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ORIGINAL ARTICLE

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# Seven cases of neonatal appendicitis with a review of the English language literature of the last century

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**Abstract** Neonatal appendicitis (NA) is a very rare surgical condition. The aim of this study is to once again draw attention to this subject by collecting our cases with NA and cases of NA reported separately in English-language literature over the period from 1901 to 2000. We performed a retrospective chart review of patients admitted to our hospital, with the clinical diagnosis of NA from 1990 to 2000. A survey of the English-language literature together with our own 7 cases revealed a total of 141 cases of NA during the period of 1901–2000. 128 cases had sufficient information for analysis. The patients are grouped and discussed according to these 3 time– periods: 1901–1975, 1976–1984 and 1985–2000. The incidence, etiology, and presenting signs and symptoms of appendicitis in newborns are discussed. Despite the similar perforation rates in the 3 time– periods (73%, 70%, 82%), mortality rate in NA has decreased from 78% in the 1901–1975 period, to 33% in the 1976–1984 period, and to 28% in the 1985–2000 period. A newborn baby presenting with continuous vomiting, refusal to feed, and , showing signs of pain through irritability, restlessness, sleep disturbance, and a distended abdomen; one should strongly suspect an abdominal disorder, perhaps appendicitis.

**Keywords** Neonatal appendicitis · Appendicitis · Neonate

## Introduction

Neonatal appendicitis (NA) was first reported by Albrecht in 1905 in a 1-month-old male infant who died of peritonitis. The first neonate to survive was a 3-week-old male with appendicitis in a scrotal hernia, described by Lillenthal in 1908 [8].

Since the review of 94 cases of NA by Surouji (1901–1975), Massad reported an additional 17 cases (1976–1984) (Table 1), and another 10 NA have later been described in the literature between 1985 and 2000 (Table 2) [6,10].

The aim of this study is to collect our cases with NA and cases of NA in English-language literature reported separately since 1985 and once again draw attention to this subject.

## Materials and methods

We performed a retrospective chart review of patients admitted to our hospital with clinical diagnosis of NA who underwent emergency appendectomy from January 1990 to December 2000. A total of 7 patients were found (Table 3).

A survey of the English-language literature from 1985 to 2000 was carried out and we combined our cases with those of the earlier reviewed and reported cases.

## Results

In a previous review from the period 1901–1984, 123 cases were collected, of which 111 were adequately documented [6]. Seventeen out of 18 cases had adequate documentation during the period 1985–2000.

A survey of the English-language literature together with our own 7 cases revealed a total of 141 NA cases during the period 1901–2000 [1,2,4,5,6,7,9.10,11]. Out of these, 128 cases had sufficient information for analysis.

The patients were grouped and compared according to 3 time– periods: 1901–1975, 1976–1984 and 1985–2000. A definitive diagnosis was made in 60%, 94%, and 100% of the cases at operation and in 40%, 6%, and none of the cases at autopsy respectively [6,10]. Seventy-five per cent of the newborn infants were male and 25% were female, 52% of these babies were pre-term and

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**Table 1** Data on 128 cases of neonatal appendicitis with adequate documentation (1901–2000)

| | 1901–1975 (94 cases) | | 1976–1984 (17 cases) | | 1985–2000 (10 cases) | | OURS (7 cases) | |
|---|---|---|---|---|---|---|---|---|
| | ABD* | HER** | ABD | HER | ABD | HER | ABD | HER |
| Sex | | | | | | | | |
| Male | 40 | 24 | 9 | 2 | 4 | 2 | 3 | 1 |
| Female | 18 | | 6 | - | 2 | | 3 | - |
| Not reported | 9 | 3 | - | - | 2 | - | - | - |
| Maturity | | | | | | | | |
| Term | 23 | 9 | 9 | 2 | 2 | - | 5 | - |
| Preterm | 31 | 11 | 6 | - | 4 | 1 | 1 | 1 |
| Not reported | 13 | 7 | - | - | 2 | 1 | - | - |
| Time of Diagnosis | | | | | | | | |
| Intraoperative | 29 | 27 | 14 | 2 | 8 | 2 | 6 | 1 |
| Autopsy | 38 | - | 1 | - | - | - | - | - |
| Status of Appendix | | | | | | | | |
| Non-perforated | 25 | - | 4 | 1 | - | 1 | 2 | - |
| Alive | 7 | - | 2 | 1 | - | 1 | 2 | - |
| Perforated | 42 | 27 | 11 | 1 | 8 | 1 | 4 | 1 |
| Alive | 7 | 7 | 8 | 1 | 6 | 1 | 2 | 1 |

*ABD: Abdominal
**HER: Hernial sac

**Table 2** Literature reports of neonatal appendicitis (1985–2000)

| Literature | Gestation, birth wt (*kg*) | Sex | Age (*days*) | Status of appendix | Location | Additional disease | Hosp. time* (*days*) | Outcome |
|---|---|---|---|---|---|---|---|---|
| Singh I, 1986 | 36wks 2,020 | M | 13 | Non perforated | Hernia | - | - | Alive |
| Arliss J, 1990 | ? | M | 14 | Perforated(base) | Abdominal | Hirschsprung's disease | - | Alive |
| Deguchi E, 1990 | 35wks 1,508 | M | 9 | Perforated(base) | Abdominal | - | 53 | Alive |
| Ruff ME, 1991 | ? | ? | ? | Perforated(mid portion) | Abdominal^ | - | - | Alive |
| Arora NK, 1991 Case 1 | 31wks 1,360 | ? | 12 | Non perforated | Abdominal | - | 11 | Died |
| Case 2 | 30wks 1,250 | M | 5 | Perforated | Abdominal | - | 30 | Alive |
| Stiefel D, 1998 Case 1 | Term | F | 21 | Perforated | Abdominal | Hirschsprung's disease | 159 | Died |
| Case 2 | Term | F | 28 | Perforated | Abdominal | Cystic fibrosis | - | Alive |
| Case 3 | 33wks | M | 5 | Perforated(base) | Abdominal | - | - | Alive- |
| Iuchtman M, 1999 | ? | M | 6 | Perforated | Hernia | - | - | Alive |

*Hosp. time: Hospitalization time

**Table 3** Cases of neonatal appendicitis operated in Dr. Sami Ulus Children's Hospital, Department of Pediatric Surgery between 1990–2000 (7 cases)

| | Gestation, birth wt (*kg*) | Sex | Age (*days*) | Status of appendix | Location | Additional disease | Hosp. time* (*days*) | Outcome |
|---|---|---|---|---|---|---|---|---|
| Case 1 | 36wks2,000 | M | 28 | Perforated(distal) | Hernia | - | 8 | Alive |
| Case 2 | 32wks2,200 | M | 22 | Perforated(distal) | Abdominal | - | 5 | Dead |
| Case 3 | 40wks4,500 | F | 18 | Perforated(distal) | Abdominal | - | 20 | Alive |
| Case 4 | 38wks3,600 | M | 4 | Perforated(base) | Abdominal | - | 1 | Dead |
| Case 5 | 38wks3,020 | M | 11 | Non perforated | Abdominal | - | 23 | Alive |
| Case 6 | 39wks3,000 | F | 11 | Non perforated | Abdominal | - | 15 | Alive |
| Case 7 | 38wks2,000 | F | 23 | Perforated(distal) | Abdominal | Operated for EA + TEF | 7 | Alive |

*Hosp. time: Hospitalization time

48% were term, 74% had perforated appendicitis and 26% had non-perforated appendicitis. Since 1981, NA was not recognised at autopsy. Perforation was diagnosed in 95 of the 128 babies (74%). Survival rate was not any better for those without a diagnosed perforation(13 of 33; 39.4%) than for those with a diagnosed perforation (33 of 95; 34.7%) ($P = 0,63$). According to information available, only 46 infants out of 128 survived (36%).

In the period 1985–2000, all patients (reported cases and our own cases) with non-perforated abdominal appendicitis and hernial appendicitis survived. Four of

the 14 neonates with abdominal appendicitis died (28%), less than when compared with 5 out of 15 (33%) in 1976–1984, and 53 out of 67 (78%) in the 1901–1975 period. Total mortality rate in abdominal appendicitis was 64% (62 of 96). Mean age at the diagnosis was 14 days, and mean hospitalization time was 30 days. Two neonates, who had perforated appendicitis, were later diagnosed with Hirschsprung's disease and one other neonate had cystic fibrosis.

## Discussion

Although acute appendicitis is the most common acute surgical condition of childhood, it is seldom considered in the differential diagnosis of abdominal distention in the newborns.

NA is a rare condition with a reported incidence of 0.04% [3]. Snyder and Chaffin proposed four factors to explain the low incidence of appendicitis in the newborns: (1) a funnel shaped appendix, (2) a diet of soft foods, (3) recumbent posture, and (4) infrequent gastrointestinal and upper respiratory infections [8].

NA is found intra abdominally in three fourths (96 of 128) and in an inguinal hernia sac in one fourth of the patients (32 of 128). A definite survival rate with a hernial appendicitis may be attributed to obvious physical findings resulting in earlier surgical intervention. Neonatal intra abdominal appendicitis has a high incidence of perforation because of a thin appendiceal wall and an indistensible caecum [6]. Perforation is a significant factor in prognosis since it frequently results in peritonitis. However, the major factor responsible for a high mortality in a newborn appears to be the delay in diagnosis, since the clinical presentation of NA does not have the characteristic features.

In our review, abdominal distention (11 of 17; 64%) and bilious vomiting (8 of 17; 47%) were the most common symptoms of NA. Induration and oedema over the abdominal wall (6 of 17; 35%) and a right lower quadrant mass (3 of 17; 17%) were also seen. Other less consistent findings were irritability, anorexia, fever and leucocytosis. Others have also confirmed these observations [3,6,7,10].

An abnormal intestinal gas pattern was found in roentgenographic evaluation of neonates with suspected appendicitis but was not specific. Other findings were presence of free peritoneal fluid and air, a right scoliosis, psoas margin obliteration, and abscess.

Early surgical intervention with appendectomy before perforation occurs is the ideal treatment, but the difficulty in establishing a diagnosis in neonates makes this an exception.

Despite the perforation rate remaining the same in the 3 time– periods (73%, 70%, 82%) ($P = 0.69$), mortality rate in abdominal NA has decreased from 78% in the 1901–1975 period, to 33% in the 1976–1984 period, and to 28% in the 1985–2000 period ($P < 0.01$) due to the rapid advancements in the field of antibiotic therapy and surgical care.

Hence, a newborn baby who continuously vomits, refuses to feed, shows signs of pain through irritability, restlessness, and sleep disturbance, and also has a distended abdomen, an abdominal disorder, perhaps appendicitis should be strongly suspected.

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