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# When uncertainty generates more anxiety than severity: the prenatal experience with cystic adenomatoid malformation of the lung

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## Abstract

**Aim:** To assess reasons for higher levels of anxiety generated by prenatal counselling of a condition with good outcome such as cystic adenomatoid malformation (CCAM) of the lung compared to a life-threatening malformation such as congenital diaphragmatic hernia (CDH).

**Materials and methods:** The Spielberger State-Trait Anxiety Inventory (STAI-S) was used to measure anxiety in two groups of mothers carrying a fetus with the respective malformation.

**Results:** Forty-four mothers completed the questionnaire (CCAM, n = 21 and CDH, n = 23). Before consultation, the mean STAI-S scores in the CCAM group (44.80 ± 5.92) and in CDH group (44.05 ± 4.96) were not significantly different but was significantly reduced in both groups after consultation (CCAM 44.80 vs. 41.60, P = 0.014 and CDH 44.05 vs. 34.35, P = 0.0001). The groups were not significantly different regarding gestational age at diagnosis.

**Conclusions:** After initial prenatal counselling, uncertainty about prenatal outcome and lack of defined management plans in CCAM seems to be more important than higher mortality rate occurring in CDH.

**Keywords:** Counselling; cystic adenomatoid malformation (CCAM); emotional reactions; prenatal diagnosis; prognostic uncertainty.

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## Introduction

The shock encountered with finding a fetal anomaly is faced by an increasingly higher number of prospective parents. Anxiety experienced by parents following diagnosis of a congenital anomaly requiring surgery at birth might be due to different factors. It is sudden and unexpected, disrupts parents' sense of control, induces a sense of helplessness, determines loss of the child that the parents have expected and imagined. It would seem reasonable that the more severe the malformation the greater is the amount of anxiety experienced by parents. However, to our knowledge, such association has never been demonstrated.

In this respect, Kemp et al. [14] observed that reduction in anxiety was more consistent in conditions in which the outcome was well-defined. On the other hand, reduction of anxiety was lower in a condition such as cystic adenomatoid malformation (CCAM) where the prenatal outcome varies widely with obvious implications on neonatal clinical conditions. These authors suggested that parents seem to relate better to defined management plans rather than the more pragmatic yet entirely honest approach: "we will see what it is like when the baby is born".

Despite a large number of studies regarding the emotional impact of prenatal diagnosis, such unexpected observation has not been further investigated [2, 15, 18]. Thus, whether such a reduction of anxiety in CCAM was due to variable *in utero* development leading to different neonatal conditions was investigated.

## Material and methods

During the period 2001–2003, all mothers undergoing surgical consultation following the diagnosis of CCAM in the fetus were asked to participate in this psychological study. Results were compared to those obtained in mothers counselled for the diagnosis of congenital diaphragmatic hernia (CDH) over the same period of time. The latter is a type of congenital malformation with significant mortality rate for which a well-defined perinatal treatment protocol is followed at our Institution [5]. Briefly, all patients are ventilated at birth, transferred to the NICU where delayed surgery is performed based on definite ventilatory setting and blood gases.

In both groups, mothers were requested to complete the Spielberger State-Trait Anxiety Inventory (STAI-S) [7, 19] before and after consultation with our multidisciplinary team. This is a

**Table 1** Characteristics of groups.

| | Patients | Mean age | Educational level | Marital status |
|---|---|---|---|---|
| CCAM group | 21 | 24<br>Range 18–34 years | 3 Low<br>15 Medium<br>3 High | 16 Married<br>5 Cohabiting |
| CDH group | 23 | 26<br>Range 20–33 years | 5 Low<br>17 Medium<br>1 High | 15 Married<br>8 Cohabiting |

CCAM = cystic adenomatoid malformation, CDH = congenital diaphragmatic hernia.

scoring of anxiety designed to be self-administered and set in two parts. STAI-S score gives the level of state anxiety at the time of completing the text and it is defined as a transitory emotional response involving unpleasant feelings of tension and apprehensive thoughts. The STAI-T score measures the inherent trait anxiety level of the subject. In both parts, scores range from 20 to 80. Mothers in which diagnosis was not confirmed were excluded from the study as were those having a fetus in severe distress or with chromosomal anomalies. The questionnaire was completed before and after the first consultation regardless of the subsequent parents' choice.

Prenatal surgical consultation was performed at the joint meeting of experts from pediatric surgery, sonography, neonatology, radiology and psychology. The consultation was structured as follows: 1) The fetus was followed serially at intervals ranging from 14 to 28 days (mean 21 days); 2) sonography was performed by the same sonographer; 3) written information with sketches were also given to the parents.

The psychologist was present at all consultations and had at least one separate consultation with the couple before birth to work through the grieving process. To provide mothers with the opportunity to talk about their experience, express feelings about what happened, connect events with emotions and explore moral dilemmas, non-directive counselling was used as an early intervention. Counselling is seen as a dynamic psychoeducational process in which the goal is to facilitate the parents' ability to use the counselling in a meaningful way which minimizes psychological distress and increases personal control [16]. Counselling was given by a pediatric surgeon together with the neonatologist and consisted of information about the perinatal natural history of the anomaly, its surgical management, and long-term outcome.

A support group was used when the prospective parents wished to meet parents having a child with a similar anomaly.

The following variables were evaluated: gestational age, educational level, parity and marital status. Socio-economic status (SES) was measured as an educational level. According to the International Standard Classification, education was categorized into three levels: $>12$ years (high), 10–12 years (medium) and $<10$ years (low). Parity was categorized as primi- and multiparity. Marital status was categorized as married, cohabiting (as in Table 1), and unmarried.

Statistical comparison was made with the Wilcoxon test for paired data and with the Mann-Whitney for unpaired data.

## Results

Twenty-four mothers having fetuses with CCAM and 30 mothers with CDH were seen during the study period. Four mothers were excluded because of fetal chromosomal anomalies, two mother did not agree to participate and four were excluded because of significant fetal distress.

Characteristics of the groups are described in Table 1. The interval between initial diagnosis and detailed counselling with the multidisciplinary team ranged between 2 and 10 days. None of the mothers decided to terminate pregnancy after prenatal counselling.

Forty-four mothers completed the STAI questionnaires: 21 in the CCAM group and 23 in the CDH group. The two groups were comparable regarding SES, parity and marital status. The mean gestational age was 20 weeks in the CCAM group (range 16–29 weeks) and 19 weeks in CDH group (range 17–27 weeks). The two groups had similar gestational age at diagnosis. There was no significant difference in STAI-T scores between the two groups (38.5 vs. 40, $P=0.59$).

Before consultation, mean STAI-S scores in the CCAM group (44.05, range 31–62) and in the CDH group (44.80, range 29–65) were not different ($P>0.05$) but there was a significant reduction in STAI-S scores in both groups after consultation (CCAM 44.05 vs. 41.60, $P=0.014$ and CDH 44.80 vs. 34.35, $P=0.0001$). After consultation, STAI-S scores wee significantly higher in the CCAM group compared with the CDH group (41.60 vs. 34.35, $P<0.05$).

## Discussion

Antenatal diagnosis of a congenital malformation leads to a crisis and effective counselling may help during this phase [12, 17]. The threat for prospective parents is real and the rational response is to be afraid. Therefore, our aim was to compare anxiety level in two groups of mothers receiving consultation for fetuses with different conditions. The opportunity to understand the nature, implications and effects of the anomaly, both on the fetus and on the child must be emphasized. Such information together with elaborating the grieving process may help parents in facing the situation at birth.

In 1998, Kemp et al. [14] suggested that parents of fetuses with a malformation with good outcome such as CCAM, exhibit higher level of anxiety when compared to

other malformations, some with a poorer prognosis. CCAM are relatively rare developmental abnormalities of the lung. They are characterized as benign hamartomatous or dysplastic lung tumors with overgrowth of terminal bronchioles. CCAM is usually unilateral involving only one lobe of the lung [1]. The majority of fetuses with CCAM detected antenatally have a good outcome. However, because of the unpredictable growth patterns for CCAM lesions, appropriate diagnosis and follow-up are required. Nevertheless, there is general agreement that this anomaly is benign with little or no sequaelae into adulthood [9, 20]. Spontaneous *in utero* regression of CCAM is also recognized as a common event even in the case of large fetal CCAM.

Not surprisingly, when comparing the two groups of mothers, anxiety levels preceding consultation were high in both groups. This was in agreement with other studies suggesting that, despite the severity of the anomaly, the discovery of a fetal malformation is stressful and emotionally demanding for the parents and, as such, can be considered the crisis of their life [11]. In this respect, some studies have assessed that the extent of fetal illness does not determine the extent of family stress [8, 13]. In particular Aite et al. [3] did not report significant difference in STAI-S score at diagnosis and at birth in mothers of children with CDH, abdominal wall defect, intestinal atresia, and abdominal masses. Docherty et al. [10] found that mothers of critically ill infants, regardless of the type of diagnosis, worry about aspects of their infant's health.

Anxiety levels were significantly reduced in both groups of mothers after consultation. It is likely that the multidisciplinary counselling approach significantly affects anxiety levels since mothers can understand the prenatal and postnatal management as well as prognosis and quality of life.

However, while anxiety levels were significantly reduced in both groups of mothers after consultation, reduction was more evident in the CDH group and the difference reached statistical significance. This confirms our clinical impression and seems to suggest that at least after the first consultation, death risk does not appear to be the major determinant for anxiety.

We speculate that maternal anxiety after consultation may be associated to the uncertainty regarding clinical development *in utero* and type of treatment rather than to the seriousness of delivered information. As a matter of fact, cystic growth potential between 18 and 26 weeks varies widely and this makes outcome rather unpredictable, yet rarely fatal.

Unfortunately, no prediction is available whether a fetus with a large CCAM will progress into hydrops or will slowly regress. Absence of a straightforward program is likely to prevent mothers from being in control of the situation.

When a CCAM is suspected, efforts should be directed to explain in detail all different available treatment options during pregnancy and after birth. In addition, once such exceedingly high anxiety levels are acknowledged, a closer follow-up program should be offered to the couple. As it has been widely demonstrated, reducing anxiety would affect the mother-baby relationship as well as infant development [4, 6]. Therefore, once a diagnosis is suspected couples should be promptly referred to a multidisciplinary team, i.e., a network for couples confronted with this delicate problem respecting their right to complete and clear information as well as to psychological support.

We conclude that in CCAM, maternal anxiety levels are not necessarily dependent on death risk: variables related to the *in utero* development and clinical outcome might be more important than mortality rate. Because of these psychological constraints, a structured prenatal consultation program with scheduled meetings throughout pregnancy is recommended. Whether or not such finding might be applied to other anomalies remains to be clarified.

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