



Sudden Unexpected Postnatal Collapse: Analysis of Some Clinical Cases and their Diagnostic Approach

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Abstract

SUPC is a clinical entity occurring in the first week of life that has been more defined recently. We here report six clinical cases defining their characteristics and discussing the diagnostic approach and preventive measures.

Keywords: SUPC; Skin-to-skin care; Hs troponin I

Introduction

Sudden Unexpected and Unexplained Postnatal Collapse (SUPC) is a relatively new clinical entity. It wants to indicate a situation in which apparently healthy newborn infants present a sudden and unexplained episode of hypotonia and/or pallor and/or apnea and /or bradycardia and/or cyanosis that requires some form of resuscitation and can evolve into death. The event happens in babies that have been given an Apgar score at 10 min >7, whose gestational age was > 35 weeks and typically occurs in the first week of life [1].

In practice SUPC includes two entities: early Sudden Infant Death Syndrome (SIDS) or Sudden Un-Expected Neonatal Death (SUEND) and early Apparent Life Threatening Event (ALTE).

Incidence of SUPC is between 1.6-133 cases/100,000 live births depending on the reports. About 33% of SUPCs occur within the first 2 hrs after delivery, 33% between 2 hrs and 24 hrs, while the remaining 33% between 24 hrs and the first week of life [1,2].

Risk factors that have been identified are [2-4]:

1. Unattended skin-to-skin care (SSC). SSC consists of giving the baby to his/her mother immediately after birth and for the two hour subsequent to the delivery (post-partum period): during such procedure the mother and the baby have not to be left unattended, and the presence of persons like husband or grandmother that are not specifically educated does not give any guarantee.
2. SSC when mother is tired, sleepy or under sedation.
3. Use of mobile smartphone during SSC: many mothers send messages and photos after birth.
4. Newborn infants that have had any form of resuscitation, or are not stable or have medical complications since birth.
5. Preterm infants < 36 weeks' gestation.
6. Primiparous mother.
7. Unattended co-bedding.
8. Prone position of the infant.
9. Hypothermia (left axillary temperature <36.3°C [5,6]).

We here retrospectively describe 6 cases of SUPC that we have registered in 2015 in our general county hospital, a secondary level perinatal center with level IIIA Neonatal Intensive Care Unit (NICU) and Pediatric Emergency Room (ER), and analyze clinical, laboratory and instrumental investigation data of such cases.

Case Series

Case 1

Female, 40⁺⁵wks' gestation, birth weight (b.w.) 3,080 gr, Caucasian, born by vaginal delivery

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from a primiparous mother. Delivery was defined as precipitous. Apgar score at 1 min was 9 and at 5 min were 10. At 46 hrs and 30 min of age she presented brief episodes of perioral cyanosis accompanied with tachypnea, and bradycardia till to 80 b/min during sleep. She was brought by the mother to the Nursery. Capillary refill time was 3.5 sec, the baby was given O₂ and was admitted to the NICU. Exams did not reveal infections, hypoglycemia, electrolytes disturbance, hemorrhage or changes in echogenicity at brain ultrasounds; QTc interval on ECG was within limits and chest X-ray was negative for diseases. The only findings were a transient increase in Hs troponin I level and a small Patent Foramen Ovale (PFO) with left-to-right shunt at Doppler echocardiography. The baby was breastfed; first attachment was done at 12 hrs. She was taken under observation and monitoring for seven days and discharged when troponin was normalized and infection excluded. Outcome after 1 year was favorable.

Case 2

Male, 39 wks' gestation, b.w. 3,360 gr, Chinese, born by vaginal delivery from a multiparous mother. Vaginal swab was positive for Group B Streptococcus and complete intrapartum antibiotic prophylaxis was given. Apgar score at 1 min was 8 and at 5 min was 9. At first visit in the delivery room minimal axial hypotonia was recorded. SUPC occurred at 3 hrs and 40 min of age with facial cyanosis, axial hypotonia, acrocyanosis, tremors under stimulation, valid suction, normal differential pulsoxymetry. He was given O₂ for few minutes and was admitted to NICU for observation and exams particularly addressed to hypotonia and cyanosis. Aminoacidic profiles and acylcarnitine were within limits. TSH was high (8.2 mU/L) at the episode, but returned to normal after 1 month without any therapy. Anti-thyroid antibodies were negative. Similarly ECG with QTc interval measurement was normal as well as brain echography. At Doppler echocardiography a small PFO was shown with left-to-right shunt. The baby was breastfed and first attachment was done at 4 min. Hospital staying lasted 8 days. He was lost at follow-up.

Case 3

Female, 40⁺wks' gestation, b.w. 3,390 gr, Caucasian, born by vaginal delivery at another hospital from a primiparous mother. Apgar score was not available. SUPC occurred at home between 48-72 hrs, i.e. after an early discharge from the nursery. At 10 pm, 2 hrs after breastfeeding, the baby was supine and presented tremors followed by stiffening of head and trunk, and then hypotonia and pallor with unconsciousness. Parents refer that the event lasted about 10 minutes. At the arrival at ER she was awake, reactive, heart rate was 133 b/min and transcutaneous SatO₂ was 99% in room air. She was admitted to NICU and given O₂ because SatO₂ subsequently lowered to 90%, with respiratory rate of 35 br/min. Mild axial hypotonia was described and bilateral ocular secretion too. Bacteriological examinations, including blood, ocular secretions, urine, pharynx and auricular samples, were negative as well CRP and the rapid test for Respiratory Syncytial Virus (RSV). Glycemia, serum electrolytes and ammoniemia were within limits. WBC and RBC count were normal but hematocrit was 62.8%. At 4 hours from the event QTc interval was elongated (450 msec) with reversed T in V₁ and V₄. After 18 hrs QTc interval was normal (382 msec), but a complete right bundle branch block (RBBB) was present. Hs troponin I level was increased (0.62 ng/ml). Doppler echocardiography did not show anomalies. To exclude seizures, an aEEG was performed and resulted within limits (then confirmed by 12 channels EEG). In the familial history there was a paternal aunt with a not well defined genetic anomaly and peripheral paralysis.

The baby remained in hospital for 4 days and was discharged with diagnosis of SUPC characterized by loss of consciousness and hypotonia. Outcome after 1 year was favorable.

Case 4

Male, 40 wks' gestation, b.w. 3,320 gr, Caucasian, born by elective CS from a primiparous mother. Apgar score was 9 at 1 min and 10 at 5 min. SUPC occurred at home between 72-96 hrs of age. The baby was discharged at 72 hrs. In the following day, while he was fed with expressed mother's milk given by bottle, he suddenly closed the mouth, became hyperemic in the face and rapidly turned to hypotonia. Mother stimulated him and he restarted breathing, however remaining hyporeactive. He was therefore brought to pediatric ER and at admission he was cold, sleepy with slow response to stimuli. The physical examination was normal and vital parameters were stable. Chest X-ray picture was compatible with inhalation of milk and, during a subsequent feed, he presented 2 short episodes of desaturation with spontaneous resolution. Hs troponin I, myoglobin and CK-MB levels resulted altered. ECG showed non-specific anomalies of ST-T in V₁ and V₄. To exclude other diagnosis the following exams were done and resulted negative: EEG, cardiorespirography, gastro-esophageal junction and pyloric echography, adrenal glands echography, fundus oculi, aminoacidemia, aminoaciduria, urinary organic acids, very long chain fatty acids (VLCFA). Only troponin resulted high (1.32 ng/ml) and persisted high for days without significant ECG and cardiac echo changes and with the baby being in good conditions. For such a reason after 12 days he was sent to a Pediatric cardiology center where, however, they could not explain the cause of such high level of the enzyme and reckoned it was due to some problem of the laboratory method. We discharged the baby with the diagnosis of SUPC in gastroesophageal reflux done on a clinical basis. The infant was subsequently admitted at another hospital at the age of 2^{1/2} months confirming the clinical diagnosis of gastroesophageal reflux. At 1 year the outcome was good.

Case 5

Female, 41⁺wks' gestation, b.w., 3270 gr, Caucasian, born by vaginal delivery from a primiparous mother at another hospital. Apgar score at 1 min was 9. Vaginal swab was positive for GBS and antibiotic intrapartum prophylaxis completed. SUPC occurred at home at 160 hrs of life. The mother referred that the previous evening the baby had refused breastfeeding. Then, in the morning, she sucked less vigorously than usual. Vomits and regurgitations at every meal were reported. Parents therefore decided of paying a visit to the hospital where the baby was delivered, but during the way the baby became cyanotic, hypotonic and hyporeactive. They therefore stopped at the first ER on their way, where the father attempted mouth-to-mouth resuscitation and subsequently chest compressions and insufflation were performed, following which the baby started recovering. She was then transferred to our pediatric ER, and when she arrived her vital parameters were within normal limits, she cried vigorously, her clinical exam, including the neurological aspect, was normal. Capillary refill time was 2 sec. In the familial history: epilepsy. Arterial blood gases, CBC, BUN, glycemia, serum electrolytes, CRP resulted negative. Blood culture, auricular swabs, urine culture, adeno-rota virus and Respiratory Syncytial Virus search resulted negative. In the pharyngeal swab *S. aureus* was isolated. Normal was also ECG, QTc, cardiorespirography, EEG, abdominal echography, cerebral ultrasounds. Chest X-ray showed reduced transparency of superior and medium right lobes, compatible with milk inhalation.

Table 1A: Laboratory examinations done and their absolute and relative positivity.

	Done	Positive	% Positivity
BGA	2	1	50
CBC	6	2	33
CRP	6	0	0
Hs troponin I	4	3	75
RSV	1	0	0
Aminoacidic profile, VLCFA, acylcarnitine	1	0	0
TSH	1	1	100
Blood culture	3	0	0
BUN, glycemia, serum creatinine, serum electrolytes	6	0	0
Ammoniaemia	2	0	0
Urine culture	2	0	0

BGA: Blood Gas Analysis; CBC: Complete Blood Count; CRP: C Reactive Protein; Hs: High Sensibility; RSV: Respiratory Syncytial Virus; VLCFA: Very Long Chain Fatty Acids; TSH: Thyroid Stimulating Hormone; BUN: Blood Urea Nitrogen.

Gastroesophageal junction echography put in evidence an open His' angle with frequent regurgitations and reduced cleaning time (>5 sec). At echocardiography PFO with right-to-left shunt was found, and Hs troponin I was modestly increased: 0.239 ng/ml. The baby was given amoxicillin + omeprazole and was discharged after seven days with the diagnosis of SUPC and gastroesophageal reflux. She was lost at follow-up.

Case 6

Female, 39 wks' gestation, b.w. 3,290 gr, Caucasian, born by vaginal delivery from a primiparous mother. Apgar score at 1 and 5 minutes was 9. SUPC was recorded at 8 hrs of life, in hospital. The neonate was brought from the mother in her arms for regurgitation and apnea. She was cyanotic, in apnea, hyporeactive and hypotonic. Mucous with digested blood was present in mouth and choanae. After aspiration of such secretions, O₂ was administered with mask, and gradual recovery of colour and reactivity followed. Hematocrit resulted 71.1%, WBC 30,000/mm³, RBC 6,470,000/mm³, Hb 22.4 g/dl. All other exams were normal apart from the discovery of a very small PFO with left-to-right shunt. The baby had already been attached at mother's breast within 1st hr and the mother had no fissures of the nipple. The baby was given iv fluids due to the very high hemoconcentration and after 5 days was discharged with the diagnosis of SUPC by hematic hyperviscosity. Outcome after 1 year was favorable.

Analysis by exams done (Table 1A and 1B)

When we analyzed the biochemical exams performed and their contribution to the diagnostic process we could observe that blood gas analysis (BGA) was altered in 50% of cases, Hs troponin I in 75% of cases and TSH in the only case in which it was determined (100%), while all other laboratory investigations resulted significant only to exclude any particular disease. Instead, if we take into account the instrumental investigations, the positivity of such exams was greater and especially important when they were done on the basis of a diagnostic suspect tied to symptoms.

Discussion

In the present report we analyze the data from 6 cases of SUPC that we observed in our county general hospital in 1 year period.

Table 1B: Instrumental investigations done and their absolute and relative positivity.

	Done	Positive	% positivity
Chest X-ray	4	2	50
Abdomen X-ray/US	3	0	0
Brain US	4	0	0
Heart echo - Doppler	5	4	80
GEJ US	2	2	100
ECG	6	2	33
QTc	6	1	16.6
EEG	3	0	0

US: Ultrasounds; GEJ: Gastroesophageal Junction; ECG: Electrocardiogram; QTc: Corrected QT; EEG: Electroencephalogram

Fifty percent occurred in hospital while the remaining 50% happened at home. In particular 33% occurred in the time period 2-24 hrs, none in the first 2 hrs, and the remaining 67% in the time interval 24-168 hrs.

From the literature data about one third of SUPC are registered in the first 2 hrs of life and have been related to unattended or poor controlled SSC, sleepy or tired mothers, unstable infants, use of smart phone [2-4].

Why SUPC is limited to the first week of life, and especially to the first hours after delivery, is not clear. One hypothesis is that transition from fetal to extra-uterine life can make the newborn more vulnerable during these first hours of life. Neuro modulators and prostaglandin that inhibits fetal movement are elevated before delivery and during birth, and fall after birth. In the normal delivery there is a fall in the inhibiting adenosine and this allows more movements of the newborn and increased breathing activity. Asphyxia during delivery determines a great release of PGE₂ in the newborn that has an inhibitory effect on the brainstem centres of breathing. Besides these levels of PGE in plasma are 20-fold higher in newborns than in older infants, and declines quickly after the first postnatal week. Here hence the hypothesis that PGE are implied in the respiratory instability of the infant in the first week of life (see ref. 2 for mini review).

Prevention of SUPC is especially addressed to a greater attention at risk factors in the first 2 hrs of life, when control can be more easily conducted by skilled personnel in the delivery room [7]. In Italy mother and infant must remain in the delivery room for the first 2 hrs after delivery by law, where they are regularly checked by skilled personnel. We can speculate that this is the major determinant of the absence of cases in the first 2 hrs of life in our series. This in turn might imply the need of a more close control in the first 24 hrs while in hospital. However it remains very arduous to prevent home events, even with prenatal and postnatal family education on correct and controlled SSC, avoidance of unattended co-bedding, avoidance of hypothermia and of supine position. Indeed in our cases none of the preventable causes of SUPC [2,3] were evident. Only in case 2 mild hypotonia probably would have required more prudence before starting rooming-in.

Besides our cases were each one very different in their presentation, varying from respiratory symptoms to symptoms related to feeding and to neurologic signs, and it is therefore difficult to identify a classical clinical picture of SUPC and therefore also to implement prevention, especially when babies are at home and a direct control cannot be carried out. The major link between them is

that 5 out of 6 were babies born by primiparous mother; however this is a non-preventable risk factor. Case 1 had a collapse with troponin movements; case 2 had hypotonia and transient hypothyroidism; case 3 had tremors followed by stiffening and loss of consciousness with RBBB, a transient elongation of QTc and troponin movement; case 4 had hypotonia and hyporeactivity episode during feeding and diagnosis was gastroesophageal reflux and persistent elevation of troponin; case 5 similarly had gastroesophageal reflux and milk inhalation, was resuscitated and also in this case troponin's value was altered; case 6 had hypotonia and hyporeactivity due to very high hematocrit.

A second link was the presence in two cases of gastroesophageal reflux, a treatable problem. Five cases out of 6 had hypotonia as presenting symptom, but only one had hypotonia before the SUPC. ECG changes found were in any case transient and can be interpreted as secondary to the symptoms instead of being the cause of SUPC.

Signs and symptoms resemble ALTE and we cannot exclude that links exist between ALTE and SUPC. Some Authors suggest a link between SUPC and SIDS or ALTE in the first week of life is strong, with analogous mechanisms and risk factors. [8-10] An apparent connection is prevention by avoidance of the prone position of the baby and by performing control during prone position of SSC.

As concern the laboratory investigations done to diagnose or at least exclude any possible cause of SUPC, in our series the very great majority of the exams performed routinely proved useless to diagnosis, but useful anyway to exclude infections or electrolyte disturbances or metabolic congenital problems. Hs troponin I resulted positive in many cases, but this finding was not accompanied with ECG abnormalities or, to be precise, was associated only with minimal aspecific changes. We retain that the movement of troponin can be related to the severity of the event more than to be a cause of SUPC. Hs troponin I, being very sensitive, can be increased even when damage to muscle of the heart is not evident on ECG, but can be an indirect sign that something was really happened even when we did not observe it. Severe infections and renal problems can alter Hs troponin I, but its value is not influenced by difficult blood sampling or muscle trauma [11]. Anyway more data are required to establish its diagnostic value in infants.

As for the instrumental investigations, these were positively involved in the diagnosis when they were done on a clinical suspect basis. Indeed, also in this case it is difficult to say which investigations could be avoided. Collapse and hypotonia and cyanosis, at this age of life, can be due to infectious, cardiac, neurological or metabolic problems as well. Clinical examination also when performed by a skilled neonatologist, can give some clues and can induce to perform some investigation before than others, trying non-invasive exams if possible, e.g. ultrasounds instead of X-rays. However if the results are negative it is necessary to search again and exclude other possible causes. In the meanwhile the baby must remain under strict observation and monitoring without interrupting the processes of bonding and milking.

Minor echocardiographic findings as PFO, were not probably connected with SUPC in our cases because in none of the patients there were persistent neurological deficits possibly tied to cerebral embolism through PFO. The increased value of troponin led to perform echocardiography but it is very probable that changes of cardiac enzymes and of ECG after a SUPC are a scar of the event and/

or of the resuscitation manoeuvres and not the cause of it.

Looking at our data, different clinical onsets in different settings (hospital vs home) were approached differently by the physicians. In any case the work-up was consistent more with clinical symptoms than with a standardized protocol for SUPC.

A protocol of investigations both for intra-hospital or outside hospital SUPC has been proposed from Wellchild, a charity endorsed by the British Association of Perinatal Medicine, to allow diagnosis of SUPC, but it looks quite complex for an extensive use, especially to screen with priority relevant and treatable problems [12].

Only in 50% of SUPCs of our series a possible and related cause was found, and this is another similarity with ALTE. In the cases in which we find a cause we can give a treatment or solve the problem. When SUPC is idiopathic, instead, return at home creates problems both for parents and physicians. Parents are worried about the possibility that the event can recur and pediatricians do not have precise answers. To propose home-monitoring in these cases might be taken into account, but there are not definite indications for SUPC cases.

Conclusion

SUPC is a relatively new acronym for an already known problem. However the new approach to physiologic delivery and birth has given an impulse to the frequency of such problem. Unattended SSC and rooming-in seem the main cause of the increased incidence of SUPC in hospital, together with erroneous indications to the procedure. Cases happening at home may have other and less definite facilitating causes.

A question arises: does the increase of SUPC recorded in the last years determine a crisis of several concepts that have been demonstrated to be useful for the infant and the mother-infant bonding as SSC, rooming-in, discharge at 48 hrs? Our actual answer is that a greater non-intrusive control during the first 24-48 hrs in the hospital targeted to safe SSC, safe breastfeeding establishment and secure positioning of the infant during sleep could reduce the risk of intra hospital SUPC, and a checklist has been proposed for evaluating SSC and the first 2 hrs after delivery that could be broaden to the entire period of staying in the hospital [2,7]. Such physiological approach requires skilled personnel in sufficient number, and not a reduction of it, how rooming-in is too often interpreted by hospital directorates. Anyhow such approach requires to be evaluated in large numbers.

Prenatal and postnatal education of parents on SSC management, co-bedding avoidance, back-to-spine sleep of the baby seems at present the only way for preventing home SUPC as well SIDS.

More studies are necessary to define better SUPC, to determine a simple protocol of first investigations and to promote new forms of prevention.

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