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(REVIEW ARTICLE)

Complicated Dextroversion of the Heart: A rare constellation of multiple congenital malformations in an infant: Case report and literature review

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Abstract

Dextroversion of the heart is a form of dextrocardia resulting from congenital malrotation of the heart about its long axis. The earliest reference to the condition is credited to Paltauf, who, in 1901, differentiated it from the other forms of dextrocardia. Since then, a considerable volume of information regarding this type of right-sided heart has accumulated. In spite of the fact that the radiologist is often the first to be confronted with the anomaly, only scant information concerning it is available in the radiological literature. Dextroversion may exist in association with an otherwise normally developed heart or it may be accompanied by numerous varieties of congenital cardiac anomalies.

Dextroversion is an uncommon congenital anomaly characterized by situs solitus (normal position) of thoracic and abdominal viscera with right cardiac apex. Isolated dextroversion, i.e., without associated congenital heart disease, is rare, but its occurrence permits adult survival, setting the stage for late development of acquired heart disease. Exceptionally, dextroversion with abdominal heterotaxy has been mentioned by Robert P. Grant after reviewing the literature of 122 patients of dextroversion of heart in 1958.

Herewith, we are reporting a case of dextroversion with situs solitus in association with a characteristic conundrum of multiple and rare congenital cardiac defects, in a 6 month old infant.

Keywords: Dextroversion; Dextrocardia; Isolated dextrocardia; Situs solitus; Mitral atresia; Double outlet right ventricle; Hypoplastic left ventricle

1. Introduction

In normal anatomy, the morphologic right atrium and ventricle are to the right of the morphologic left atrium and ventricle. However, in congenital cardiac malposition, chamber location is variable in relation to its morphologic counterpart. Dextrocardia is malpositioning of the heart, with the cardiac apex in the right hemithorax [Figure 1, 2].

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Figure 1 Dextrocardia cadaveric specimen. 1-pulmonary trunk; 2-ascending aorta; 3-superior vena cava; 4brachiocephalic trunk; 5-left common carotid artery, 6-left subclavian artery: 7-thoracic aorta; 8-hepatic tissue covering the inferior vena cava

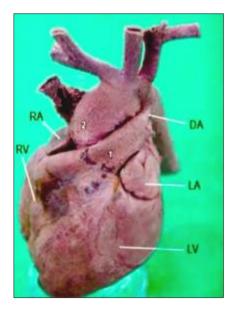


Figure 2 Dextrocardia cadaveric specimen illustrating cardiac chambers: RA-right auricle; RV ventricle: DA-ductus arteriosus; 1-right ventricle; LA-left auricle; LV-left-pulmonary trunk; 2-ascending aorta

Dextrocardia is a rare congenital abnormality in the general population, with an estimated incidence of 1 in 12,000 live births [1] and may be associated with significant additional cardiac malformations. The male-to-female incidence is 1:1. Dextrocardia with a normal abdominal situs has a high incidence of associated congenital cardiac anomalies, on the other hand, dextrocardia with situs inversus is associated with a lower incidence of congenital heart disease, 0-10% [2].

The arrangements of the position of the abdominal viscera in dextrocardia may be normal (situs solitus), reversed (situs inversus), and indeterminate (situs ambiguous or isomerism) in 32–35%, 35–39%, and 26–28% of cases, respectively [1, 3]. It is noteworthy, that with situs solitus and situs inversus, the atrial situs always corresponds to the visceral situs [4, 5].

Dextroversion refers to a type of dextrocardia in which the orientation of the chambers is such as would be obtained where the heart was rotated or pivoted to the right along the horizontal plane with the atria as a fulcrum [6] (Figure 3).

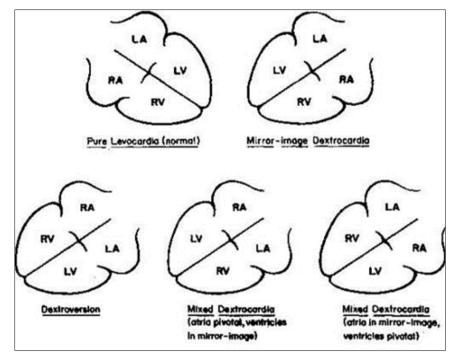


Figure 3 Diagrammatic sketches of positions of chambers in various types of dextrocardia, using the position of chambers in the normal position of the heart (pure levocardia) as a reference point. The various chambers should be referred to fundamentally from the standpoint of their morphology, and secondarily from the standpoint of position. RA-morphologic right atrium; RV-morphologic right ventricle; LA-morphologic left atrium; LV-morphologic left ventricle

Dextrocardia with situs solitus, D-loop ventricles, and normally related great arteries is termed dextroversion which results from failure of the final leftward shift of the ventricles during embryologic development (Figure 4). The morphologic right atrium and morphologic right ventricle remain to the right of the corresponding left-sided chambers [7, 8]. This is the 2nd most common type of dextrocardia and has a 90% incidence of additional cardiac malformations including corrected transposition of great arteries, anomalous pulmonary venous return, tetralogy of fallot's and septal defects [7, 8].

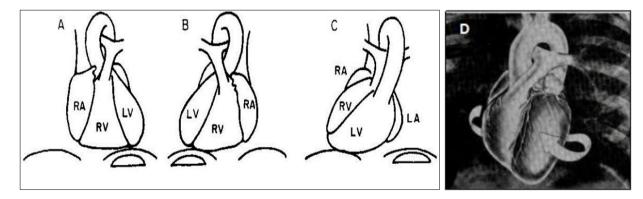


Figure 4 (A) the normal heart, (B) classical mirror-image dextrocardia, (C) classical dextroversion. The position of the 4 cardiac chambers, the great vessels, and the gastric gas bubble are shown for each. Note the difference in cardiac

silhouette between mirror-image dextrocardia and dextroversion. In the latter, the apex is blunter, there is a prominent shadow high on the right side due to the right atrium, and the transposed aorta forms a prominent slow curve on the upper left side of the silhouette in the typical ease. (D) Drawing of heart in dextrorotation superimposed on frontal angiocardiogram. The ventricular septum (identified by the anterior descending branch of the left coronary artery) is displaced to the right and is perpendicular to the frontal plane of the thorax. The ventricles lie side by side,

the right atrium is posterior and superior, and the aortic loop is wider than normal. The "apex" of the heart is on the right and is produced by the right ventricle

Dextroversion is a rare congenital abnormality with an estimated incidence of 1/2800 [9, 10]. This condition is different from dextrocardia with situs inversus, where a mirror-image reversal but preserved relationship exists between the heart, great vessels, and abdominal organs [10].

There are 3 major differences between dextroversion and mirror-image dextrocardia [11]

- In dextroversion the atria have a normal position but the ventricular heart lies rightward, as if swung like a pendulum through an arc of 120 degrees in the frontal plane from its normal position; there is also a 90 degree counter-clockwise rotation of the ventricular heart on its long axis, with the right ventricle lying superiorly to the left ventricle. In contrast with this, in mirror-image dextrocardia all cardiac structures are rightward mirror-images of the normal heart.
- The vast majority of cases of dextroversion (90 per cent of autopsied cases) have additional intracardiac malformations, usually of the conotruncal region, moulding transposition of the great vessels which is present in over 80 per cent of cases. while in mirror-image dextrocardia additional intracardiac anomalies are probably no more frequent than in the population at large.
- An abdominal heterotaxy is often present in dextroversion, which is different from the situs inversus that accompanies mirror-image dextrocardia: instead of inversion of abdominal organs there is a tendency for abdominal structures to be primitive and bilaterally symmetric, frequently with congenital absence of the spleen.

Moreover, among the 69 cases of dextroversion in which autopsy data were adequate, at least 16 had some degree of abdominal heterotaxy [11]. Of these 8 had cor bilocular (2 chambered heart) and 5 of these had the combination of a single pulmonary vein, single atrium, single atrioventricular ring, single ventricle, and single outflow organ (truncus arteriosus) [12-17].

2. Diagnostic modalities for detection of dextroversion

There are multiple techniques available to diagnose dextroversion of heart as mentioned below:

- Resting supine ECG
- Xray chest (PA view)
- Color echocardiography
- Cardiac CT
- Cardiac MR
- Cardiac angiography

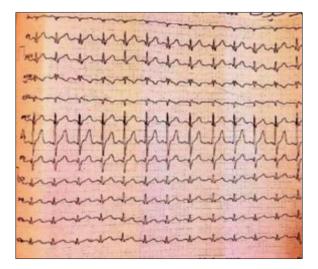


Figure 5 Resting ECG in dextroversion. Patient ECG showing extreme counterclockwise rotation of the heart, and a high R wave in all leads from the left and right side of the chest (due to the anterior position of the left ventricle). Notice positive P wave in lead II and negative P wave in lead aVR denoting situs solitus



Figure 6 X-ray chest PA view in dextroversion. Chest X-ray showing the heart in the right chest with the descending aorta located to the left of midline (arrow). Ao, aorta

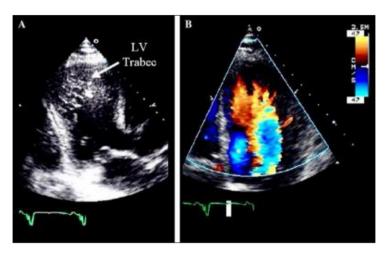


Figure 7 Color echocardiography in a patient of dextroversion with left ventricular non-compaction. Two-dimensional echocardiogram (apical four chamber view), depicting prominent trabeculations in the left ventricle (arrow) (A). Color Doppler examination (B) shows penetration of blood flow into the sinusoidal recesses. LV, left ventricle

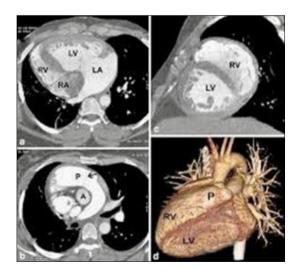


Figure 8 Cardiac CT in a patient of dextroversion. Images from ECG-gated cardiac CT. (a) Axial CT image shows that the cardiac apex is in the right hemithorax with the left ventricle (LV) in a substernal location anterior to the right ventricle (RV). The left ventricle and left atrium (LA) remain to the left of the right ventricle and right atrium (RA) respectively. (b) Axial CT image at a more superior level demonstrates that the pulmonary outflow tract of the right

ventricle (P) passes anterior to the aorta (A) so the pulmonic valve (arrow) remains anterior to the aortic root. (c) Reformatted CT image in the short axis plane relative to the cardiac chambers shows that most of the right ventricle (RV) is posterior and superior to the left ventricle (LV). (d) Volume-rendered image in an anterior view provides a three dimensional perspective of the locations of the right ventricle (RV), left ventricle (LV) and pulmonary outflow tract of the right ventricle (P)

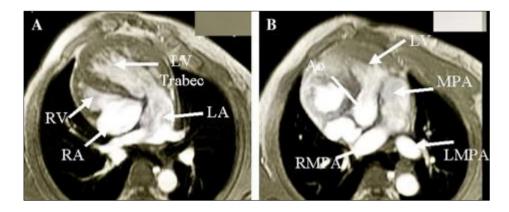


Figure 9 Cardiac MRI in a patient of Dextroversion. MRI long axis depicting normal location of the cardiac atria and great vessels, situs solitus, with the heart's apex rotated into the right chest, dextroversion (A). The pulmonary artery is seen in the left chest, lateral to the aorta, situs solitus (B). Ao, aorta; RA, right atrium; RV, right ventricle; LA, left atrium; LV, left ventricle; MPA, main pulmonary artery; LMPA, left main pulmonary artery; RMPA, right main pulmonary artery

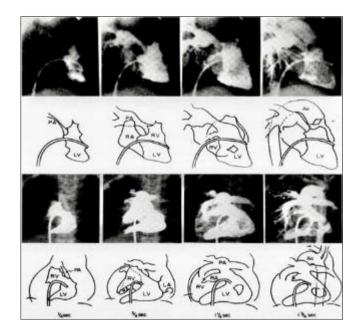


Figure 10 Angiocardiography of a patient of dextroversion, VSD and transposition of great arteries. Simultaneous lateral (top) and anterior-posterior (bottom) biplane angiocardiogram. The opacification for each film is diagrammed below it, and the time of the film after injection of the contrast material is indicated at the bottom of the figure. The position of the catheter through which the contrast material was injected is shown. Both ventricles fill in the first film, demonstrating the presence of a ventricular septal defect. In the second and third anterior-posterior films, the plane separating the right and left ventricles is well defined, indicating that the ventricular septum lay more or less perpendicular to the frontal plane of the body, with the right ventricle superior to the left ventricle. In the fourth lateral film, it can be seen that the aorta is anterior to the pulmonary artery, demonstrating the presence of transposition of the great vessels; usually the aorta has a more leftward position than in this case

3. Case Report

A 6 month old male infant was referred to us for comprehensive cardiac evaluation and transthoracic echocardiography. The child was a full-term normal delivery born out of non-consanguineous marriage. There was no history of maternal risk factors of CHD (obesity, diabetes, febrile illness, smoking, alcohol intake, teratogenic drug use, or radiation exposure). The history was narrated by the parents. They informed that the child was cyanotic since birth and moreover, the cyanosis became more apparent when the child used to cry. Additionally, they gave history of severe breathlessness, recurrent intercostal retractions, failure to thrive and recurrent chest infections. However, they denied any history of loss of consciousness or swelling over feet / face.

On clinical examination, the patient was thin built, apparently very sick and markedly breathless (Figure 11A). Cyanosis was recognised by bluish colouration of the lips, tips of fingers and toes. There was pectus excavatum deformity of the chest (Figure 11B). The infant's weight was 3.5 kg, height was 23 cm, pulse rate was 132/min, blood pressure was 60/40 mmHg, respiratory rate was 30/min and SPO2 was 77% at room air. All the peripheral pulses were normally palpable without any radio-femoral delay. However, no clubbing was detected.

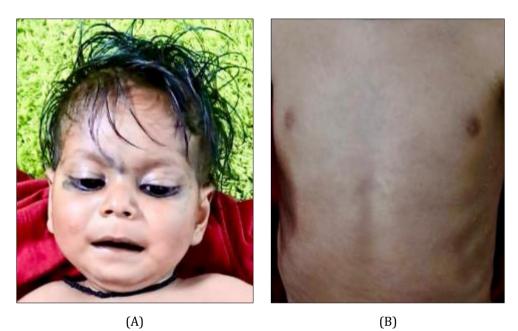


Figure 11 (A) Apparently sick looking child with bluish colouration of lips. (B) Thin, protruding chest with pectus excavatum deformity

On cardiovascular examination, there was presence of grade 2/6 ejection murmur in the pulmonary area. The first heart sound was normal and the second heart sound was loud & closely split. There was no clicks or gallop sound heard. Rest of the systemic examination was unremarkable.

Xray chest PA view (Figure 12A) was suggestive of dextroversion with apex pointing towards the right. Some features of abdominal heterotaxy were appreciated; midline liver, right sided gastric bubble and intestinal malrotation.

Ultrasound of the abdomen (Figure 12 B-D) confirmed the presence of midline liver alongwith solitary spleen demonstrated in the left hypochondrium.

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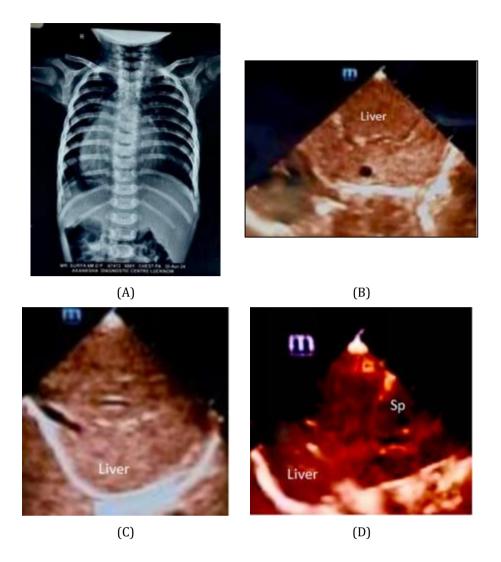


Figure 12 (A) X-ray Chest (PA). Dextroversion is recognized with an apex pointing towards the right. Some features of abdominal heterotaxy is characteristically appreciated, with the presence of midline liver, right gastric bubble and suspected intestinal malrotation; (B) & (C) Ultrasound of abdomen identifies midline liver with right side of liver lying in the right hypochondrium and left side of liver lying in the the left hypochondrium; (D) Solitary spleen is demonstrated in the left hypochondrium

Resting ECG showed P wave was positive in lead I with deep Q waves in lead I and II (Figure 13). There was gradual diminution of R waves from V₁-V₆, thus suggesting dextroversion.

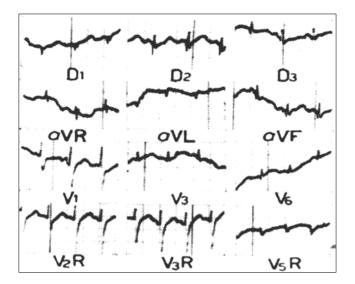


Figure 13 An electrocardiogram showed P wave in lead I is positive with deep Q waves in leads I and II. The voltage of ventricular complexes decreases from V₁ through V₆, suggesting dextroversion.

4. Transthoracic Echocardiography

All echocardiography evaluations were performed by the author, using My Lab X7 4D XStrain echocardiography machine, Esaote, Italy. The images were acquired using a pediatric probe equipped with harmonic variable frequency electronic single crystal array transducer while the subject was lying in supine and left lateral decubitus positions.

Conventional M-mode, two-dimensional and pulse wave doppler (PWD) and continuous wave doppler (CWD) echocardiography was performed in the classical subcostal, parasternal long axis (LX), parasternal short axis (SX), 4-Chamber (4CH), 5-Chamber (5CH) and suprasternal views. Contemporary sequential segmental approach for echocardiographic analysis of our index patient was accomplished and the characteristic features were outlined (Figures 14-19).

4.1. Mode Echocardiography

M-mode echocardiography of right and left ventricle was performed and the estimated measurements are outlined.

Table 1 Calculations of M-mode echocardiography

Measurements	RV	LV
IVS d	2.5 mm	1.7 mm
LVID d	23.3 mm	9.0 mm
LVPW d	4.8 mm	3.1 mm
IVS s	4.8 mm	3.3 mm
LVID s	16.1 mm	6.7 mm
LVPW s	5.0 mm	4.0 mm
EF	61 %	56 %
%LVFS	31 %	26 %
LVEDV	18.8 ml	1.5 ml
LVESV	7.2 ml	0.684 ml
SV	11.5 ml	0.858 ml
LV Mass	14 g	2 g

4.2. Summary of M-mode echocardiography

The RV was dilated with normal systolic function - RVEF 61%. RV mass was 14 g. Conversely, LV was hypoplastic with normal systolic function - LVEF 56 %. There was paradoxical systolic motion of ventricle septum. The LV mass was 2 g.

4.3. Dimensional Color Echocardiography

Transthoracic color echocardiography exhibited multiple features as mentioned below:

- Levocardia
 - Situs Solitus
 - AV concordance
 - o D-loop ventricles
 - Normally related great arteries (NRGA)
 - Left aortic arch
 - Confluent pulmonary arteries.
 - Normal systemic venous drainage
 - Normal pulmonary venous drainage
- Atresia of the mitral valve
- Atrial septal defect (moderate)
 - Size : 4.3 mm
 - Ostium secundum type.
 - Peak/mean gradient across ASD = 39/17 mmHg
 - Lt to Rt shunt.
 - There is biatrial enlargement
- Intact ventricular septum
 - Non inverted ventricles
- Double outlet right ventricle (DORV)
 - Both the great arteries are totally arising from the right ventricle with normal spatial relationship of great arteries -NGRA
 - Sub aortic or subpulmonic conus were absent.
- There is an absence of pulmonary valvular / sub pulmonary / subaortic obstruction.
- Pulmonary arteries are markedly dilated
 - Pulmonary valves was normal
 - PV annulus (D) 18.7 mm, AV annulus (D) 8.3 mm
 - Main pulmonary artery MPA (D) : 14.0 mm
 - Left pulmonary artery LPA (D) : 6.6 mm
 - Right pulmonary artery RPA (D) 4.7 mm
- Tricuspid regurgitation (moderate)
 - TV are elongated
 - TR velocity = 4.06 m/sec (gradient 66 mmHg).
 - o On color flow mapping, TR jet area was 1.23 sqcm; 25 % of RA area, eccentric anterior jet .
- Dilated RV, hypoplastic LV with normal biventricular systolic function (RVEF = 61%, LVEF = 56%).
- There was absence of ventricular septal defect, patent ductus arteriosus, coarctation of aorta, aortic or pulmonary stenosis.

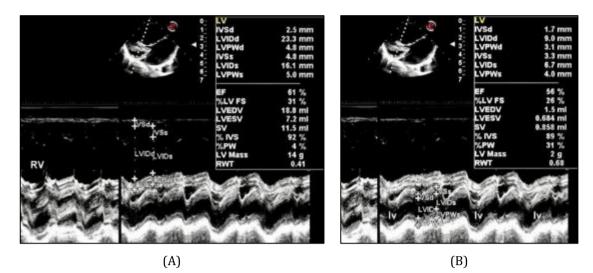
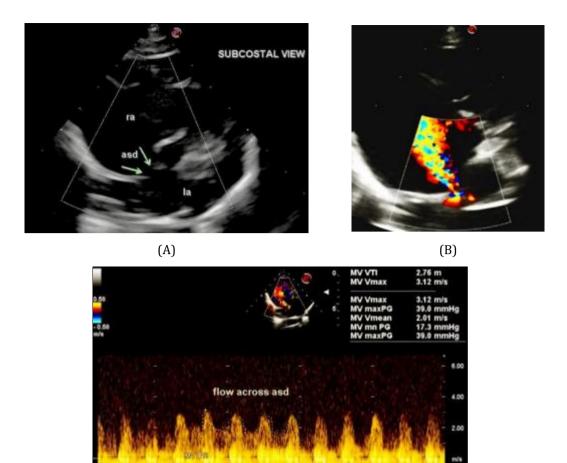


Figure 14 M-mode echocardiography of (A) dilated right ventricle and (B) hypoplastic left ventricle

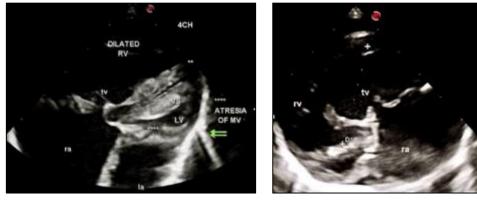


Figure 15 2-dimensional transthoracic echocardiography. In the subcostal view right atrium is lying to the right of left atrium, consistent with situs solitus



(c)

Figure 16 In the subcostal view; (A) Moderate sized ASD is visualised; (B) with left to right shunt; (C) peak and mean gradient across ASD was 39.0/17.3 mmHg



(A)

(B)

Figure 17 (A) 4CH view illustrates dilated LA, RA and RV. Conspicuously, mitral valve atresia is depicted by 4 asterisk (****) and 2 horizontal green arrows ; consequently there is presence of hypoplastic left ventricle with intact ventricular septum (vs). 2 asterisk (**) pinpoints to attachment of chordae of tricuspid valve to the RV apex; (B) Charecteristic dilatation of RV and RA is present.

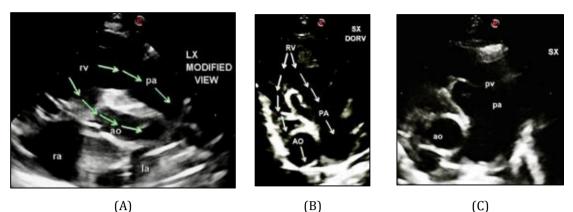


Figure 18 (A) In modified LX view and (B) SX view, DORV is demonstrated; (C) In the SX view, there is presence of normally related great arteries with markedly dilated pulmonary artery and its branches

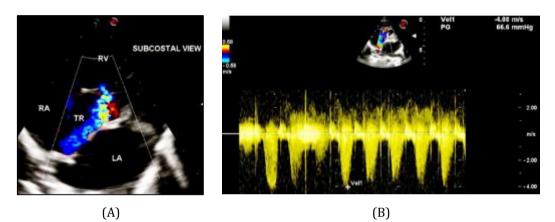


Figure 19 (A) In the subcostal view on color flow mapping, a moderate tricuspid regurgitation is visualised with an eccentric anterior jet; (B) On continuous wave doppler analysis across the tricuspid regurgitation jet, a peak velocity of 4.06 m/sec with a peak gradient 66 mmHg was noted

4.4. Summary of 2-Dimensional color echocardiography

Our index patient was afflicted with complex cyanotic congenital heart disease comprising of atresia of mitral valve, ASD, DORV, dilated non obstructed pulmonary arteries, dilated RV, hypoplastic LV with normal biventricular systolic functions Furthermore, moderate tricuspid regurgitation was detected with TR gradient across TV being 66 mmHg suggesting severely elevated right ventricular systolic pressure / pulmonary artery pressure.

4.5. Future course of action

Because of above mentioned afflictions, our index patient was severely symptomatic with cyanosis and breathlessness and hence, the infant was referred to a tertiary care pediatric cardiovascular institute for suitable palliative/ corrective surgery.

5. Discussion and review of literature

In general, dextroversion consists of a rotation of the ventricular part of the heart to the right, as in turning the page of a book, with the atria remaining in normal position. Usually there are transposition of the great vessels and a ventricular septal defect. Since the atria are in normal position, the direction of spread of atrial depolarization is the same as in the normal subject and therefore the P wave in lead I is upright, differentiating it from mirror-image dextrocardia. The QRS and T waves in dextroversion depend upon the type and degree of associated intracardiac malformation [11].

5.1. Embryology; Pathologic Anatomy [18]

The basic developmental defect in dextroversion complex occurs in the second week of fetal life. At that time, the primary cardiac tube begins to grow more rapidly than the surrounding thoracic structures. For this reason, in the normal fetus, the cardiac anlage doubles on itself to form an S-shaped curve. If, instead, the primordial cardiovascular

structure bends in a countersigmoid direction, dextroversion complex with complete inversion of cardiac chambers results.

In uncomplicated dextroversion, this initial embryologic phase is believed to be entirely normal, and development progresses without complication until approximately the sixth week of intrauterine life. The heart is then well differentiated and occupies a position in the thorax close to the mid-line. At this time the next normal step in development occurs, namely, rotation of the heart to the left about its long axis. This results in normal cardiac position and configuration. If rotation about the long axis occurs to the right instead of to the left, dextroversion results.

The pathologic anatomy resulting from this error in development varies with the degree of the malrotation. In fullblown cases of dextroversion, the left ventricle is rotated to an anterior position, forming the major portion of the anterior surface of the heart. The right ventricle lies well to the right and constitutes the right cardiac border. The right atrium occupies the posterior aspect of the heart and the left atrium comprises most of the left cardiac border. The ascending aorta and pulmonary artery are apparently rotated to a relatively smaller degree than the heart itself.

5.2. Dextro-version

Dextoversion is a fairly frequent malposition; Anselmi et al [19] described in their series that 12 of 41 cases of malposition were dextroversions (29%). It has been recognized under different names: dextrootation [20], isolated dextrocardia [21], dextrocardia with atria in situs solitus [22]; in fact, some of the published descriptions call dextroversion other malpositions, which accounts for the confusion shown in many statistics as to the actual incidence of this malformation.

Dextroversion is complicated in 90 per cent of the cases by cardiac malformations [11]. Only 10 per cent show no evidence of cardiac abnormality; of 12 cases, 2 showed no associated malformations [11]. This is probably related to the fact that dextroversion is an abnormal position of the heart in a situs solitus malformation that appears very early in development, and for this reason it is often complicated by complex malformations; 83 per cent of cases show severe cyanotic abnormalities and only 7 per cent mild and noncyanotic malformations. Likewise, our patient was an extremely complex case, presenting to us with dextroversion complicated by atresia of the mitral valve, ASD, DORV, dilated RV, hypoplastic LV with severe tricuspid regurgitation.

5.3. Truncoconal morphologies

In dextroversion, truncoconal morphologies are of three basic types [19]:

- Type (a) without TGA
- Type (b) with TGA is the most common and
- Type (c) with a common trunk is the least frequent.

5.4. Position of ventricles

In dextroversion the ventricles [19] can be in their normal positions, that is the ventral ventricle situated on the right and the dorsal ventricle situated on the left, this being the ventricular position corresponding to the situs in which the dextroversion has occurred (situs solitus). The second possibility is ventricular inversion. This means that the ventral ventricle is situated on the left and the dorsal ventricle on the right. In 50 % of our cases there was ventricular inversion, which proves the high incidence of this abnormal ventricular position in dextroversion.

The three truncoconal morphologies described above can be seen in dextroversion with or without ventricular inversion.

5.5. Unequal partition of truncoconus and lateral positions

In dextroversion, truncoconal morphologies can occur (which is usual) complicated by unequal partition and/ or by lateral positions [19]. When there are two vessels, the most common is an unequal partition at the expense of the pulmonary artery; this can be expressed anatomically as stenosis of the pulmonary artery, and occasionally atresia. Fifty per cent of complicated cases and 48 per cent of the published cases had pulmonary stenosis.

5.6. Associated malformations

Dextroversion is frequently associated with other disorders which, owing to their number and characteristics, cause a very complex malformation [19].

5.7. Associated malformations are outlined

- Septal defects
- Atrial septal defects
- Single ventricle
- Transposition of great arteries
- Pulmonary stenosis
- Pulmonary atresia
- Mitral atresia
- Tricuspid atresia
- Anomalous pulmonary venous drainage (partial/total)
- Anomalies of drainage of the systemic veins
- Patent ductus arteriosus

6. Conclusion

Three types of clinical dextrocardia are recognized; mirror image dextrocardia with situs inversus, cardiac dextroversion with or without situs inversus, and extrinsic dextrocardia.

The most common cause of dextrocardia is situs inversus where the thoracic and abdominal viscera are in mirror image positions relative to their normal state. The vast majority of such patients are otherwise normal without associated congenital cardiac malformations. Dextroversion is the second most common type of dextrocardia representing extreme right-sided cardiac rotation relative to normal. In the most common variety of dextroversion a D-ventricular loop is formed with the apex pointing to the right but the apex of the primitive heart fails to migrate from the right into the left hemithorax. Although the right atrium and right ventricle remain to the right, they are located posterior to the corresponding left sided chambers. It is as if, looking from below, the normal heart is rotated counterclockwise to the patient's right on an axis passing through the right atrium. In the less common form of dextroversion. L-ventricular loop forms (with the apex pointing to the left) and the primitive heart migrates into the right thoracic cavity. These patients have corrected transposition. In all forms of dextroversion there is a 90% incidence of additional cardiac malformations including corrected transposition, anomalous pulmonary venous return, tetralogy of Fallot and septal defects.

Due to the increased possibility of associated congenital anomalies associated with dextroversion, diagnosing dextroversion could be a bridge to the diagnosis of other congenital cardiac defects. Physicians should encourage routine medical examination for their patients which could help identify this anomaly and, upon suspicion, a thorough evaluation that involves imaging, is necessary to assess for other accompanying abnormalities, to prevent an incorrect diagnosis or death due to delayed management. Surgeons, radiologists, and radiographers should be watchful for this anomaly during the preoperative and surgical management of their patients.

Compliance with ethical standards

Disclosure of conflict of interest

No conflict of interest to be disclosed.

Statement of ethical approval

Ethical approval was obtained by the ethical committee of Prakash Heart Station, Lucknow.

Statement of informed consent

Informed consent was obtained from the parents of the infant.

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