Abstract

Objectives: Evaluation of an aggressive policy for the treatment of phrenic nerve palsy (PNP), following cardiac operations, with emphasis on early diaphragmatic plication. Attention was given to the incidence and predisposing factors for PNP and the potential for recovery following plication. Methods: From 1 June 1991 to 1 January 1996 we prospectively screened patients for PNP following cardiac surgery. The diagnosis was suspected if difficulty was experienced in weaning the child from the ventilator. If abnormal elevation of the hemidiaphragm was present diaphragmatic plication was performed. Echocardiography was used to assess subsequent return of diaphragmatic function. Results: Seventeen children (nine boys, eight girls), out of 867 (1.9%) children younger than 16 years of age, undergoing cardiac operations were found to have PNP. The mean age was 66 days (range 1–17 months) with 16 patients below 1 year out of a total of 285 patients (incidence 5.6%) and one patient 17 months old. The incidence following open procedures was 11/190, following closed procedures 2/95 and following reoperation 4/83. PNP was diagnosed from 2 to 44 days (mean 14 days) following surgery. It was present on the right side in seven cases, the left in nine and was bilateral in one patient. Two patients were extubated at the time of diagnosis, one patient could be extubated shortly thereafter. Fourteen children underwent diaphragmatic plication, at a median 5 days post diagnosis. Extubation was possible 1–60 days (mean 4 days) after plication. Mean follow-up was 19–5 months. Subsequent recovery of diaphragmatic movement was documented in seven (41%) children. Time to recovery following plication was 16 months, without plication 38 months. Conclusion: Prospective screening for PNP revealed an incidence in children younger than 1 year of 6%. Early plication substantially reduces the duration of ventilation, with its associated reduced morbidity and ICU stay. © 1998 Elsevier Science B.V. All rights reserved

Keywords: Phrenic nerve palsy; Diaphragmatic plication; Paediatric cardiac surgery

1. Introduction

Phrenic nerve palsy (PNP) after cardiac surgery is a recognized complication which is increasingly reported as more complex operations are performed earlier in infancy [1–3]. The serious morbidity and increased hospital costs associated with PNP in infancy are well documented with a tendency to opt for early surgical plication [2–7]. The aim of this study was to review the incidence and predisposing factors for the development of PNP following cardiac operations and attempt to evaluate an aggressive policy of diaphragmatic plication in addition to assessing the potential for recovery of phrenic nerve function.

2. Methods and materials

From 1 June 1991 to 1 January 1996, 867 cardiac procedures were performed on children below 16 years of age, out of which 285 were in infants younger than 1 year with 190 procedures using cardiopulmonary bypass, 95 without and 83 reoperations. No patient had an abnormal elevation of the diaphragm on preoperative chest X-ray. All patients had a central venous line attempted or positioned via the right internal jugular vein for intraoperative monitoring and medication administration.

If indicated autologous pericardium was harvested cleaning the pericardium superiorly to the level of the innominate vein. A longitudinal incision was made in the pericardium 1 cm anterior...
to the right phrenic nerve. Next a transverse incision is made along the diaphragm extending to within 1 cm of the left phrenic nerve. Superiorly a similar but convex incision is made. A left-sided longitudinal incision is made parallel to the left phrenic nerve.

Cardiopulmonary bypass was instituted with moderate hypothermia (28°C) via an aortic (Medtronic DLP cannula) and bicaval venous cannulation (Medtronic DLP cannulae). Myocardial protection was performed with antegrade crystalloid cardioplegia solution (Plegisol™) initial dose 150 ml/min per m² BSA for 2 min, throughout the procedure the pericardial cavity was irrigated with cold Ringers lactate solution (4°C) to maintain a septic temperature <15°C. Operative notes were reviewed for all patients in an attempt to elucidate a possible cause for injury to the phrenic nerve. PNP was suspected when weaning from ventilatory support was unsuccessful, respiratory insufficiency occurred or an elevated hemidiaphragm was noted on chest X-ray, in the presence of stable haemodynamics. The diagnosis was confirmed by ultrasound examination. Once the diagnosis of PNP had been made and the child could not be readily weaned from the ventilator diaphragmatic plication was performed at the next routine operating list. All plications to flatten the diaphragm in its inspiratory position were performed through a lateral thoracotomy through the 7th or 8th intercostal space. Diaphragm plication was performed using a purse-string stitch to reduce the dome followed by mattress sutures placed in radial orientation on the paralyzed hemidiaphragm [8].

Follow-up echocardiography was used to assess return of diaphragmatic function. Our diagnostic management is outlined in Fig. 1.

3. Results

Out of 867 cardiac procedures performed in patients younger than 16 years of age, 17 (1.9%) children age range 1–510 days (mean 66 days) were found to have PNP. There were nine boys and eight girls. In the group less than 1 year old the incidence was 16 (5.6%) out of 285 cardiac procedures. Patient characteristics are listed in Table 1. The time from surgery to PNP diagnosis was a median of 14 days (range 2–44 days). The incidence following procedures using cardiopulmonary bypass or circulatory arrest was 11/190 (6%), following closed procedures 2/95 (2%) and following reoperation 4/83 (5%). The diagnostic characteristics, operative approach, procedure, PNP side and most likely reason of PNP are documented in Table 1. Extensive dissection of the great vessels and thymus was presumed to be the cause in seven patients, while attempts to obtain a large piece of autologous pericardium was thought to be the reason in nine. The arterial switch operation (8/37) and correction for IAA (2/7) were the procedures thought to be the reason in nine. The arterial switch operation (8/37) and correction for IAA (2/7) were the procedures thought to be the reason in nine. The arterial switch operation (8/37) and correction for IAA (2/7) were the procedures thought to be the reason in nine. The arterial switch operation (8/37) and correction for IAA (2/7) were the procedures thought to be the reason in nine. The arterial switch operation (8/37) and correction for IAA (2/7) were the procedures thought to be the reason in nine. The arterial switch operation (8/37) and correction for IAA (2/7) were the procedures thought to be the reason in nine. The arterial switch operation (8/37) and correction for IAA (2/7) were the procedures thought to be the reason in nine. The arterial switch operation (8/37) and correction for IAA (2/7) were the procedures thought to be the reason in nine.

Subsequent recovery of diaphragmatic movement following plication was documented in four patients, average was 9 ± 3 months (range 16 days to 16 months). One child with PNP who underwent diaphragmatic plication died during the hospital stay. This child known with total anomalous pulmonary venous drainage and progressive pulmonary vein obstruction requiring reoperation, died due to pulmonary hypertension and cardiopulmonary failure. Four patients died during follow-up. One after 3 years following correction of a complex DORV, one after 1 year due to pneumonia following Norwood I palliation, one after 1.5 years due to ventricular failure and persistent desaturation following total cavopulmonary connection and one after 1 year due to pulmonary atresia and lung hypoplasia for which a modified Blalock–Taussig shunt was the initial palliation.

4. Discussion

The exact incidence of PNP after cardiac surgery in the pediatric age group is unknown. Reports of the prevalence of PNP after cardiac operations in children vary from 0.4% [2] and 16% [9] to 49.5% [10]. According to Chan [11] PNP is frequently undiagnosed. If the child is asymptomatic the diagnosis may be missed, if the child is ventilated the diagnosis may not be considered, or is difficult to confirm in the ICU setting. Prospective series suggests that PNP is significantly more prevalent than previously reported in
retrospective series, especially as more complex operations are being performed in even younger children [9]. Chest X-ray and fluoroscopy tend to underestimate the true prevalence. However, children under 1 year seem to have a higher incidence. Although in adults with respiratory difficulty after cardiac operation it has been proposed that phrenic latency should be part of the routine post-operative assessment [12]. No such data have been described for children.

In our prospective series PNP was found in 5.6% of pediatric cardiac surgical procedures done in children under 1 year. This suggests that PNP in this age group might be considerably more prevalent than that so far reported.

With regard to aetiology PNP occurs most frequently from technical accidents during various operative proce-
great vessels [1]. Correction of transposition of the great arteries; Mustard [15], Switch [1], total anomalous pulmonary venous drainage and tetralogy of Fallot [1,15] have a significantly increased risk on PNP. In the group of closed procedures the Blalock–Taussig shunt was the main cause of PNP.

The mechanism of hypothermic damage has been well studied. Experiments in dogs have shown that application of ice chips to the nerve leads to demyelination and axonal degeneration [16]. In man the use of cold solutions within the pericardium has been associated with phrenic injury [17]. Ice slush seems to be a greater risk factor than other cold solutions. Internal jugular vein cannulation is also identified as a possible causative factor. There is little doubt that hilar dissection in a previously operated area such as when taking down a shunt is hazardous.

Our series confirms these observations. The incidence in arterial switch operation in this study is higher than often reported, we believe this to be as a result of the prospective nature of the study and the relative small number of operations performed. As regards the aetiology, extensive autologous pericardial resection, hilar dissection for the Lecompte manœuvre and the use of cold saline are all implicated.

The diagnosis of PNP in infants can be particularly difficult as they have to be capable of spontaneous ventilation to identify paradoxical movement of the affected diaphragm. Early diagnosis has traditionally relied on a high index of clinical suspicion supported by radiographic evidence of elevation of the injured hemidiaphragm and confirmed by fluoroscopy. As chest radiography alone is sensitive to many interpretation errors, due to factors such as positive pressure ventilation and diffuse lung disease and the practical problems of obtaining a good correct and rotated inspiratory film. This has tended to lead to a delay in confirming the diagnosis [12].

Direct percutaneous stimulation of the phrenic nerve probably provides the most accurate diagnosis, and can be applied to patients on full ventilation. The technique can,
Review of the literature concerning incidence of PNP, time from diagnosis to plication and time from plication till detubation

<table>
<thead>
<tr>
<th>Author</th>
<th>Year</th>
<th>Period (years)</th>
<th>Patients (n)</th>
<th>PNP (n)</th>
<th>%</th>
<th>Time to plication (days)</th>
<th>Time to detubation (days)</th>
</tr>
</thead>
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<tr>
<td>Shoemaker [14]</td>
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<td>1500</td>
<td>7</td>
<td>0.4</td>
<td>12</td>
<td>4</td>
</tr>
<tr>
<td>Watanabe [15]</td>
<td>1986</td>
<td>12</td>
<td>76/100</td>
<td>125</td>
<td>1.6</td>
<td>14</td>
<td>2</td>
</tr>
<tr>
<td>Affatato [8]</td>
<td>1988</td>
<td>8</td>
<td>300</td>
<td>18</td>
<td>6</td>
<td>18</td>
<td>3</td>
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<tr>
<td>This series</td>
<td></td>
<td>4</td>
<td>867</td>
<td>17</td>
<td>1.9</td>
<td>5</td>
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</table>

Table 3

References


5. Conclusions

The true incidence of PNP in neonates and small children may be greater than has been realized in the past. Harvesting autologous pericardium and extensive dissecting of the great vessels is associated with a higher rate of PNP.

When PNP is present and weaning from the ventilator is not possible early diaphragmatic plication should be considered.

After diaphragmatic plication recovery of diaphragmatic movement was seen in 4/12 within 9 ± 3 months.

However be painful especially in infants and the presence of venous cannulae may make direct stimulation technically difficult [3]. In our opinion such an approach offers little practical advantage over prompt ultrasound detection. Ultrasound examination is technically straightforward, involves no patient discomfort and is readily repeatable.

The respiratory function of infants who develop PNP is usually compromised to a greater degree than in older children or adults, weak intercostal musculature, horizontal orientation of the ribcage, a mobile mediastinum and recumbent position aggravate the paradoxical motion of the paralyzed diaphragm which can be life threatening [18].

Optimal management of PNP in children who have undergone cardiac surgery remains controversial and consists of prolonged ventilation or diaphragmatic plication. A major difficulty is that the natural history of PNP following cardiac surgery is unknown. Watanabe [15] reported recovery of the affected diaphragm between 5 and 51 days and is thus unpredictable. Further follow-up suggested that 16% never recovered [15], Iverson [19] showed that in many cases of traumatic injury of the phrenic nerve, normal diaphragmatic function would return after 6–12 months. Mickell [13] noted radiographic or fluoroscopic resolution of phrenic paralysis in 95% of their pediatric cases up to 3.5 years after operation.

There are as many protagonists for medical management [2] as there are for surgical plication of the hemidiaphragm [3,5]. The advocates of early plication argue that prolonged ventilation is associated with substantial morbidity and extended hospital stay with extremely costly intensive care (Table 3). Further delay implies multiple and unsuccessful attempts at weaning from the ventilator allowing a potential for increase in pulmonary infection rate and with a substantial mortality [14].

Although the selected use of plication has been accepted the appropriate timing has been more controversial [7]. As indication for plication an arbitrary figure of 2–3 weeks of symptomatic PNP is advocated. Of course plication may be appropriate earlier if the phrenic nerve is known to be divided, or may be delayed if the patient is gradually improving.

Little information exists regarding long-term function after plication of the diaphragm. Most authors report normal function, with the majority of patients remaining free of respiratory symptoms [7,14]. While early plication may reduce ventilation time and hospital stay there is concern that long-term follow-up of these patients demonstrates that only a small proportion shows normal diaphragmatic movement [2]. More sophisticated assessment using phrenic nerve function, pulmonary function or exercise tolerance testing has yet to be reported.

Our aggressive policy of early plication (median 5 days) shortens the total time of hospital stay. The ventilation time after plication (median 4 days) is comparable to other studies (Table 3). The pronounced negative psychological effects of this period on both parents and other family members are not to be underestimated. Subsequent recovery of diaphragmatic movement was documented in 4/12 at an average follow-up of 9 ± 3 months. Acknowledging this our policy has favored surgical plication.