries lead to poor outcomes.[13] Fatunde et al[14] identified some of the harmful effects of traditional practices on the child such as prolonged hospitalisation, aspiration pneumonitis and burns among others.[14] Many studies have been conducted on the etiology, manifestations and management approaches of convulsions, but very little information is available about parental knowledge, attitudes and practices.[15] This informed the objectives of this study; specifically, we aimed at investigating the knowledge of parents on convulsion, their attitudes, concerns and the measures they take when faced with an episode of convulsion in the child. This study will provide a framework which will serve as a guide for health care providers especially nurses to plan and deliver appropriate health education to parents.[5] This will go a long way to ensure that required health information regarding FC is given to parents. This will in turn reduce harmful traditional practices, and hence, curb complications associated with febrile seizures in less developed countries.

MATERIALS AND METHODS

A community-based cross-sectional study was conducted from the 28th of October, 2017 to 30th of June, 2018 to investigate the knowledge of parents on convulsion, their attitudes and practices regarding convulsion in the child. Both qualitative and quantitative approaches were employed to collect and process data. A questionnaire made-up of both open and closed ended questions was used to collect data. The target population was made up of all parents residing in Muea Community in the Buea Health District, Cameroon having children aged 0-5-years and who had experienced their child convulsing. All parents who have managed convulsion in a child age 0-5-years and gave their consent to participate in the study were included; eligible participants were recruited from the Muea Community, a rural area in Fako Division of the South West Region of Cameroon. Most of the people living in Muea are of diverse tribes and cultures with most of the inhabitants being farmers.

A sample of 100 participants living in Muea community who were parents or caregivers of children age 0-5-years selected by purposive and consecutive convenient sampling participated in the study. Parents of children aged 0-5-years were purposively selected to participate in the study according to their availability. In addition, the snow-ball approach was used whereby after discussion with a parent; he/she directed us to another parent who had managed convulsion in a child under 5-years of age. Data was collected on participants' knowledge, attitudes and practices regarding convulsion in children under the age of 5-years. Participants' knowledge on convulsion was evaluated using ten questions each given a point, making a total of ten points. A score of 0-4 on 10 (00-40%) was referred to as not knowledgeable (bad knowledge) while a score of 5-10(50-100%) was referred to as knowledgeable (good knowledge). Similarly, attitudes and practices were scored on a scale of 9 and 14 respectively. For attitudes, participants with scores of 5 and above on 9 (56-100%) were considered to have positive (good) attitudes while those with scores of 4 and below on 9 (00-44%) had negative (bad) attitudes towards convulsion. Fair practice was rated as 7-9 on 14 (50-64%) and positive (good) practice was given a score of 10 and above on 14 (71-100%) while negative (bad) practice was given 6 and below points on 14 (0-43%).

Before administering, the questionnaire was pre-tested by administering 10 copies to ten parents who were not part of the study population. Their responses confirmed the clarity and validity of the questions. Copies of the questionnaire were then administered to the study participants who completed the various sections of the questionnaire. The investigator read the questions for those who could not read and their responses were written down.

This study was authorised by the Department of Nursing, Faculty of Health Sciences, University of Buea, Cameroon. Administrative authorisation was first obtained from the Regional Delegation of Public Health (No. 477/107) and then from the heads of the various health facilities. Before responding to the questionnaire each respondent gave her consent by signing the consent form.

Data collected was entered into Epi Data Version 3.1 and analysed using statistical package for the social sciences (SPSS) version 21.0. Data was analysed using the quantitative method. Open-ended questions were analysed using the systematic process of thematic analysis where ideas or viewpoints were grouped under umbrella terms or key words. Chi-Square ($\chi 2$) test of equality of proportion was used to compare proportions for significant difference as well as to measure the association between the study indicators and background information. Data was presented using frequency table and charts. All statistics was discussed at the 95% confidence level (CL), Alpha (α)=0.05.[16]

Using frequency tables and charts, the percentages of responses were determined.

RESULTS

All 100 respondents enrolled, participated in the study giving a response rate of 100%. The mean age of the participants was 29.2 and median 29.0, implying that half of the respondents were aged less than 29-years. Female respondents were the most represented 93(93.0%) and most respondents 69 (69%) were married. Fifty-seven percent (57%) of the participants had attained secondary level of education while 1(1%) of them had never been to school. Only 18 (18%) of the respondents were employed while most respondents 81(81%) were Christians (Table 1).

Table 1. *Demographic Characteristics of Participants*

Characteristic		No (%)
Age	>29 years	50(50)
	<29 years	50(50)
Gender	Female	93(93)
	Male	7(7.0)
Marital Status	Married	69(69)
	Single	31(31)
Level of education	Primary	31(31)
	Secondary	57(57)
	Tertiary	11(11)
	No Schooling	1(1)
Occupation	Employed	18(18)
	Not Employed	82(82) 20(27.0)
Religion	Christianity	81(81)
	Others	19(19)

In aggregate, slightly more than half of the respondents were knowledgeable about convulsion with weight of 53.7%. Most respondents 74 (74%) indicated that fever is the cause of convulsion while others 26 (26%) believed it was caused by malaria. Majority of the participants 70 (70%) said shivering is a manifestation of convulsion in the child. Eighty-one subjects (81%), perceived that traditional herbs were needed for the management of convulsion, 76 (76%) perceived that it was a life-threatening event and up to 34 (34%) indicated that convulsion can cause brain damage. Also 80% of the respondents added that there is stiffening of the child's extremities and eyes roll backwards (Table 2).

The study revealed that knowledge of convulsion had an association (p=0.05) with gender and marital status but was not dependent (p>0.05) on age and level of school attained. It was

Table 2. *Participants' Knowledge on Convulsion*

Items		Responses		
		Yes	No	Don't Know
Causes	Fever is the cause of convulsion	74.0% (74)	26.0% (26)	0.0% (0)
	Malaria is the cause of convulsion	26.0% (26)	74.0% (74)	0.0 % (0)
Perceptions	Convulsion is epilepsy	11.0% (11)	88.0% (88)	1.0% (1)
	Convulsion is a life threatening event	76.0%(76)	21.0%(21)	3.0%(3)
	Convulsion can cause brain damage	34.0%(34)	53.0%(53)	13.0%(13)
	Traditional herbs are needed for the management of convulsion	81.0%(81)	14.0%(14)	5.0%(5)
	Convulsion is rare after age five	26.0%(26)	17.0%(17)	56.0%(56)
	Children with convulsions can receive immun-ization on schedule	11.0%(11)	13.0%(13)	75.0%(75)
	Every child with convulsion will have another convulsion attack	39.0%(39)	20.0%(20)	40.0%(40)
Manifestation	Fainting spell	25.0%(25)	75.0%(75)	0.0%(0)
	Shaking	70.0%(70)	30.0%(3)	0.0%(0)
	Suffocation	1.0%%(1)	99.0%(99)	0.0%(0)
MRS		Correct	Wrong	Non-response
		53.7%(644)	30%(360)	16.3%(196)

statistically obvious that female were more knowledgeable than the male with proportions of 71 (76.3%) and 3 (42.9%) respectively (Table 3). The married were also more knowledgeable 56 (81.2%) than those who were single 18 (58.1%) (Table 4).

Table 3. *Association between Causes of Convulsion and Gender*

Gender	Stats	What Causes Convulsion		Total
		Fever	Malaria	
Male	n	3	4	7
	%	42.9%	57.1%	100.0%
Female	n	71	22	93
	%	76.3%	23.7%	100.0%
Total	n	74	26	100
	%	74.0%	26.0%	100.0%
χ^2-test: χ^2=3.794; df=1; p=0.049				

Table 4. *Association between Causes of Convulsion and Marital Status*

Age	Stats	What Causes Convulsion		Total
		Fever	Malaria	
Married	n	56	13	69
	%	81.2%	18.8%	100.0%
Single	n	18	13	31
	%	58.1%	41.9%	100.0%
Total	n	74	26	100
	%	74.0%	26.0%	100.0%
χ^2-test: χ^2=5.930; df=1; p=0.015				

In aggregate, more than half 61.9% of the respondents had positive attitudes towards convulsion. For participants with positive attitudes towards convulsion, majority 95 (95%) attested to the fact that children with convulsion required frequent temperature monitoring and also that the child needs more attention and care 93 (93%). Among those with negative attitudes 87 (87%) believed that folk medicine was needed during the convulsive attack, while some 13 (13%) believed it's a shameful thing to have a child with convulsion, 30.0% believed it's due to possession of evil spirit. Other attitudes reported were panic and fear (95%) and run away from the child (2%) (Table 5).

In aggregate, 51.4% of parents had good practices towards convulsion. Majority agreed to lower the child's body temperature 97 (97%), give Paracetamol syrup 92 (92%) and 99 (99%) said they will rush the child to the hospital. However, some respondents 92 (92%) reported opening child's clenched teeth to put something in the mouth, others 67 (67%) reported restraining the convulsing child. Sixty-six (66%) of the respondents did not place the child on a safe surface and 68 (68%) did not place the child on his or her side. Majority 73 (73%) did not keep calm during the event. Other interventions that the parents reported were putting the child's head into the toilet pit 40 (40%), applying black "Mayanga and masepo" on the child's body 32 (32%), bleeding the child on the forehead 2 (2%) amongst others (Table 6).

Fear of death was the major concern for most participants 94 (94%), followed by recurrence 92 (92%), fear of future epilepsy 61 (61%), mental retardation 37 (37%) and physical disability 32 (32%).

DISCUSSION

This study aimed at investigating the knowledge, attitudes and practices of parents regarding the home manage of convulsion in children under five-years of age. Worthy of note is the fact that proper planning and delivery of appropriate health education to parents by nurses will create awareness in parents and upgrade their knowledge. This will in turn reduce harmful traditional practices, and hence, curb the morbidity and mortality associated with febrile seizures in less developed countries.

According to the findings of this study, female respondents were the most represented. This is in line with the study of Abeysekara et al[15] who had a larger proportion of female respondents. This could be explained by the fact that females are more

Table 5. *Participants' Attitudes towards Convulsion in Children*

Items	Responses		
	Yes	No	Don't Know
Convulsion is due to possession by evil spirits	74.0% (74)	26.0% (26)	0.0% (0)
Convulsion will become epilepsy	26.0% (26)	74.0% (74)	0.0 % (0)
Convulsion is contagious	11.0% (11)	88.0% (88)	1.0% (1)
Parents should take their child's temperature fre-quently	76.0%(76)	21.0%(21)	3.0%(3)
"Country medicine" is also necessary during a convulsive attack	34.0%(34)	53.0%(53)	13.0%(13)
More attention and care are needed for a child with convulsion	81.0%(81)	14.0%(14)	5.0%(5)
It is shameful to have a child with convulsion	26.0%(26)	17.0%(17)	56.0%(56)
Parents of children with convulsion should be avoided	11.0%(11)	13.0%(13)	75.0%(75)
MRS	Positive	Negative	Undecided
	495	203	102
N=100; $N_{responses}$=800	61.9%	25.4%	12.7%

Table 6. *Participants' Attitudes towards Convulsion in Children*

Items	Responses		
	Yes	No	Don't Know
Lower the child's body temperature	97.0%(97)	1.0%(1)	2.0%(2)
Using cold water	66.0%(66)	44%(44)	0.0 %(0)
Using luke warm water	34.0%(34)	66.0%(66)	0%(0)
Give paracetamol syrup	92.0%(92)	7.0%(7)	1.0%(1)
Protect the child on a soft and safe surface	21.0%(21)	66.0%(66)	13.0%(13)
Place the child on his or her side	16.0%(16)	68.0%(68)	13.0%(13)
Keep calm	27.0%(27)	73.0%(73)	0%(0)
Observe seizure manifestation and duration	15.0%(15)	80.0%(80)	5.0%(5)
Rush the child to the hospital	99.0%(98)	1.0%(1)	0%(0)
Call for help	99.0%(99)	1.0%(1)	0%(0)
Shake and rouse the convulsing child	34.0%(34)	62.0%(62)	4.0%(4)
Pry (open) the convulsing child's clenched teeth apart and put something in his/her mouth	92.0%(92)	6.0%(6)	2.0%(2)
Attempt to do mouth to mouth resuscitation	13.0%(13)	77.0%(77)	10.0%(10)
Suck discharge from the child's nose and mouth	14.0%(14)	77.0%(77)	9.0%(9)
Restrain the convulsing child	67.0%(67)	25.0%(25)	8.0%(8)
I don't know what to do	1.0%(4)	99.0%(96)	0%(0)
MRS	Good	Bad	Don't know
	51.4%(823)	44.4%(710)	4.2(67)

involved in child care. Again, the results showed that females were more knowledgeable than the men. This could be probably due to the fact that females are more engaged in child care and as such, more experienced in matters concerning children's health. Also, it was found in this study that, the highest level of school attained by majority of the respondents was secondary education. This finding is in line with that conducted by Bogne et al[12] whereby 57% of the study participants were found to have attended secondary education. Nevertheless, only a fewer proportion was found to have attended primary school as compared to this study. This is probably due to the fact that this study was conducted in a rural area were literacy rate is lower.

Concerning the participants' knowledge on FC, the study showed that most participants were knowledgeable. For instance, majority of the participants indicated fever being the cause of convulsion. This result coincides with those of Abeysekara et al[15] and Agin et al[17] where majority (91.45% and 55.1% respectively) of their respondents attributed fever as the cause of FC. These finding contradicts that of Anjum et al[18] who found out that 58% of the respondents did not know convulsion could be due to fever. Also, the findings of this study showed that parents have good knowledge of the signs and symptoms of the condition such as shaking, stiffening of the extremities and eyes rolling backwards. This supports the findings of similar studies where the majority of the mothers (70-96.5%) gave good description of the illness.[9,19] This could be due to the fact that they are always present at the time of seizure. Furthermore, most parents did not think that convulsion is epilepsy which contradicts that of Agin et al[17] in which a high proportion of their study population perceived febrile convulsion as epilepsy. Our study revealed that most respondents perceived convulsion to be life-threatening. This finding ties with that of Agin et al[17] where majority of their subjects (87.9%) believed convulsion is life-threatening. This could be due to the fact that parents are usually not aware of the benign nature of convulsion.[17] This study also showed that only one third of the subjects thought FC could cause brain damage. This slightly contradicts that conducted by Agin et al[17] where almost all (96%) of respondents felt FC could cause brain injury.[17] However, there is no association between any type of febrile seizure and later development of neurological deficit.[20] Nevertheless, some parents perceived convulsion to occur again. This is in accordance with the findings of Agin et al[17] in which majority of the respondents showed significant concerns of recurrence. However, recurrence rate for FS is low for most children but one-third will; with age being the strongest and most consistent risk factor Hall-Parkinson et al.[5] More than half of the risk is realised during the first year after initial FS and over 90% recur within two years. Risk factors for recurrence are family history of febrile seizures,[21] low temperature at initial FS and being less than 18-months of age.[5,22] Therefore, it should be remembered that an episode of fever is in fact the only time that the child is at risk of recurrence. Overall, this study found that parents had good knowledge on FC, this overall finding is similar to those of Abotsi et al[9] and Anigilaje et al[13] who found that parents have good understanding of FC in children under five. However, this finding contradicts that of Shibeeb et al[23] in Babylon, where less than half of the participants (43%) had good knowledge on FC. In addition, this finding contradicts that of Abeysekara et al[15] who found that more than half (77.9%) of the parents were unaware of the entity of febrile convulsion. This difference in findings may be due to a variety of reasons including increasing parental awareness.

With respect to the attitudes, most respondents agreed to the fact that the child's temperature should be monitored fre-

quently and giving more attention and care to the child. These findings concord with that of Agin et al[17] where, most parents believed that the children with FC required frequent temperature monitoring. This could be explained by the fact that most parents are aware that fever is the immediate cause of the convulsion and checking the child's temperature will help guard against the attack. Also, this study revealed that majority of the respondents said that convulsion is not a contagious condition and consequently parents of the child with convulsion should not be avoided. This finding could be due to the fact majority of the respondents knew that convulsion was due to fever. In addition, the findings revealed a fewer proportion of respondents perceived it was shameful having a convulsing child. However, this proportion was lower than that of Agin et al[17] where close to half of his subjects were ashamed of having a convulsing child. This slightly negative attitude could be explained by the fact that parents are not knowledgeable enough on FC. Moreover, almost half of the participants said that convulsion will become epilepsy which is not true because the risk of developing epilepsy following simple FC or complex FC is 1-2.4% and 4.1-6%, respectively.[17]

Furthermore, majority of the parents reported that folk medicine ("country medicine") was necessary during a convulsive attack. This may be due to the fact that they feel folk medicine has a rapid effect on aborting the convulsion. It is important for parents to be informed that some of these traditional practices may lead to the child's death or long-term neurological deficit.[14] This study also revealed that more than a quarter of the study population still perceived that convulsion could also be due to evil spirit possession. This finding supports that of Abotsi et al[9] in which 30% of the respondents described convulsion as a sickness in children which is normally caused by witchcraft, evil spirits that fly as birds at night. These findings also tie to the findings of Anigilaje et al[13] who also found that participants attributed angry gods, evil spirits (49%), constipation and black blood to the causes of FC.[13] These gross misconceptions about FC by parents, may favor decisions to take inappropriate measures in an attempt to control the convulsion. This study also found that almost all, the participants mentioned the feeling of panic and fear whereas very few confessed that they will run away from the child. Panic and fear may come as a result of the child exhibiting sudden body contraction followed by loss of consciousness. Running away from the child may be due to the frightful manifestation of the illness. Overall, the parents' attitudes towards the convulsing child was positive. This contradicts the findings of Bogne et al[12] who concluded in his study that parents had inappropriate attitudes towards convulsion.

With regards to respondents' practices on convulsion, almost all the subjects in the study knew what to do during the time of the convulsion. This result contradicts those of Agin et al[17] and Anjum et al[18] in which some proportion of respondents (44% and 58.18% respectively) did not know what to do at the time of the attack. The results of this study showed that parents tried lowering the child's body temperature. This finding is in good accord to that of Abotsi et al[9] and Agin et al[17] where most and majority of the participants respectively reported tried reducing the fever in an attempt to prevent convulsion. Also, majority of the participants in our study agreed on giving Paracetamol syrup to the child given that it is an antipyretic. Unfortunately, most parents did not consent on the fact to place the child on soft and safe surface, placing the child on his or her side or even observing the seizure manifestation and duration. These findings reflect that of Anigilaje et al[13] whose respondents did not acknowledge the correct practice of putting the child on his or her. It is well-known that children with convulsion are expected to be laid on their side to prevent injury resulting from falls and dangerous aspiration of secretion that may be regurgitated into their mouths. A majority of the subjects rushed the child to the hospital while a similar majority called for help. These results coincide with that of Anigilaje et al,[13] where 82.8% of the participants rushed the child to the hospital while 81.6% called for help. Nevertheless, the study also revealed that, most parents attested to the fact that they opened the convulsing child's clenched teeth so as to put something inside the mouth. This is a similar finding to the studies carried out by Anigilaje et al[13] and Agin et al.[17] However, the proportion of participants of this study was higher than those of the other study (9% and 61.2%) respectively. Other major traditional practices reported were putting the child's head into the toilet pit, applying "*black mayanga and masopo*" on the child's body, making scarification amongst others. Similar traditional practices were reported by another study.[1] Most parents believed that when a child is convulsing, all efforts must be put in place in order for the child not to bite the tongue, an event that may lead to instant death of the convulsing child. Thus, any item readily available including the parents hands is put into the mouth of the convulsing child. Other items including spoon, spatula or even sticks are quickly placed into the child's mouth to "prevent" death. Furthermore, the concept of making a therapeutic incision on the fore head of the convulsing child was practiced only by a few as they believed that it could be due to evil forces so by scaring the child will help liberate the child from these forces. Some subjects also burned the feet of the child in open fire. This proportion was lower than that reported (8.3%) by Anigilaje et al[13] in Nigeria but similar to that of Fatunde et al[14] still in Nigeria who reported a value of 1.4%. This mode of treatment appears as one of the practices that have been passed down from one generation to another to another, without any logical explanation and which have been abandoned by many explaining why only a few carried out the intervention.

Pertaining to the common concerns of participants, the study showed that the most common concern expressed among subjects was the fear of their child dying. This finding is similar to that of Abeysekara et al[15] in India who also reported that 90% of the parents worried about possible demise of the convulsing child. Also, these findings are in good accordance with that of Anjum et al[18] where 82.72% of subjects thought the child may die due to the convulsion. However, Kumar, et al[24] in Turkey reported a lower proportion of 33.5% parents nursing fear of death of the convulsing child. The abrupt onset of the abnormal motor activities and the accompanying impairment or loss of consciousness is a dreadful and frightening experience and may readily explain the parental concern of imminent death.[24] It should be noted that there are almost no deaths reported in a thousands of cases of FS that have been studied and the risk for mortality is not higher in children with FC.[25] Furthermore, the study revealed that most subjects feared recurrence of convulsion, a finding that was higher

than that of Abeysekara et al[15] and Kumar et al[24] who reported fear of recurrence in 19.3% and 91.4% respectively in the participants. Another significant concern for parents was fear of feature epilepsy which tied to the findings of Agin et al[17] and Anjum et al.[18] Accumulated epidemiological evidence indicates that FC are the most common recognised precursor for epilepsy in childhood, although the exact risk for developing epilepsy after a febrile seizures is uncertain.[21] However, factors that increase the risk for developing epilepsy following FC include; a family history of epilepsy, complex features and the presence of early onset of neurodevelopmental abnormalities.[26]

Talking about the association between knowledge of convulsion and demographic characteristics, the study revealed that knowledge of convulsion was associated (p=0.05) with gender and marital status respectively but was not dependent (p>0.05) on age and level of school attained. In addition, it was statistically obvious that female were more knowledgeable than the male with proportions of 76.3% and 42.9% respectively. Furthermore, the married were also more knowledgeable 81.2% than those who were single 58.1%. The findings of our study however, disagree with the findings of Wuni et al[1] and Shibeed et al.[23] Wuni et al[1] noticed that knowledge was associated with belief that witchcraft could cause FC (p=0.44), first aid (p=0.021) and intervention after first aid (p=0.040) while Shibeed et al[23] found that knowledge was associated with residence (p=0.047) and educational level (p=0.001).

CONCLUSION

This study revealed that overall, parents' knowledge on convulsion was good, and more than half of the respondents had positive attitudes towards convulsion. However, it was found that some parents whose children had suffered from febrile convulsion even though, had adequate knowledge regarding the causes, signs and symptoms of convulsion, still showed negative attitudes such as panic and fear towards convulsion. In addition, the study revealed that generally their practices towards convulsion were good. Nevertheless, there still exist some inappropriate practices like putting the child's head into the toilet pit. Most parents feared that they will lose their child during the convulsive attack. Based on the findings of this study, it can be seen that there is still a need for parents to be properly educated by nurses especially on convulsion and its benign nature. This will go a long to allay their fears and concerns, and improve their first aid measures of convulsion in children under 5-years. This will in turn reduce prolonged hospitalization and aspiration pneumonitis among others resulting from inadequate practices.

LIMITATIONS OF THE STUDY

This study used a questionnaire to collect data; the disadvantage is that it does not produce rich data; in this case participants' feelings cannot be fully captured. To compensate for the limitation open-ended questions were included and copies of the questionnaire were administered on a face-to-face basis. The data from the close and open ended questions together gave us a broad understanding of the participants' knowledge, attitudes and practices.

In addition, the study was only conducted in Muea Commuinity and as such the findings cannot be generalised to other areas. Hence, it is suggested that a more robust studies on parents' knowledge, attitudes and practices be carried out to increase the validity of our study.

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AUTHORS' CONTRIBUTION

Both authors participated in all steps of the study from its commencement to writing. That is, conception and design, acquisition of data, analysis and interpretation of data as well as drafting and or revising and approving the final manuscript.

CONFLICTS OF INTEREST

The authors declare that they have no conflicts of interest.

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