Surgical treatment of atrial septal defect under hypothermia

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Since the development of cardiac surgery, defects of the atrial septum have been recognized with increased frequency. During the past 15 years, various surgical techniques have been used, and even to-day the surgical treatment of this lesion is not uniform. In 1948, Murray first closed an atrial septal defect, suturing together the anterior and posterior atrial walls in the septal plane. Søndergaard, Götzsche, Ottosen, and Schultz (1955) introduced his circumclusion suture, applied after dissection of the interatrial groove between the cavae and the right pulmonary veins. Bailey, Bolton, Jamison, and Neptune (1953) described his technique of atriopexy, and the following year Gross, Pomeranz, Watkins, and Goldsmith (1952) reported his atrial wall operation. Lewis and Taufic (1953) were the first to close an atrial septal defect with direct vision under hypothermia following the experimental work of Bigelow, and soon after Swan (1953) applied this technique to a large series. The modern management of atrial septal defects has been with the use of extracorporeal circulation, first achieved by Gibbon in 1954, by Lillehei, Cohen, Warden, and Varco (1955), and subsequently used by many workers.

Although it is now generally agreed that closed operative techniques are unsatisfactory, there is considerable disagreement as to whether hypothermia or cardiopulmonary bypass should be the method of choice for dealing with the uncomplicated, simpler type of atrial septal defect. Many surgeons, for example C. A. Hufnagel (personal communication, 1960), Gross (1962), and a number of surgeons in this country, recommend bypass for every case of atrial septal defect. They consider the main disadvantage of conventional hypothermia at 30°C to be the time limit of circulatory arrest thereby imposed, usually accepted as eight to 10 minutes. On the other hand, this should be an adequate period for simple suture of an atrial septal defect, and the use of hypothermia avoids heparinization, the considerable volume of donor blood, and the more elaborate apparatus involved in the use of bypass.

In the Department of Thoracic Surgery, Newcastle upon Tyne between 1956 and 1961, 158 patients were treated for an atrial septal defect associated with or without partial anomalous venous drainage and valvular pulmonary stenosis. Of these, 133 were operated on under hypothermia, and two patients with an ostium primum defect were treated under extracorporeal circulation using the AGA, Senning Crawford apparatus. It is the purpose of this paper to present the findings and results in this series of cases treated by this method of open-heart surgery, and to discuss the place of hypothermia in the surgical treatment of atrial septal defect.

THE SERIES

The age and sex distribution of the patients is shown in Table I.

Table I

<table>
<thead>
<tr>
<th>Age and sex of 135 patients with atrial septal defect</th>
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<tbody>
<tr>
<td>Age (years)</td>
</tr>
<tr>
<td>Females . .</td>
</tr>
<tr>
<td>Males . . .</td>
</tr>
<tr>
<td>Total . . .</td>
</tr>
</tbody>
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As in other reports in the literature, it will be seen that the lesion was more common in the female and that most of the patients were under the age of 20 years. Atrial septal defect was the commonest single cardiac anomaly seen in the Unit, and this corresponds with the experience of Paul Wood (1956), of Bedford and Sellors (1960), and of Nadas (1963).
ANATOMICAL TYPE OF DEFECT

It is customary to classify atrial septal defects into four main categories: secundum or fossa ovalis defect; sinus venosus defect; ostium primum defect; and atrio-ventricularis communis.

SECUNDUM TYPE This is the commonest type of atrial septal defect and accounts for 85 to 90% of cases in all published series. It includes two sub-types. The commonest type is the fossa ovalis defect, which occupies part or whole of the fossa ovalis and which may be fenestrated. The inferior cava form has no lower border so that the inferior cava opens directly into both atria.

SINUS VENOSUS DEFECT This is embryologically distinct; it lies at the upper end of the atrial septum and is associated with an abnormal connexion of the right, upper, and middle lobe veins. The fossa ovalis may be intact or may contain a second defect. We have seen only one case of sinus venosus defect with a second low defect of the foramen ovale variety. Seven other cases of sinus venosus defects were encountered in the present series. It has been described by Lewis (1958), Brock and Ross (1959), and McCormack, Marquis, Julian, and Griffiths (1960).

OSTIUM PRIMUM DEFECT This defect lies low in the atrial septum in front of the coronary sinus, and its inferior margin is formed by a ridge of endocardium between the septal cusps of the atrio-ventricular valves and lying immediately above the superior edge of the ventricular septum. The mitral and tricuspid valves are normal. This defect accounts for 5 to 10% of atrial septal defects.

ATRIO-VENTRICULARIS COMMUNIS This is the least common type of defect and the most complicated. There is an ostium primum and it is associated with a cleft in the septal cusp of the mitral valve and sometimes also with a similar cleft in the septal cusp of the tricuspid valve. In the most serious abnormality, the ostium primum and these two valve clefts are associated in addition with a defect in the upper part of the ventricular septum.

ASSOCIATED ANOMALIES As in other series, it was found that the two commonest associated defects were pulmonary valvular stenosis and partial anomalous connexion of the pulmonary veins. This occurred in four and in 16 patients respectively in this series. A less common associate was mitral stenosis (Lutemabacher's syndrome), and there were only two examples. However, Bedford and Sellors (1960) found an incidence of rheumatic mitral stenosis of 8.5% in atrial septal defects. The anatomical types of the defect are summarized in Table II.

FUNCTIONAL EFFECTS OF THE DEFECT

The important functional effect of an atrial septal defect is the occurrence of the left-to-right shunt of blood at atrial level. This shunt occurs because the right ventricle is more easily distended, and it has been described by Dexter (1956). As a result of the shunt, the right ventricular output and pulmonary blood flow are increased, commonly to two to four times that of the systemic flow. Pulmonary vasodilatation usually prevents a serious rise of pulmonary arterial pressure, but Bedford and Sellors (1960) reported the development of pulmonary arterial obliterative changes in about 12% of cases. The left-to-right shunt is increased when any of the pulmonary veins connect directly to the right side of the heart or to the cavae. Mitral stenosis increases the left-to-right shunt. The result of the right ventricular overload is progressive hypertrophy of the right side of the heart and eventually right-sided failure.

CLINICAL FEATURES

All the patients in this series presented the typical clinical features of an atrial septal defect first described by Bedford, Papp, and Parkinson (1941).

The soft pulmonary systolic murmur of relative pulmonary stenosis was associated with fixed splitting of the pulmonary second sound, and in those with a fairly large shunt a mid-diastolic tricuspid flow murmur was heard. Those with large shunts were under-developed and had mild peripheral cyanosis. In uncomplicated cases, the jugular venous pulse had a normal configuration. The right ventricle was hyperdynamic depending upon the degree of shunt. In cases complicated

<table>
<thead>
<tr>
<th>Type of Defect</th>
<th>No.</th>
</tr>
</thead>
<tbody>
<tr>
<td>Simple secundum defect</td>
<td></td>
</tr>
<tr>
<td>Fossa ovalis</td>
<td>81</td>
</tr>
<tr>
<td>Inferior type</td>
<td>31</td>
</tr>
<tr>
<td>Sinus venosus defect</td>
<td>8</td>
</tr>
<tr>
<td>A.S.D. with partial anomalous venous drainage of the right lung</td>
<td>8</td>
</tr>
<tr>
<td>A.S.D. and pulmonary stenosis</td>
<td>4</td>
</tr>
<tr>
<td>A.S.D. and mitral stenosis</td>
<td>1</td>
</tr>
<tr>
<td>Ostium primum defect</td>
<td>2</td>
</tr>
<tr>
<td>Total</td>
<td>135</td>
</tr>
</tbody>
</table>
FIG. 1. Pre-operative E.C.G. of a patient with an ostium secundum defect which shows right axis deviation and right ventricular hypertrophy. There is a QR in $V_1$, which is seen in 20% of patients. An rSR' in $V_1$ would be the more typical finding.

FIG. 2. Typical E.C.G. of a patient with an ostium primum defect. Note the left axis deviation and rSR' in $V_1$. There is evidence of biventricular hypertrophy. The broad P wave in lead II and deeply negative P in lead $V_1$ suggest left atrial enlargement.
with pulmonary stenosis, the above signs persisted, but the right ventricle was heaving and the systolic murmur was of grade 3 to 4 with a systolic thrill felt in the pulmonary area.

The two patients with ostium primum defects had the following, in addition to the signs of those with uncomplicated atrial septal defects. The left ventricle was hyperdynamic and there was a marked systolic murmur, usually of grade 2 to 3, best heard in the mitral area. This was either pansystolic or late systolic.

Fifty of the patients presented without symptoms. Of the remainder, the commonest presenting symptom was exertional dyspnoea, present in 65 patients, and others complained of recurrent bronchitis, fatigue, or pain and tightness in the chest. Three patients were in cardiac failure when first seen; one of these had an ostium primum defect. The electrocardiogram showed incomplete right bundle branch block with right axis deviation in almost all patients, and in those with elevation of pulmonary arterial pressure or in those with an associated pulmonary stenosis, there was E.C.G. evidence of right ventricular hypertrophy (Fig. 1). In the two cases of primum defect, there was left ventricular hypertrophy and left axis deviation (Fig. 2). The characteristic radiological features consisted of enlargement of the right atrium, right ventricle, and main pulmonary artery and considerable pulmonary plethora. These appearances were not seen in those with small shunts and were, as would be expected, most marked in the patients with the largest shunts.

**INDICATIONS FOR SURGICAL INTERVENTION**

Our recent increase of knowledge of the natural history of atrial septal defect, the risks of operation, and the results achieved has given us a clearer understanding of the indications for surgical treatment. We have accepted the principle that, when a diagnosis of atrial septal defect is made and when the pulmonary/systemic blood flow ratio is more than 2:1, the defect should be repaired. We have followed the advice of Bedford and Sellors (1960), Wood (1956), Brom and Kalsbeek (1959), Gross (1962) and others who have stated that the proper time to close an atrial septal defect is before the age of 20, i.e., before the heart and pulmonary vessels have suffered irreversible changes. This allows operative treatment to be undertaken with the smallest risk and the greatest chance of subsequent success. The only absolute contra-indication to surgery is obstructive pulmonary hypertension with a vascular resistance exceeding 4 to 5 units and a pulmonary to systemic ratio of less than 2:1. However, in patients over 40 with marked cardiac enlargement, pulmonary hypertension, atrial fibrillation, and cardiac failure, operative morality is high, probably in the region of 15% or more. Although many of these patients are still suitable for operation, the risk is greater and the results are less good.

**PRE-OPERATIVE INVESTIGATIONS** In addition to the standard clinical examination and radiographic films of the heart and lungs, all patients had an electrocardiogram made and underwent cardiac catheterization. Angiography was performed only in cases associated with partial anomalous venous drainage or pulmonary valvular stenosis. A left-to-right shunt was calculated in all patients with a simple atrial septal defect. Four patients with associated pulmonary valvular stenosis had a right-to-left shunt. A pulmonary artery mean systolic pressure of 50 mm. Hg was present in only 12 patients, and none had a significantly increased pulmonary vascular resistance.

**OPERATIVE TECHNIQUE** After induction with thiopentone, anaesthesia was continued with a nitrous oxide oxygen mixture, and a relaxant was routinely used. The anaesthetic was maintained through an endotracheal tube, and controlled ventilation was used throughout. The temperature was monitored with thermocouples in the mid-oesophagus and in the nasopharynx, and a continuous E.C.G. tracing was maintained throughout the procedure.

Hypothermia was induced by surface cooling achieved by ice packs placed on the skin in the recovery room where the patient remained until his oesophageal temperature had reached 32° C. Thereafter the patient was transferred to a re-warming blanket on the operating table through which water at 37° C. circulated. By this method it was almost always possible to have the patient's temperature very close to 30° C. at the time of inflow clamping.

The operative approach was through the bed of the fifth rib with division of the sternum. Both internal mammary arteries were divided between ligatures and the pericardium was cleared. The pericardium was opened and retracted to the right over the lung. The heart was inspected for associated anomalies and the atrial septal defect was palpated through the atrial wall. Preliminary trans-aortic exploration has not been practised. Tapes were placed around the cavae, and blood samples were taken from the cavae and atria for oxygen saturation measurements. A 5 cm incision was made in the wall of the right atrium within the jaws of a Satinsky clamp. The caval tapes were tightened, and, after a few beats to allow cardiac emptying, the great arteries were
occluded with a clamp through the transverse sinus. The Satinsky clamp was then removed and the right atrium was cleared of blood by suction. The lower margin of the defect was sutured first with a 3–0 suture of silk. A similar stitch was then passed through the upper margin, and the defect was closed with a double layer of everting stitches (Fig. 3). In the case of the inferior vena caval defect, care was always taken to ensure closure of the defect to the left of the cava. Before completing closure of the defect, the anaesthetist inflated the lungs, thereby filling the left atrium with blood and expelling all air from it. After closure of the defect, the inferior caval tape was released to demonstrate that the inferior cava was flowing into the right atrium. When the right atrium had filled with blood, the Satinsky clamp was reapplied to the atrial incision, and the superior caval tape and the transverse sinus clamp were removed. Normal heart action usually resumed spontaneously. The atrial wall was closed with a double everting suture. In order to determine whether the defect had been completely closed, 10 minutes after restoration of the circulation blood samples were taken from the superior vena cava and from the right and left atria and were analysed for oxygen content. The pericardium was closed and the chest wall was closed with pleural drainage.

No case of secundum defect has required a prosthesis for its closure, and there has been no observed breakdown of the suturing line.

The size and site of the simple secundum defects were as follows. All were of the fossa ovalis variety. There were 94 between 3 and 4 cm. in size occupying the fossa ovalis, eight of which were fenestrated; 31 were a large defect of the inferior type of 5 to 7 cm., and 16 of these had no posterior septal ring. Surface cooling was applied in all but one patient, in whom venovenous cooling was performed according to Brock's technique.

Associated partial anomalous pulmonary venous drainage In all 16 patients with partial anomalous pulmonary venous connexion, operative correction was undertaken under hypothermia. Eight patients had a sinus venous defect and the characteristic associated anomaly, so that the right upper and middle pulmonary lobes drained into the dilated lower part of the superior vena cava. In five of these the technique of caval partitioning was used (Bahnsen, Spencer, and Neill, 1958). In two patients the technique described by Brock and Ross (1959) was used; in this the crescentic lower edge of the defect is sutured horizontally to the posterior caval wall above the anomalous veins. In the remaining patient the right upper vein was cut off from the superior vena cava and anastomosed to the lower vein. During this patient's convalescence there was radiological evidence of pulmonary infarction, but a satisfactory recovery was made. The importance of occlusion of the appropriate pulmonary artery during the venous anastomosis was not at that time appreciated, and this may have contributed to the occurrence of infarction.

In eight patients there was either partial or complete anomalous connexion of the right pulmonary veins to the right atrium, and in all of these the pulmonary venous drainage was corrected by sutting the medial edge of the atrial septal defect to the right atrial wall in front of the anomalous venous openings.

Atrial septal defect combined with pulmonary valvular stenosis The four patients who had both pulmonary valvular stenosis and an atrial septal defect have had both anomalies corrected at the same operation through a vertical sternotomy. The valvulotomy was performed first and the circulation was re-established for 10 minutes to allow cardiac recovery. The atrial septal defect was then repaired during a second period of inflow occlusion. The valvular stenosis was most often due to a funnel shape, and two cuts were made converting it to a bicuspid valve. We have the clinical impression that less insufficiency may result if two cuts are made. The pressure gradient across the valve was recorded before and after valvulotomy.

Ostium primum defects The two patients who had a primum defect were operated upon with the standard technique of extracorporeal circulation combined with hypothermia at 18° C. body temperature. In both, a teflon prosthesis was used to close the defect, and in one the cleft mitral
valve was repaired (Fig. 4). Convalescence was uneventful in both.

Arrhythmia during operation The only important arrhythmia was ventricular fibrillation, and this occurred in 18 patients, in all but one immediately after the atrial closure. No patient who ultimately developed ventricular fibrillation had a temperature below 28°C, and most were kept between 29 and 30°C. All patients who developed ventricular fibrillation had undergone circulatory arrest for six and a half minutes or longer. Four patients died as a result of ventricular fibrillation. In three, normal rhythm was never re-established, and in the fourth, although the rhythm eventually reverted to normal after some two hours of massage, the patient never regained consciousness and died from cerebral anoxia. In the remaining 16 patients, normal rhythm was re-established by means of a period of cardiac massage followed by electrical defibrillation and sometimes associated with re-warming of the heart with warm saline. Ventricular fibrillation occurred more often in our early experience with hypothermic anaesthesia. It is well known that the lower the temperature the more likely is the complication; 30°C is probably the safest lower limit. Other factors inducing ventricular fibrillation are the prolongation of the period of inflow occlusion and probably the over-distension of the right side of the heart by the simultaneous release of the venae cavae. Our experience indicated that there is good correlation between low temperature, period of inflow occlusion, and ventricular fibrillation.

The use of coronary perfusion for protecting the myocardium during the inflow occlusion has not been practised; we do not believe that coronary perfusion is necessary. Experience with extracorporeal circulation has shown that 10 to 15 minutes hypoxic cardiac arrest is well tolerated by the myocardium. If cardiac arrest of longer than eight minutes is required, then the intermittent inflow occlusion is advisable. As experience has grown, the incidence of ventricular fibrillation has decreased, so that it is now a rare occurrence.

Operative mortality There were six deaths in the series. Apart from one 6-year-old child, all the deaths occurred in adults, and the commonest cause of death was ventricular fibrillation occurring during operation. As detailed above, three patients died on the operating table with irreversible ventricular fibrillation, and a fourth died of cerebral anoxia after a long period of ventricular fibrillation. One 42-year-old patient died of cerebral embolism on the day after operation, never having regained consciousness, and the remaining patients died on the seventh post-operative day of staphylococcal septicaemia uncontrolled by antibiotics.

Post-operative complications Apart from the fatalities, the only important complication was empyema, which developed in three patients, two of whom subsequently needed drainage by rib resection. Atrial fibrillation was a common occurrence and appeared post-operatively in more than half of the patients. It was usually of short duration, but in three patients it persisted for six months. In two, a sinus tachycardia of 160 per minute persisted for six months. Two patients had transient atrial flutter, and one had heart block for 24 hours.

Discussion

Though the surgical management of atrial septal defect has become increasingly satisfactory, surgical opinion is still divided as to whether the best approach is visual closure of the defect under 30°C hypothermia or with the use of extracorporeal circulation.

Several clinics in the United States of America and in Europe have effectively used moderate hypothermia alone without the additional use of cardiopulmonary bypass for the surgical correction of an atrial septal defect and pulmonary valvular stenosis.
FIG. 5. Postero-anterior view of the chest showing the cardiac silhouettes of a patient who underwent open-heart surgery for the closure of a secundum type defect: (above) pre-operative, (below) two years post-operatively.
Although an efficient pump oxygenator for open-heart surgery has been in use in our clinic since 1959, we have continued to use the inflow occlusion technique under $30^\circ$ C. hypothermia for all patients with an atrial septal defect of the secundum variety, whether or not it is associated with partial abnormal insertion of the pulmonary veins and pulmonary valvular stenosis.

When the clinical, radiological, E.C.G., and haemodynamic studies were all consistent with the presence of a secundum defect, this diagnosis was invariably substantiated at operation. If any of the features suggest a primum defect, operation should be performed using extracorporeal circulation.

We believe that the surgical correction of an atrial septal defect of the secundum variety can easily be achieved under $30^\circ$ C. hypothermia without the use of prosthetic materials. The expulsion of air from the left atrium might be easier with the use of cardiopulmonary bypass but this has not been a problem in our series.

The unexpected discovery of partial anomalous venous drainage at operation does not give cause for concern, for the operation is the same, and the technique described by Brock and Ross (1959), Bahnson et al. (1958), and Risch and Hahn (1958) can be used effectively with minimal operative risk and good results. When pulmonary valvular stenosis is present it is not necessary to use the heart–lung machine. The hypothermia technique with intermittent inflow occlusion can be used successfully for the correction of both anomalies in one operation within the time limits imposed by hypothermia at $30^\circ$ C.

Our experience indicates that the technique of inflow occlusion under $30^\circ$ C. hypothermia has an acceptable low operative mortality with good clinical results and is applicable to all atrial septal defects of the secundum variety with the associated anomalies.

This method has the merits of simplicity and economy, and requires minimal personnel; in this respect it offers distinct advantages over extracorporeal circulation. We believe that cardiopulmonary bypass is necessary for the correction of the most complicated defects of the primum variety.

After operation almost all symptomatic patients noticed an improvement in their physical condition with a lessening or disappearance of breathlessness and fatigue. Underdeveloped children showed an improvement in growth, weight gain, and exercise tolerance. Clinical examination revealed that splitting of the second sound usually became closer on expiration but in some cases did not become single on expiration. The tricuspid dia-

stolic murmur always disappeared, and a pulmonary systolic ejection sound, when present, always persisted. Post-operative cardiac catheterization was performed in five patients two years after operation. In none of these patients were signs of communication between the atria found. Radiological examination showed that large hearts became smaller in one-third of the cases, and the right atrium was also less prominent (Fig. 5). In the remaining patients the size of the heart was unchanged. Of the patients who developed atrial fibrillation and atrial flutter, post-operative E.C.G. reports showed that both the atrial fibrillation and the atrial flutter had reverted to normal rhythm within 48 hours in the majority of patients. All patients were treated with digitalis. However, normal reversion in three patients took six months, and in one patient paroxysmal tachycardia occasionally develops.

**SUMMARY**

The technique of inflow occlusion under $30^\circ$ C. hypothermia was adopted for direct visual correction in 133 consecutive cases of atrial septal defect of the secundum variety and the associated anomalies.

The use of extracorporeal circulation combined with hypothermia was reserved for the repair of two cases of ostium primum defects.

The over-all operative mortality was 4%.

Post-operative evaluation showed that the results have been good in almost all patients, including those with pulmonary hypertension.

Since an ostium primum defect requires cardiopulmonary bypass for its closure, the pre-operative recognition of the defect is highly desirable in the selection of patients for operation and in planning the procedure itself.

The information derived from the clinical examination and appropriate diagnostic studies has been found to be consistently accurate in the pre-operative differentiation of the primum from the secundum defect. Therefore, the unexpected discovery of a primum defect at operation was not a problem.

In our experience, open-heart surgery under $30^\circ$ C. hypothermia for the repair of an atrial septal defect of the secundum type and the associated anomalies is a safe and practical technique and effectively corrects the anomaly with minimal risk.

My thanks are due to Mr. George Mason, senior surgeon who operated on these cases, to Dr. Joan
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Millar, senior anaesthetist, and to Dr. C. B. Henderson, cardiologist to the Regional Thoracic Surgical Clinic at Shotley Bridge General Hospital, Newcastle upon Tyne, for their help and advice in reporting these cases. I also wish to thank Dr. Khakoo for the follow-up of the cases.

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