Paediatric video-assisted thoracoscopic clipping of patent ductus arteriosus: experience in more than 700 cases

Emmanuel Villa*,1, Frédéric Vanden Eynden, Emmanuel Le Bret, Thierry Folliguet, François Laborde

Département de Pathologie Cardiaque, L’Institut Mutualiste Montsouris, 42 Boulevard Jourdan, 75014 Paris, France

Received 1 September 2003; received in revised form 9 December 2003; accepted 15 December 2003

Abstract

Objective: To overcome drawbacks of thoracotomy and percutaneous techniques, we have performed video-assisted thoracoscopic surgery (VATS) since 1991 to treat patent ductus arteriosus (PDA). This retrospective study aims to analyse morbidity and outcome in order to add data in the evaluation of minimally invasive operations. Methods: From September 1991 to March 2003, 703 patients underwent PDA clipping by VATS. The indications for operation were clinically significant patency or failure to close in older children. Diameter > 8 mm, previous thoracotomy, calcifications, infection, or aneurysm were contraindications to VATS. In right decubitus, only two thoracostomy incisions (for 5-mm trocars) and a smaller one (for direct hook insertion) were required. In the post-operative stay two echocardiographic controls were scheduled, the first before extubation and the second before discharge. Referring cardiologists assured follow-up. Results: Mean age was 3.0 ± 3.8 years (5 days–33 years), mean weight 10.7 ± 8.0 kg (1.2–65 kg), and 3.1% of our activity were low birth-weight infants (LBWIs) weighting ≤ 2.5 kg. Operative and 30-day mortality was nil. Median operative time was 20 min and median stay 2 days. Unfavourable events globally occurred in 6.8% of cases (13.6% of the LBWIs, RR 4.0, CL 95% 1.5–10.4). Recurrent laryngeal nerve injury was noted in 3% (13.6% of the LBWI, RR 5.1, CL 95% 1.6–15), but only 0.4% had long lasting dysfunction. Incidence of chylothorax was 0.6%, thoracotomy 1%, transfusion for bleeding 0.1%, pneumothorax 1.3%. LBWIs were at increased risk for the latter two events. Residual patency was detected immediately in 1.4% (all non-LBWI) and underwent additional surgery. Incidence of residual patency at follow-up was 0.6% (0% LBWI, 0.6% in 2.5–25 kg group, 5.0% in > 25 kg group, P = 0.001). Conclusions: This study records a long experience of PDA treatment in a wide range of body size and age. VATS clipping is safe, but LBWIs are at augmented risk of complication. It may be carried out with a high degree of efficacy in all the ductus diameters < 9 mm. VATS clipping requires minimal operating time and avoids morbidity related to chest wall trauma, percutaneous vascular access, and intravascular foreign bodies.

© 2004 Elsevier B.V. All rights reserved.

Keywords: Patent ductus arteriosus; Video-assisted thoracic surgery; Minimally invasive surgical procedures; Paediatrics; Low birth-weight infant

1. Introduction

Patent ductus arteriosus (PDA) was the first congenital heart lesion to undergo surgical intervention [1] and also the first where a transcatheter approach was utilised [2]. Consequently, PDA treatment may be considered a benchmark for new modalities of heart disease therapy.

In published series [3], and in the unreported experience of congenital surgical teams, the interruption of PDA via open thoracotomy is a safe surgical procedure that provides reliable results. It represents the standard against which all other surgical or interventional therapeutic approaches are compared.

Closure in the catheterisation laboratory has been increasingly used, but some restrictions on body weight and ductus morphology limit the indication. Furthermore, interventional techniques have shown a variable rate of residual patency and continuous adoption of different devices prevents having long-term data [2,4]. On the surgical side, advances in technology have produced instruments and equipment that now allow the so called
minimally invasive surgery. In the early 1990s, video-assisted thoracoscopic surgery (VATS) began to be employed also in the paediatric field [5,6]. Less invasive operations have gained popularity but have not yet achieved widespread acceptance and the number of applications in paediatrics remains limited. Therefore, additional data regarding the outcome of PDA closure with VATS are necessary in order to compare the results with those of conventional treatment and those of rapidly evolving non-surgical approaches.

VATS clipping for PDA has been performed at our institution since 1991. Techniques and patient care underwent only slight modifications over the years and the procedure has become highly standardized. The present paper summarizes and analyses overall results.

2. Materials and methods

2.1. Patients

Indication to close a PDA in low birth-weight neonates/ premature infants was the presence of a clinically or instrumentally significant shunt. In older children, the indication was persistent patency. Paediatric cardiologists of referring hospitals determined whether a surgical or non-surgical procedure was performed on each patient. All children presented for surgical treatment were evaluated for inclusion in the VATS group. Ductus diameter > 8 mm, previous thoracotomy, presence of calcifications, active infection, or aneurysm were considered contraindications. From September 1991 to March 2003, a total of 754 patients were referred to our institution for isolated ductus arteriosus surgical treatment. PDA closure with VATS clipping was performed in 703 of these cases, and the outcomes were retrospectively analysed. Of the remaining patients, 28 were enrolled in a study previously conducted by our department [7], and the final 23 were considered for standard thoracotomy approach due to contraindications to the VATS technique.

2.2. Pre-operative protocol

Standard pre-surgical laboratory analysis and chest X-ray were performed primarily on an outpatient basis. Trans-thoracic echocardiography, however, was systematically repeated on the admission day for complete cardiac re-evaluation and determination of ductus size. Angiography and cardiac catheterisation were not routinely employed if accurate anatomical and functional definition was obtained with ultrasounds. The degree of pulmonary hypertension not allowing PDA closure by VATS referred to the standard limits adopted in the other procedures [2].

The protocols were evaluated by the local institutional committee and informed approval from patients or children’s parents was obtained.

2.3. Surgery

Technique and equipment used have been described in previous works, and have remained unchanged for the 12-year period with a few exceptions (modality of dissection, chest tube drainage) [5,8]. The principal steps included two thoracotomy incisions for the introduction of 5-mm trocars (L-shaped electrosurgery and camera), one smaller incision for direct nerve hook insertion (two hooks for lung retraction, one for pleural reflection), positioning of two 9-mm titanium clips through the central incision after trocar removal, and pleural drainage by one small-diameter wound drain catheter (type Redon-Jost) placed through the same incision.

2.4. Anaesthesia

Anaesthesia management was recently modified to include a new drug scheme (propofol, sufentanil, atracurium besylate for induction; sevoflurane and sufentanil for the maintenance). However pressure monitoring, perfusion lines, and ventilation strategy were employed as previously described [5,8].

2.5. Post-operative protocol and complication recording

Colour flow Doppler echocardiography was performed immediately after surgery, either in the operating room, in the recovery room, or in the intensive care unit depending on the clinical status and age of the patients. If a persistent shunt was detected, the patient was returned to the operating theatre. In case of complete flow suppression, chest X-ray and extubation were performed. Another ultrasound evaluation was conducted prior to discharge.

All morbid events were recorded with particular attention given to the following unfavourable occurrences: residual ductus patency, VATS redo clipping, thoracotomy, transfusion for bleeding, clinically detected transient laryngeal nerve dysfunction (hoarseness, stridor, voice changes), persistent laryngeal nerve dysfunction (ORL confirmed), phrenic nerve injury, pneumothorax (if requiring placement of a new drain), chylothorax, wound infection, aneurysm formation, pulmonary artery stenosis, inadvertent clipping of another vessel, endarteritis/endocarditis, or haemolysis. A complicated course was defined if any of the cited morbid events occurred at any moment after the operation.

Intensive care unit plus surgical paediatric ward stay at our department were considered post-operative stay. Continuation in neonatal nursery due to the premature condition (lung immaturity, weight deficit, etc.) was not considered as requiring further stay due to surgical morbidity. Referring paediatric cardiologists assured follow-up visit on 7–10th post-operative day and after 6 months. Antibiotic prophylaxis of infective endocarditis was prescribed only in the presence of residual shunt.
2.6. Cost analysis

During the last years, a complex monitoring system of hospital costs had been available. The cost of hospitalisation at our department (direct charges, meals, laundry, sterilization, general logistic, medical logistic) and the cost of the facility were retrospectively determined and a daily mean charge was obtained. Instead, technical consumption was prospectively recorded using the scoring system of the French National Health Organization (www.le-pmsi.org). Every performed medical act was scored considering consumption of medical, nursing, and technical resources.

2.7. Statistical analysis

Data were analysed using a statistical software program (SPSS for Windows 11.5, SPSS Inc., Chicago, IL, USA). Means are expressed ± one standard deviation; range was added if considered helpful to the interpretation of data.

Patients were divided into groups by body weight according to cut-off reported in the literature[9,10]. Pearson χ² test, or Fisher’s exact test when appropriate, was used to evaluate differences in the incidence of complications. Statistical significance between data was considered achieved when P was < 0.05 except for degrees of freedom >1 when appropriate smaller values were calculated and signalised. Relative risks were calculated and presented with 95% confidence limits.

3. Results

Mean age at surgery was 3.0 ± 3.8 years ranging from 5 days to 33 years (Fig. 1). The overall mean weight was 10.7 ± 8.0 kg (range 1.2–65 kg). A particular group of patients was identified: 22 low birth-weight infants (LBWIs), weighing <2.5 kg (mean 2.1 ± 0.4 kg), constituted 3.1% of our VATS activity (Fig. 2). The male/female ratio was 0.6 and 0.8 in the LBWI group.

Associated cardiac anomalies were present in 4.1% of patients (29/703) but all defects were of mild severity and none requiring conventional treatment by sternotomy or open thoracotomy when indication for PDA closure was done. Conversely, 0.6% (4/703) of the patients had previous cardiac surgery (two VSD, one ASD, one APVR) with a residual ductal shunt detected in the post-operative course. Among other pathologic findings, Down’s syndrome comprised 4.0% (28/703) with none in the LBWIs group.

Operative and 30-day mortality was nil. Median operative time was 20 min (incision to skin glue application). Post-operative stay ranged from 1 to 20 days and the median was 2 days. Direct home discharge was performed only in 77.2% of patients (543/703), reflecting a preference to transfer, on the first post-operative day, to secondary structures for ‘protected’ convalescence in cases where the home was a long distance away from our centre.

Unfavourable events occurred in 6.8% of patients (48/703) while incidence of complicated course in LBWI was 22.7% (5/22). Suboptimal outcomes were recorded in 17.9% of the Down’s syndrome group (5/28) (P = 0.036; RR 2.0, CL 1.2–7.5).

Residual patency of the ductus was detected immediately in 1.4% of cases (10/703) and none in LBWI group. In eight of these patients, a second VATS clipping was conducted while the remaining two were operated on by a limited thoracotomy. The second procedure was successful in both patients of the thoracotomy group but a persistent leak remained in two of the eight using the VATS strategy (25%, 2/8). These two known cases and another five tiny residual shunts that were not detected immediately after the first operation were documented at the echocardiography check before discharge (1.0%, 7/703). The exiguity of all the shunts allowed us to tolerate them and only control visits were prescribed. At 6-month visit, all the PDAs tolerated at discharge showed no leak with the exception of one that underwent ligature and clipping by minithoracotomy. Three other patients had newly discovered shunts at follow-up (two of these underwent closure with thoracotomic entry and one did not undergo further procedure), for a final figure of 0.6% (4/703) for overall late residual patency.

Chylotorax was managed conservatively with the exception of one patient that required thoracoscopic treatment. In this group, mean post-operative stay was 10.0 ± 6.7 days (range 6–20).

Table 1 depicts the incidence of complications other than residual patency.
Table 2 reports suboptimal outcomes, analysed according to body weight repartition. Low birth-weight was associated with an increased risk of post-operative laryngeal nerve dysfunction but long lasting consequences were not as likely to occur compared with the non-LBWI group. Furthermore, once the problem occurred, the LBWIs were not at significantly increased risk to develop persisting symptoms ($P = 0.4$, RR 3.0, CL 0.4–23.7). Low weight was significantly associated with an increased risk of pneumothorax, transfusion, and complicated course.

Table 3 Subanalysis of residual patency and thoracotomy

<table>
<thead>
<tr>
<th>LBWIs, %</th>
<th>2.5–25 kg, %</th>
<th>&gt;25 kg, %</th>
<th>$P^*$</th>
</tr>
</thead>
<tbody>
<tr>
<td>Immediate residual patency</td>
<td>0</td>
<td>1.4 (9)</td>
<td>2.5 (1)</td>
</tr>
<tr>
<td>Residual patency at discharge</td>
<td>0</td>
<td>0.8 (5)</td>
<td>5.0 (2)</td>
</tr>
<tr>
<td>Residual patency at follow-up</td>
<td>0</td>
<td>0.3 (2)</td>
<td>5.0 (2)</td>
</tr>
<tr>
<td>Thoracotomy</td>
<td>4.5 (1)</td>
<td>0.6 (4)</td>
<td>5.0 (2)</td>
</tr>
</tbody>
</table>

Weight as risk factor for residual patency and for thoracotomy, was still further evaluated in Table 3 (because the literature did not offer pertinent weight subdivisions, the non-LBWI group was split at the 95th percentile of the weight scale).

Finally, Table 4 reports the mean cost of a VATS clipping operation at our institution in the last 85 patients (analysis was limited to the last 18-month period due to administrative reasons and not to case selection).

4. Discussion

PDA is reported to be the third most common congenital cardiac lesion. Closure of an isolated PDA diagnosed in infancy removes the haemodynamic burden of the shunt that may be responsible at times for severe congestive heart failure. In older children/adults suppression of the shunt is advised to prevent infective endarteritis, pulmonary hypertension, heart failure, aneurysm formation, pulmonary/systemic thromboembolism and to avoid the associated shortened life expectancy [2,4]. PDA in premature ones presents some particularities [11] and surgery for a symptomatic ductus is generally indicated in case of failure or contraindications to the pharmacological attempt [2].
After more than 60 years of practice worldwide, conventional surgical approach has reached a high level of safety and efficacy and it has been credited as the gold-standard of treatment [3]. PDA may be simply ligated/clipped or it may be sectioned and sutured. The feasibility of conducting the procedure at any age and the versatility offered to correct varied anatomical occurrence make thoracotomy approach largely adaptable, but it has resulted in some concerns about long-term sequelae (ribs deformity, scoliosis, shoulder dysfunction) [12] besides post-operative pain, pulmonary impairment, and duration of hospital stay [13]. Today cosmetics and length of incision are questionably more and more important but it is a fact that muscle cutting and rib spreading are invasive components of the standard surgical procedure.

To reduce the morbidity of open thoracotomy, we have developed the procedure of VATS clipping and the series analysed in this manuscript is the largest reported to date. Instruments are introduced through only two trocars and, with a supplemental incision of a few millimetres, exposition, dissection, clipping of the ductus can be easily carried out. Damage to the chest wall is substantially reduced by the limited numbers of thoracostomies and by a median operative time of only 20 min. In the absence of wide pleural opening, temperature management, particularly relevant to the younger patients, is easier with VATS than with thoracotomy. No wound infections were registered and the use of skin glue has shown to be very beneficial.

Therapeutic results are also required to validate new minimally invasive operations: a demonstration of at least a comparable level of efficacy and morbidity to standard technique are awaited from VATS.

Recurrent laryngeal nerve injury was noted as a complication in 3.0% of our patients. The incidence with standard PDA ligation is reported to be 4.2% by Fan et al. [14] while other authors indicate 2.5% using VATS clipping [15]. Symptoms attributable to vocal cord paralysis regressed in most of our cases and only 0.4% had long lasting dysfunction. Clip entrapment of the nerve has been cited to be the mechanism of injury [16], but we believe that trauma induced by traction (or thermal injury by electrocautery) may better explain the observed clinical outcome. The complication seems to be, at least functionally, a reversible disorder. The incidence of laryngeal nerve dysfunction in LBWIs was significantly higher (13.6%, \( P = 0.025 \)) and they have an increased risk (5.1 times) compared with non-LBWIs (CL 1.6–15.0). Zbar et al. [17] reports a series of PDAs treated using open thoracotomy and indicates an incidence of recurrent laryngeal nerve injury of 22.7% in extremely low-weight babies, confirming the importance of the issue in premature infants. Decreased incidence of this complication appears to have been achieved with VATS and may be a consequence of improved vision from the video camera image.

The incidence of pneumothorax requiring de novo drain placement was 0.8%. We believe that routine adoption of a small-bored drain as chest tube and a Redon wound drainage system as conventional drainage modality may be safely utilized. Moreover we preferred to always drain the pleural cavity unlike other practices [18,19].

Conversion to thoracotomy for haemostatic control was necessary in very few cases (two patients). Overall, thoracotomy was required in 1% of patients: in addition to the two previously mentioned, it was performed in five for difficult closure of the ductus (two immediate and three residual shunt). LBWIs were at increased risk (RR 4.1, CL 0.7–30.5), however this increase was not statistically significant. Other authors report a number of unplanned thoracotomies ranging from 1 to 7% in mixed patient populations (neonates, children, adults) [18,19]. In our opinion, a careful echocardiographic evaluation of the size and anatomy of the ductus is essential to adequately select patients for VATS. In this way it is possible to minimize the incidence of conversion and address PDA of larger size directly to thoracotomic closure.

Selecting the best treatment in greatly premature babies is a challenging task. We do not have inferior weight limit or minimal gestational age for VATS, but our patients’ recruitment is limited by the absence of a neonatal intensive care unit in our hospital. Therefore, our VATS experience in great prematurity is still limited and we look forward to improving our knowledge in this field.

Echo Doppler studies have detected flow in clinically silent ducts that have been only ligated [20]. Consequently, because we were not dividing the ductus, we routinely performed a rigorous echographic evaluation immediately after surgery and before discharge. This protocol has allowed us to detect 1.4% of incomplete closures. Each of these patients underwent additional surgery and the second procedure was unsuccessful in one-fourth of patients re-operated using VATS. Therefore, it seems wise to remedy with an open surgery in the event that the first VATS operation fails. This would allow shortcomings in clip size, anatomic anomalies and difficult visualization to be more easily overcome. The majority of patients with immediate residual flow belonged to non-LBWI group. However, despite no significant difference between weight groups, the heavier patients require greater care when disposing of a single size clip because of thicker vessel walls. Indeed, residual patency at follow-up (total incidence 0.6%) was significantly associated with body weight over 25 kg (\( P = 0.001 \)) as was the overall incidence of thoracotomy (\( P = 0.006 \)).

Only after adoption of coil use and accumulation of sufficient manipulation experience, transcatheter occlusion systems results have been comparable to surgery. Intermediate (1 year) residual shunt was present in 5% of patients enrolled in the European Registry [21]. However, immediate residual patency was 41% and it is unknown in how much time complete occlusion occurs. Consequently concerns about repeated examinations, duration of follow-up, and necessity of long-term antibiotic prophylaxis
exist [22]. Superseding the Rashkind double umbrella PDA occluder, the Amplatzer occluder is an alternative to coils in larger ducts. All of these devices, however, maintain disadvantages and potential intra-procedural risks: embolization of the occluder to systemic or to pulmonary circulation and peripheral vascular injury. Intravascular foreign corps may protrude creating iatrogenic pulmonary or isthmic stenosis or they may cause thromboembolic events, haemolysis, infection, aneurysm, or re-canalization [2,4,22]. Cases of vocal cord paralysis have also recently been attributed to closure by coils [23].

The VATS technique is safely applicable to cohorts of patients younger and lighter than those generally treated in cardiological series [21,24]. We successfully carried out clip closure in a wide range of body size and age with a simple set of instruments. Further, in our study a high degree of efficacy was demonstrated for ductus diameters up to 8 mm. Multiple coils, conversely, are usually required for ductus diameters ranging from 3 to 8 mm, increasing the risk of complications.

Sedation and local anaesthesia generally allow very short hospital stays in interventional practice. We deem home discharge the morning of the second day advisable to assure a safe early post-operative course and we consider this practice to have an acceptable cost. Although we did not systematically survey pain or cosmetic evaluation of scars from this operation, children and adults often have reported satisfaction of the smooth and rapid course of repair and recovery.

No extensive cost/effectiveness comparisons of VATS clipping versus open surgery or versus currently adopted transcatheter techniques are available in the literature. Prieto et al. [24] stated that coil occlusion is as effective and less costly than conventional surgery. Their conclusions are principally based on the cost of post-surgical stay while considering the presence of persistent leaks clinically negligible. Regarding these issues, we estimate that thoracoscopic approach may favourably influence the cost-effective therapeutic balance thanks to shorter inpatient hospital stay (compared to conventional surgery) and less residual shunts (compared with transcatheter procedure).

Our study presents many intrinsic limitations primarily due to its retrospective nature and because it involved the analysis of a single institution only. Furthermore, conclusions from cost comparisons between different systems and institutions are always something hazardous. Nonetheless, this study records a long experience of continuous practice ranging from neonates to adults. We wish to provide additional data for evaluation of VATS in congenital practice. Shortcomings of this procedure may be overcome by adopting current endoscopic techniques of general thoracic surgery or other available techniques [25].

In conclusion, VATS clipping for PDA is a safe procedure that has broken new ground in the field of less invasive congenital cardiac surgery. It is effective in LBWIs, but these patients are at augmented risk of complicated course. With correct pre-operative evaluation, VATS clipping may be carried out with a high degree of efficacy in all the ductus diameters < 9 mm. The technique requires minimal operating time and avoids morbidity related to chest wall trauma, percutaneous vascular access and intravascular foreign bodies.

References

Appendix A. Conference discussion

Dr J. Monro (Southampton, United Kingdom): I’m amazed at the age of your patients. I rarely have to deal with a duct in a patient above 1.2 kg, which was your smallest. Do your cardiologists not close ducts? And do you use this technique in the smaller babies under 1.2 kg?

Dr Villa: We have younger series compared to other published cardiological series and we have no weight limitation for the indication.

Dr Monro: But you didn’t have any patients under 1.2 kg.

Dr Villa: No. We don’t have VATS experience in these patients. Referring cardiologists have other options for lowest baby.

Dr Monro: Well, not many. You can try medical treatment, but many of them come to need surgery. The cardiologists can’t close them. They can only close the ones in bigger children. And that’s why I’m surprised you are dealing with these and the cardiologists haven’t closed all the bigger ones.

Dr Villa: Can you repeat the question.

Dr Monro: Well, I think, the mean weight of your patients was 10.7 kg and your age was 3 years. Our cardiologists close the ducts in all those patients. Do your cardiologists not close the older patients that you’re dealing with now?

Dr Villa: No. We collaborate with many cardiologists and lots of them have referred us older patients, but these are selected cases today.

Dr Monro: Well, I’m amazed that you have so many patients. Perhaps this is an unusual experience.

Dr G. Stellin (Padova, Italy): We are sharing similar experience of about 150 cases. First question: Your incidence of laryngeal nerve injury is quite high. I’m just wondering whether or not it’s related to the fact that you apply two clips on the PDA rather than one. What’s the reason for applying two clips, rather than one? Do you think that double clipping may interfere with the laryngeal nerve? Second question: I know that Francois Laborde has closed several ductus arteriosus with robotic assistance. I would like you to make a few comments about robotic assisted PDA closure.

Dr Villa: We think that this transient dysfunction is due to thermal injury and cauterization may provoke it. This kind of dysfunction is usually reversible. And we think that there is no entrapment of the nerve into the clip. Furthermore two clips are better than only one for safer occlusion. We reported last year the robotic experience with the Zeus system, and we found no difference between the robotic group and the thoracoscopic group in term of clinical results. Only duration of the procedure was longer. We think that thoracoscopic approach is a good option.

Dr Stellin: Which are the ductus that you have excluded from VATS closure? And you electively have performed a thoracotomy? How small is the smaller patient? How big is the patient you consider only duct diameter: more than 8 mm is a contraindication to our procedure. We have no limitation due to size of the baby, only the duct diameter is a contraindication.

Dr Villa: You consider only duct diameter; more than 8 mm is a contraindication to our procedure. We have no limitation due to size of the baby, only the duct diameter is a contraindication.

Dr Stellin: Is in premature babies the PDA closed through a thoracotomy?

Dr Villa: Yes, almost always.